There's more to life than perfect plosives:

Assessing the psycho-social impact of late intervention for cleft lip/palate using an adapted scale of the Therapy Outcome Measure (TOM)

By Joanna Rees (MSc, MFDSrcps, MSurgDent, BChD)

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The University of Sheffield
Department of Human Communication Sciences
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I must also extend my thanks to my husband Martin for continually having more confidence in me than I did in myself and to our ever patient dog Enzo for his hours of waiting and wondering why a computer screen was so much more entertaining than a run on the fells.

Finally I would like to remember all the participants in Assam and Hyderabad who shared their experiences with such kindness and from whom I have learned so much. Thank you each and every one of you.
Thesis statement

This paper contests that, although physiological parameters such as speech and intelligibility have value in assessing the efficacy of late treatment for cleft lip and palate (CLP), they do not account for important psychosocial impacts of intervention. It suggests that an adapted version of the Therapy Outcome Measure (TOM) (Enderby and John, 2015) can be used as a culturally sensitive instrument to demonstrate benefits which are potentially being underestimated and, as such, are currently potentially depriving this cohort from intervention. The thesis also argues a greater emphasis should be placed on overcoming the barriers individuals face in accessing treatment and that provision of comprehensive treatment for CLP can be beneficial for people of all ages irrespective of age and socio-economic status.
Abstract

Objective: To ascertain whether late intervention for CLP is associated with beneficial psychosocial changes and whether these can be identified and measured reliably in clinical practice.

Method: This research explores the lived experiences of older individuals with CLP in India and investigates the barriers which prevent them from accessing care. It uses framework analysis to evaluate semi-structured interviews and examines how an adapted version of the TOM, the TOM-clp, can be used to demonstrate both physiological and psychosocial change for individuals undergoing intervention later in life.

Results: The TOM-clp showed excellent inter-rater reliability after only a short training session and was quick and easy to implement in a clinical setting. It proved culturally, age and gender sensitive and could be delivered in multiple language settings with neither financial implications nor additional demand on local resources. Framework analysis and TOM-clp data indicated positive change in all domains following late intervention and revealed the importance that secondary surgery, dental care and orthodontic intervention played in enhancing the lives of these participants. Barriers to treatment included superstition and lack of awareness about CLP, which echoed those already highlighted in the literature. Discussion with service providers indicated that auditing and benchmarking could have a key role in improving quality of care at a wider level.

Conclusions: The TOM-clp shows promise as an outcome measure to demonstrate that late intervention for CLP leads to an improved quality of life, irrespective of the limited change in perceptual speech described in the literature. Discussions also suggest that barriers to intervention need to be overcome and treatment options for CLP in older patients should be extended to include the provision of secondary care in line with international standards for treatment of CLP.
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<tr>
<td>ASHA</td>
<td>Accredited Social Health Activists</td>
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<tr>
<td>CLP</td>
<td>Cleft lip and/or palate</td>
</tr>
<tr>
<td>CP</td>
<td>Cleft palate</td>
</tr>
<tr>
<td>CSAG</td>
<td>Clinical standards advisory group</td>
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<tr>
<td>DALYs</td>
<td>Disability adjusted life years</td>
</tr>
<tr>
<td>EUROCAT</td>
<td>European Surveillance of Congenital Anomalies</td>
</tr>
<tr>
<td>GBD</td>
<td>Global burden of disease</td>
</tr>
<tr>
<td>GDP</td>
<td>Gross domestic product</td>
</tr>
<tr>
<td>ICBDMS</td>
<td>International Clearing House for Birth Defects Monitoring System</td>
</tr>
<tr>
<td>ICC</td>
<td>Intra-class correlation</td>
</tr>
<tr>
<td>ICD</td>
<td>International Classification of Disease</td>
</tr>
<tr>
<td>ICDH</td>
<td>International Classification of Impairments, Disabilities and Handicaps</td>
</tr>
<tr>
<td>ICF</td>
<td>International Classification of Functioning, Disability and Health</td>
</tr>
<tr>
<td>ICP</td>
<td>Isolated cleft palate</td>
</tr>
<tr>
<td>IMF</td>
<td>International monetary fund</td>
</tr>
<tr>
<td>IPDTOC</td>
<td>International Perinatal Database of Typical Orofacial Clefts</td>
</tr>
<tr>
<td>LIC</td>
<td>Low income countries</td>
</tr>
<tr>
<td>LMIC</td>
<td>Lower-middle income</td>
</tr>
<tr>
<td>MDT</td>
<td>Multi-disciplinary teams</td>
</tr>
<tr>
<td>MIC</td>
<td>Middle income countries</td>
</tr>
<tr>
<td>NBDPN</td>
<td>National Birth Defects Prevention Network</td>
</tr>
<tr>
<td>NGO</td>
<td>Non government organisation</td>
</tr>
<tr>
<td>OOP</td>
<td>Out of pocket payments</td>
</tr>
<tr>
<td>PI</td>
<td>Primary investigator</td>
</tr>
<tr>
<td>QOL</td>
<td>Quality of life</td>
</tr>
<tr>
<td>SLT</td>
<td>Speech and Language Therapist</td>
</tr>
<tr>
<td>SSI</td>
<td>Semi-structured interviews</td>
</tr>
<tr>
<td>TOM</td>
<td>The therapy outcome measure</td>
</tr>
<tr>
<td>TOM-clp</td>
<td>The Therapy Outcome Measure adapted for cleft lip and palate</td>
</tr>
<tr>
<td>WHO</td>
<td>World health organisation</td>
</tr>
</tbody>
</table>
PREFACE

Justification for this research

Cleft lip and palate can present tough physical, emotional and social challenges even with the best care in the world (CSAG, 1998, Shaw et al., 2005). For individuals born in rural India, lack of understanding and limited availability of care makes these challenges harder, and leaves many unsupported and untreated (Campbell et al., 2014, Mossey and Little, 2009a, Ward, 2014). Conflicting evidence on the efficacy of late intervention for these older individuals potentially restricts their access to care, however, there are limitations in this evidence which could be leading to an underestimation of the benefit of treatment later in life, particularly in relation to psychological well being and social inclusion. The goal of this research is to establish the barriers which prevent individuals obtaining care in India and gain a better understanding of the psychosocial impacts which can result when CLP is treated later in life.

The concept of this research emerged whilst working in the Philippines where personal encounters with individuals with untreated and late treated CLP indicated that this cohort was underserved. Anecdotal evidence indicated that, whilst untreated they felt marginalised from their community, depressed and unable to realise their true potential, whereas post surgery they had greater self-esteem, felt included and had improved prospects from a personal, social and financial perspective. This prompted a provisional research project in Karnataka, India, to assess the requirements of an outcome measure to gauge psychosocial changes following late intervention for CLP.

This research explores the viability of implementing an adapted version of the TOM, the TOM-clp (Rees et al., 2016b), in four cleft centres in India and compares the results with qualitative data derived using framework analysis on 40 interviews with individuals undergoing late repair at three of the centres.
The aims of this study are threefold:

- To identify the underlying reasons why cleft lip and/or palate (CLP) remain untreated in India and to explore what services are currently available for CLP repair, how are these services promoted and what facilitates or limits their accessibility?

- To explore the psychosocial impact of untreated CLP for individuals and investigate the impact of late repair on activity, participation and well-being from the perspective of the individual and close friends and family.

- To examine the value and viability of an adapted TOM to explore the impact of untreated CLP and evaluate how well the TOM-clp can be implemented as a culturally sensitive tool to demonstrate change in relation to intervention?
Focus

This thesis is divided into a preface and 10 chapters followed by a reference and appendices section: The preface introduces the terminology used in this thesis and explains the choice of the term 'developing world' to describe regions where untreated CLP prevails. Chapter 1 will give an insight into the presentation of CLP, its aetiology, global epidemiology and management strategy. This will be followed by a chapter describing the impact of CLP and the consequences of limited treatment opportunity in the developing world. Chapter 3 compares and contrasts the research relating to late intervention available to date and discuss why there are limitations in many of these studies which may be leading to this cohort being under served. This is followed by a chapter which outlines the requirements of an ideal outcome measure and discusses the advantages and disadvantages of instruments currently available for the assessment of outcome for CLP management. Chapters 5, 6 and 7 detail the methodology, design and procedure associated with this research and are followed by a chapter describing the development of the TOM-clp. Chapter 9 presents the results of the demographic data, the framework analysis and the TOM-clp data respectively which is followed by a chapter including a discussion of the results and alluding to strengths and limitations of the study and a conclusion including the projected value of this research. Additional documentation and expanded details of the data will be included as appendices following the bibliography.
Terminology and glossary of terms

Choice of terminology can prove controversial in academic writing so this brief section will provide some background information behind the chosen terminology for this thesis, followed by a glossary of terms for cross reference purposes.

History of terminology for the developing world

Finding the correct terminology to ensure empathy and compassion whilst avoiding controversy has in itself become controversial. Whilst it is appreciated that, owing to the ranging diversity within any large group, labelling and categorising will never be wholly perfect, it does serve a function by allowing similarity to be discussed en masse. However, in doing so there is a risk of insinuating that all members of the group are similar in other ways which may not be wholly accurate. There is little surprise therefore that such activity is often considered branding and can be seen as offensive if not carefully considered. This has been clearly demonstrated when discussing multifarious geo-political, economic and developmental global comparisons.

One of the first global classifiers to receive wide media attention was the term 'third world'. This term was first coined by the Alfred Sauvy in 1952. Sauvy, a French economist, was considering an economic model where wealthy countries were wrestling for power at the expense of poorer nations. This was the time of the cold war and he divided countries into three categories or 'worlds'; the first 'world' related to capitalist countries, mainly the USA but also Western Europe, the second 'world' consisted of communist countries, mainly targeting the Soviet Union and China, which left the remainder as the 'third world' (Tomlinson, 2003, Wolf-Phillips, 1987). The classification was intended to describe geo-political status; however, because many of the countries in the Third World category were impoverished, it gradually came to insinuate that the latter group were poor and third class. Unsurprisingly the term lost favour, particularly with wealthy nations in the Middle East and Australasia who were affluent and progressive but implicated by this term (Solarz, 2014).
The terms Developing World / Developing Countries became popular to describe nations where educational establishments and healthcare services needed to develop in order to better serve their population. It is still popular with many, such as the World Health Organisation (WHO), since it encompasses countries such as India where wide social and economic inequality is apparent despite rapid globalisation and industrial expansion. However, in the eyes of many the term has connotations of being backward and irresponsible and thought to be too hierarchical. The United Nations (UN) tends to have preponderance for the term Less developed nations but this is also seen by many to have negative connotations.

The term Global South was a product of the United Nations Global Migration Database to describe movements between continents but is often been taken as synonymous with developing world (Rigg, 2007,Dados and Connell, 2012). It is widely used in political and economic circles but has obvious inaccuracies and inequalities since it includes rich countries such as Australia, New Zealand and Argentina in the Southern Hemisphere as well as more impoverished nations (Hollington et al., 2015).

The World Bank and International Monetary Fund (IMF) classify countries according to fiscal wealth based on global economic statistics. This divides countries into low income (LIC), lower-middle income (LMIC) and middle income (MIC) according to gross domestic product but gives little information regarding social status. More recently the term Majority World has become popular based on the fact that 80%, the majority, of the world’s population survives on less than ten dollars a day. The term has become popular as an indicator of low family income whilst still appreciating the wealth of culture within these communities.

There is unlikely to be total agreement on the use of terminology and different terms may be appropriate for certain circumstances but not others depending on the connotation. In the words of John Lydgate “You can please some of the people all of the time, you can please all of the people some of the time, but you can’t please all of the people all of the time”. Since the core of this thesis relates to issues of disparity and inequity of health and welfare provision and the foundation of the TOM-clp lies in the WHO-International Classification of Functioning, Disability and
Health (WHO-ICF) a decision has been made to use the terms 'developing countries' and the 'developing world' in accordance with the terminology choice of the WHO.

**Glossary of terms**

<table>
<thead>
<tr>
<th>Term</th>
<th>Definition</th>
</tr>
</thead>
<tbody>
<tr>
<td>Abduction</td>
<td>A comment supported by both induction and deduction so the conclusion is derived from all known information</td>
</tr>
<tr>
<td>Aetiology</td>
<td>The cause or reason behind a disease or condition</td>
</tr>
<tr>
<td>Analytical framework</td>
<td>A framework of codes organised into categories to organise qualitative data</td>
</tr>
<tr>
<td>Analytical memo</td>
<td>A reflection on an issue emerging from the data</td>
</tr>
<tr>
<td>Atomistic ontology</td>
<td>Philosophical theory that the universe is composed of an infinite number of building blocks.</td>
</tr>
<tr>
<td>Case</td>
<td>Individual participant</td>
</tr>
<tr>
<td>Categories</td>
<td>Clusters of codes based on similar or interrelated ideas</td>
</tr>
<tr>
<td>Charting</td>
<td>Entering summarised data into the framework</td>
</tr>
<tr>
<td>Code</td>
<td>Descriptive label assigned to raw data</td>
</tr>
<tr>
<td>Data</td>
<td>1. Qualitative textual data and observations</td>
</tr>
<tr>
<td>Deductive reasoning</td>
<td>Testing a theory and deriving a conclusion by logical reasoning</td>
</tr>
<tr>
<td>Genotype</td>
<td>Genetic makeup of an organism</td>
</tr>
<tr>
<td>Heterogeneous</td>
<td>Sample consisting of different or assorted parts</td>
</tr>
<tr>
<td>Homogeneous</td>
<td>Sample consisting of parts all of the same kind</td>
</tr>
<tr>
<td>Indexing</td>
<td>Systematic application of codes</td>
</tr>
<tr>
<td>Induction</td>
<td>Deriving meaning from what is observed</td>
</tr>
<tr>
<td>Inductive reasoning</td>
<td>Making broad generalization as a result information emerging from observation</td>
</tr>
<tr>
<td>Matrix</td>
<td>Spreadsheet summarising cases (rows) and codes (columns)</td>
</tr>
<tr>
<td>Metaphysics</td>
<td>The branch of philosophy that deals with the first principles of things, including abstract concepts regarding the nature of reality</td>
</tr>
<tr>
<td>Non-syndromic cleft</td>
<td>Cleft arising as an isolated event not in association with a known syndrome</td>
</tr>
<tr>
<td>Ontological</td>
<td>The study of existence</td>
</tr>
<tr>
<td>Orofacial</td>
<td>In relation to the face and mouth</td>
</tr>
<tr>
<td>Pathogenesis</td>
<td>The origin and development of a disease or condition</td>
</tr>
<tr>
<td>Phenotypes</td>
<td>The physical appearance and behaviour of an organism based on its genotype and environment.</td>
</tr>
<tr>
<td>Syndromic cleft</td>
<td>Cleft arising in association with a known syndrome</td>
</tr>
<tr>
<td>Themes</td>
<td>Proposed concepts based on merging of coded data</td>
</tr>
<tr>
<td>Transcript</td>
<td>Verbatim account of interview process including observation notes</td>
</tr>
</tbody>
</table>
CHAPTER 1: INTRODUCTION

1.1. Electronic bibliographic review

Aim – to gather information on factors which influence the incidence and prevalence CLP, outcome measures linked with CLP and literature pertaining to late intervention for CLP with a focus on India.

The preliminary review of the literature involved the search engines; MEDLINE, CINAHL, OVID, Cochrane library and Google Scholar. Where possible non-English papers were translated using Google-translate and bibliographic reference searches were undertaken to ensure that papers not highlighted by the electronic search were also included. Sources included; Peer reviewed journals, academic books, conference proceedings, policy documents from the Government of India, WHO and Clinical Standards Advisory Group (CSAG); information on demographics and service provision from non-government organisations such as Smile Train and Operation Smile; statistical data from web sources including Geohive and Nationmaster. The first searches began in September 2013 but were repeated on a regular basis until December 2017 to ensure the literature remained up to date.

Searches relating to background information such as incidence, prevalence and outcome measures was restricted to articles published since 2010 but was expanded to include earlier publications where appropriate. An open inclusion criteria was followed to search for literature relating to un-treated and late operated CLP with articles being traced using free text searches in 'all-fields' for any year. Multiple synonyms, wildcards, truncations, Boolean operators and cited works were employed to ensure as wide a search area as possible. Where full text was not available directly documents were sought either through research gate from the author directly or via the University and British Libraries.
1.2. CLP Background

This chapter start with an overview of the current thinking behind why clefts develop, aetiological influences and standard management of the condition.

1.3. Development

Orofacial clefts are a heterogeneous group of disorders, including CLP and isolated cleft palate (CP) which are considered one of the most common congenital anomalies, affecting over ten million people worldwide (Mossey et al., 2009, Mossey, 2007, Doray et al., 2012). They develop between the fourth and eleventh weeks of pregnancy as a result of the maxillary and nasal processes failing to fuse causing incomplete formation of the face and oral cavity (Berkowitz, 2006, Diego, 2002). The severity and appearance of the cleft depends on which part of the embryological process is deficient and ranges from an almost imperceptible notched lip or submucous cleft palate to a bilateral cleft lip and palate or midfacial cleft (Figure 1) (Dixon et al., 2011).

Seventy percent of CLP and fifty percent of CP arise as isolated anomalies and are referred to as non-syndromic, the remainder arise in combination with additional congenital anomalies and as such are known as syndromic (Schutte and Murray, 1999, Genisca et al., 2009, Jugessur et al., 2009).

1.4. Gender ratio

CLP is thought to occur more frequently in males, with a male: female ratio of 1.8:1, whereas isolated CP has been reported as slightly more common in females, with a male: female ratio of 0.93:1 (Mossey and Catilla, 2003), although other studies report an even distribution (Christensen et al., 1992).
1.5. Pathogenesis

The exact pathogenesis of oral clefting is complex and not fully understood however, it is known to be multifactorial, involving a complex combination of genetic and environmental factors (Agbenorku, 2013, Dixon et al., 2011, Berkowitz, 2006).

1.6. Genetic component

International collaboration and advances in molecular biology and genomics have substantially increased our understanding of CLP and lead to the identification of
more than 275 associated syndromes (Rahimov et al., 2012, Amberger et al., 2015). Our knowledge regarding non-syndromic clefts is less precise; traditionally CLP and CP have been considered as two distinct phenotypes with divergent aetiologies (Mossey et al., 2009), with recent studies indicating the possibility that isolated cleft lip also signifies a specific phenotype (Harville et al., 2005); Other schools of thought consider all three to represent a spectrum of genetic variance rather than explicit independent entities (Grosen et al., 2010, Ludwig et al., 2012). So, whilst is generally agreed that there is a strong genetic element in the formation of oral clefts, further research is required before it can be used more widely to reduce the risk of orofacial clefting in clinical practice (Rahimov et al., 2012, Grosen et al., 2010, Jugessur et al., 2009).

1.7. Environmental component

There is mounting evidence that numerous environmental factors influence the development of orofacial clefts a few of which are outlined below (DeRoo et al., 2008, Boyles et al., 2010, Rahimov et al., 2012).

- **Tobacco products**

Both active and passive exposure to tobacco, including nicotine replacement therapy and snuff, have been shown to significantly increase the risk of developing CLP, however, the risk associated with isolated CP is less conclusive (Gunnerbeck et al., 2014, Shi et al., 2008, Little et al., 2004, Honein et al., 2007). The danger is highest during early foetal development and thought to be more pronounced with snuff products than smoking (Gunnerbeck et al., 2014). In vitro studies have shown that it is the nicotine which inhibits embryonic plate fusion and the affect is dose dependent (Baroni et al., 2010). This is important because, although it remains important to encourage smoking cessation during pregnancy, nicotine replacement therapy may potentially pose a greater risk than conventional cigarette smoking, since the duration of nicotine delivery is far longer (Gunnerbeck et al., 2014, Coleman et al., 2012).
• **Low socio-economic and educational status**

Low socio-economic and educational status has been implicated in increased incidence of orofacial clefting (Womersley and Stone, 1987, Yang et al., 2008, Reddy et al., 2010). However, this cohort also has an increased preponderance for nutritional deficiency, smoking and consanguineous marriage which, as outlined below, are all risk factors associated with CLP, so it is difficult to extrapolate specific components from the data.

• **Dietary deficiencies**

Anaemia and vitamin deficiencies including low folic acid, iron and zinc are thought to play a role in the development of clefts, however the evidence remains inconsistent and inconclusive (Wu et al., 2010, Wehby and Murray, 2010, Munger et al., 2009, Tamura et al., 2005, Yazdy et al., 2007, Johnson and Little, 2008). In India, a staggering 40-60% of pregnant women are considered anaemic due to poverty, poor diet, multi-parity and hook worm infestations (WHO, 2015a) and Neogi et al. (2017) demonstrated a strong link with development of CLP amongst parents in India who were vegetarian which they linked to folate and B12 deficiency. If these implications are correct, they could have a significant bearing on the incidence of orofacial clefting particularly in less affluent and poorly educated communities.

• **Consanguineous marriage**

Consanguineous marriage, where siblings or first cousins are married, has consistently been shown to increase the risk of CLP (Elahi et al., 2004, Sadri and Ahmadi, 2007, Reddy et al., 2010, Sabbagh et al., 2014). Although consanguinity is rare in western culture, it accounts for over 50% of marriages in parts of middle eastern society (El-Hazmi et al., 1995, Ravichandran et al., 2012) and 20- 45% in some Indian communities (Dutta et al., 2010, Bhagya, 2013, Sah et al., 2016) which could account for some regional elevation in orofacial clefting.
1.8. Incidence and prevalence of CLP

The exact incidence of orofacial clefting is much debated, but is generally considered to occur between 1 in 500-1200 live births (Mossey and Little, 2009a, Wyszynski et al., 1996, Sommerlad, 2002, Kadir et al., 2017, Tanaka et al., 2012). Figures fluctuate widely in the literature but generally indicate a substantial regional variation, suggesting that orofacial clefting occurs most commonly in Asian populations, least frequently in African populations with a Caucasian population featuring almost equidistant between the two (Mossey and Catilla, 2003). However, closer analysis of the literature reveal that apparent differences in incidence and prevalence may well relate as much to the plethora of different inclusion criteria and research methodologies than ethnic origin alone (Cooper et al., 2006, IPDTOC Working Group, 2011, Kadir et al., 2017). It is important to understand these confounding factors in order to establish an accurate approximation of incidence and prevalence of orofacial clefting and develop management strategies on a global scale (Cooper et al., 2006) and a few will be outlined below.

1.9. Source of the data

A great deal orofacial cleft data is collected in a hospital setting or in specialist referral centres (Murray et al., 1997). This can be beneficial, in that there is likely to be qualified expertise, which should result in more accurate diagnosis and documentation than might be expected in a non-specialist unit or rural community setting. For example, research which relies on information collected in non-specialist centres may fail to include certain palatal clefts which may evade detection at birth since they are not externally visible (Cooper et al., 2006, Mossey et al., 2009, Habel et al., 2006, Tsangaris et al., 2017). Conversely, a potential problem with hospital based studies is that, in many countries the majority of births occur in the home or rural community setting, so data collected in hospitals will not be representative of the population as a whole (Mossey and Catilla, 2003); for example, according to UNICEF, approximately 25 million children are born in India every year, assuming the incidence of CLP/CP to be 1:500, this corresponds to 50,000 new cases of CLP/CP annually. It is estimated that more than two thirds
of these children will be born away from hospital (Narang et al., 2013, Prinja et al., 2014) which presupposes that 33,000 cleft cases would remain unaccounted for in hospital data. Since many parents are unaware of the possibility or potential importance of seeking treatment, particularly in relation to CP (Wilson and Hodges, 2012), they may well remain excluded from hospital based research (Dreise et al., 2011, Cubitt et al., 2014).

This fact is corroborated by a study by Dvivedi & Dvivedi (2012) at a cleft clinic in Northern India, which reported that three quarters of the cleft patients presenting for treatment were home births from rural families living below the poverty line. It is possible that even these data underestimates the problem, as the figures do not account for individuals who failed to thrive or remained undiagnosed (Campbell, 2014a; Dutta & Bawari, 2007). Since patient of low socio-economic status are more likely to have poor nutrition, dietary deficiencies, consanguineous marriage and multiple partum they are at statistically increased risk of orofacial clefting (Reddy et al., 2009) this suggests that hospital based data is likely to represent a considerably lower incidence of clefting than would be expected of a more representative national population.

1.10. Inclusion criteria

Studies vary widely in their inclusion/exclusion criteria which make the results difficult to compare: Incidence statistics are often quoted in terms of live births, however for many studies it is unclear whether perinatal mortality is included whereas data from other studies might specifically include or exclude data from miscarriage, abortion, still birth and neonatal deaths. This is particularly pertinent because, children born with orofacial clefts have a ten to fifteen fold higher mortality rate than the general population, even where state-of-the-art treatment facilities are readily available (Kang et al., 2012, Druschel et al., 1996). Mortality for syndromic clefts, particularly where there is a major associated anomaly, are often reported with even higher rates of mortality, ranging between 16 - 34% (Bell et al., 2015, Hujoel et al., 1992, Kang et al., 2012). Exclusion of this cohort could therefore result in a significantly lower reported incidence rate than might be expected when compared with inclusive studies.
Studies which rely on retrospective information from birth records, neonatal records, specialist assessments, postmortem and school records are difficult to compare with a general population since there is often targeted sampling and a assumption of complete and accurate record keeping (IPDTOC Working Group, 2011, Honein et al., 2001). A study scrutinizing the accuracy of cleft documentation on birth certificates in California identified that more than 50% of clefts diagnosed clinically were not represented whereas 20% were stated as present on the birth certificate but not clinically (Hexter et al., 1990). These results were from a state-of-the-art hospital so opens the question of the accuracy of retrospective CLP data collection, particularly from less established facilities.

1.11. Terminology

Studies often pay little regard for separating the different cleft types; isolated cleft lip (ICL), CLP and isolated cleft palate (ICP) which, as mentioned, are generally considered to have distinct aetiologies. Even where internationally accepted coding systems such as the WHO classification of disease (ICD-10) are implemented, reliability is questionable; systematic reviews of National Health Service (NHS) data collection using 3 figure ICD-10 codes estimate an accuracy of 84 - 87% (Campbell et al., 2001, Burns et al., 2012) but was as low as 50%, across specialties for four figure codes (Cleary et al., 1994). Four figure codes are required to differentiate between cleft types, so data selected in this way may well be of limited accuracy. In addition, ICD-10 become less reliable where more than one defect is recorded separately and may fail to denote minor anomalies such as lip pits or submucous clefts which may not be clinically visible.

1.12. Global databases

The difficulty of such widely varying data means that making an accurate global estimation of incidence of orofacial clefting extremely difficult. In order to introduce some parity there are now many international registries such as International Clearing House for Birth Defects Monitoring System (ICBDMS), European Surveillance of Congenital Anomalies (EUROCAT) and National Birth Defects Prevention Network (NBDPN) aimed at collecting more accurate
information (Mossey and Catilla, 2003). In an attempt to assimilate this information on a global scale a collaborative project International Perinatal Database of Typical Orofacial Clefts (IPDTOC) has been initiated by The World Health Organisation (WHO) as part of its human genetic programme, however even these show unexplained variations in data (IPDTOC Working Group, 2011). Whether this is due to geographic clustering or indicative of erroneous information is unclear however, it is hoped that, in the long term, the collaboration of data collection will help to clarify the true extent of the global burden of orofacial clefting (Druschel et al., 1996).

1.13. Gold standard for management of CLP/CP

A great deal of research and expertise has been directed at perfecting intervention for individuals with CLP/CP, and there is a general Global agreement regarding best practice for its management: The considered gold standard demands; specialist surgical intervention within the first year of life and continuing multidisciplinary support throughout childhood and adolescence into adulthood (CSAG, 1998, Shaw et al., 2001). Such a team should include surgical specialties, speech and language therapy (SLT), dental and orthodontic services, specialist nursing, ear nose and throat (ENT), audiology and psychological support as a minimum (CSAG, 1998, Shaw et al., 2001). This approach takes into consideration not only the physical and medical aspects of CLP/CP but also appreciates and addresses the psychosocial connotations which are associated with the condition.

This treatment policy is generally accepted globally leading to excellent outcomes and good prospects for many individuals with CLP/CP. However, in many developing countries such timely and comprehensive care is not always possible and many individuals with CLP remain un-treated into adulthood (Murthy et al., 2010, Adeyemo et al., 2009, Schwarz and Bhai Khadka, 2004). The following chapter will discuss the implications of living with untreated CLP with a focus on India.
CHAPTER 2: IMPLICATIONS OF CLP AND CP

This chapter starts by outlining the general impacts of CLP at a personal, community and global level. This is followed by a more detailed section pertaining to CLP in India and a vignette describing the life of a child born into a rural family in Assam which is designed to help put a personal perspective on the problems associated with CLP in the developing world.

2.1. Living with CLP

The impact of orofacial clefting on the child and family becomes apparent from the moment of birth and can endure throughout the life of the individual. The effects of CLP can be considered in three basic categories; direct functional difficulties and indirect psychosocial and financial problems.

2.2. Functional Impacts

Apart from the immediate aesthetic implication of CLP, the first major hurdle for parents to overcome is the difficulty in suckling and swallowing which can prove fatal if not managed correctly (Schreiber Willcox, 1994, Pandya and Boorman, 2001, Reid et al., 2007, Cubitt et al., 2012). Such problems are now less of a problem for children born in areas where trained staff and resources are available to assist and educate new mothers (Reid, 2004). Unfortunately feeding devices and guidance are generally poorly disseminated into rural communities (Lang et al., 1994) therefore the inability to feed remains a common cause of infant death or failure to thrive in many parts of the developing world (Schreiber Willcox, 1994, Pandya and Boorman, 2001, Tungotyo et al., 2017, Mandrano et al., 2017). Wilson and Hodges (2012) propose that the frequency of infant death due to malnourishment might account, in some part, for lower reported incidence of CLP in some African countries.

The implications of CLP on speech and language development are well documented (McWilliams et al., 1990, Chapman, 1993, Schulz et al., 2015, Oyama et al., 2016, Sell et al., 2001) and will be discussed further in relation to untreated CLP in the following chapter. CLP also makes children susceptible to otological
pathology and a study by Møller (Møller, 1981) revealed that almost 100% of participants in their study suffered hearing and vestibular problems at some point during their childhood. If undiagnosed and left untreated this can lead to long term hearing problems and deafness in adulthood which, in turn, accentuates difficulties with speech and communication (D’Mello and Kumar, 2007). Individuals with CLP also commonly suffer nasal regurgitation of food and fluid which can be both problematic and embarrassing (Sadhu, 2009, Cohen et al., 1991).

2.3. Psychosocial impacts

Psychosocial issues relating to CLP have been widely documented, even for individuals who get timely intervention for their cleft: Ramstad et al. (1995) reported reduced psychological well-being amongst young adults with clefts in Scandinavia, whilst others report low self-esteem and difficulty relating to peers (Cochrane and Slade, 1999, Sandy et al., 2001, Pillemer and Cook, 1989). CLP has been associated with behavioural inhibition and difficulty assimilating into society and family life (Richman and Eliason, 1982, Hunt et al., 2005, Kapp-Simon et al., 1992, Strauss et al., 2007), also depression and anxiety relating to problems with communication (Millard and Richman, 2001). Communication difficulties can lead to bullying and marginalisation, which subsequently segregation hampers individuals ability to make friends, participate normally in education and ultimately restricts their ability to realise their true potential in terms of social and financial autonomy (Blas et al., 2011, Wehby et al., 2006, Wehby and Cassell, 2010). Although such marginalisation spans a wide spectrum of society it may be more pronounced in underdeveloped regions (Nayar, 2007) promoted by many factors including gender, caste, age and poverty (Nayar, 2007, Peter et al., 1975, Hunt et al., 2005, Pelchat et al., 1999).
2.4. Financial impacts

The personal financial implications of a CLP are multifarious and the effects are often both direct and enduring. For many, the cost of attending hospital appointments can prove prohibitively expensive due to travel, accommodation, hospital fees and loss of earnings (Hodges and Hodges, 2000, Singh, 2009, Sitzman et al., 2016). In the long term, if children are kept in isolation owing to family embarrassment and lack of community acceptance, they will have limited access to education and may fail to reach their educational, employment and social potential (Raju, 2000, Brydon et al., 2014). As a consequence, they may never become financially autonomous and become domestically reliant on their family for the remainder of their lives (Weatherley-White et al., 2005, Gupta et al., 2010, D’Antonio and Nagarajan, 2003).

Financial implication of CLP can also be considered in terms of its Global Burden (GBD) to the Nation and has been analysed through economic modelling (Magee et al., 2010). This type of analysis considers the mortality and morbidity associated with CLP with respect to region, age and gender and socioeconomic variables (Murray and Lopez, 1996, Mathers and Loncar, 2006, WHO, 2006). By comparing the disability adjusted life years (DALYs), estimated the GBD related to CLP in a well established cleft unit in Nepal, Corlew (2010) demonstrated that the improved financial position and well-being attained after intervention for CLP, had a significant micro-economic gain as compared with the burden of the disability (World Health Organization, 2002b, Corlew, 2010). This technique has been repeated in a number of other locations with similar projections, showing cost-benefit favouring intervention for this cohort (Corlew, 2010, Alkire et al., 2011a, Alkire et al., 2014, Poenaru, 2013, Hughes et al., 2012). Although this type of modelling is contentious as it is considered simplistic and makes broad assumptions in terms of weighting criteria (Corlew, 2010), the figures indicate that providing intervention for CLP and CP can reduce the national financial burden, which would be associated with reduced productivity, and prove economically beneficial in the long term.
2.5. Cleft lip and palate: An Indian perspective

Despite the ruling of the Indian Supreme Court guaranteeing the right to health (Ranjan, 2014), the social circumstance and geography of the population makes this extremely difficult to achieve (Ward, 2014). As mentioned in section 1.8, approximately 50,000 children are born with CLP/CP each year, which represents a huge burden of clinical care (Mossey and Little, 2009a, Singh, 2009) so, whilst excellent services exist for treatment of CLP in many areas throughout India, a relatively high percentage of cases remain untreated (Campbell et al., 2014).

There are many reasons for individuals not receiving treatment; undoubtedly there is a lack of both physical and financial capacity within the health system to cope with the high volume treatment need which affect both individuals and service providers alike (Singh, 2009, World Health Organization, 2002a, Mossey, 2004, Campbell et al., 2014). According to the 2011 Indian census, 68% of the population live in rural and remote areas, so may lack the time or means to travel the great distances required to seek medical care. Many studies have identified behavioural characteristics and cultural factors such as caste and gender preferences and social stigma which inhibit families’ willingness to seek intervention (Schwarz and Bhai Khadka, 2004, Feinstein, 1993). Naram, Makhijani et al. (2013) undertook a survey of one of the cleft centres in Hyderabad and in agreement with other researchers (Olasoji et al., 2007, Oginni et al, 2010, Olaitan, 2012), they found that poor education and superstition played a major role in limiting access to cleft care. They identified that 50% of their cohort thought that seeing a solar eclipse during pregnancy had caused the CLP and 13% thought it was the will of the Gods. Such cultural beliefs can be highly influential in inhibiting people from seeking treatment in India; when families believe that CLP was an act of the God's, they often display a passive acceptance, assuming it as their fate (Weatherley-White et al., 2005, Loh and Ascoli, 2011, Oginni et al., 2010) or they believe that seeking surgery will interfere with God's will and they risk being punished further (El-Shazly et al., 2010). Considering the genetic component of CLP (Rahimov et al., 2012, Marazita, 2012), this means that many families go on to have a second child with CLP, which, for them, merely perpetuates their way of thinking. Fortunately there do not appear be such connotations placed on the solar
eclipse, so, whilst this is a most commonly held superstition, it is not perceived as a barrier to treatment (El-Shazly et al., 2010, Naram et al., 2013). Such superstition can be so strongly held as to lead to a child and family being isolated and shunned from society, accentuating their deep seated guilt and shame over the defect (Weatherley-White et al., 2005, Raju, 2000, Oginni et al., 2010). This can also make them disinclined to seek assistance (Udwadia, 2003) and increases the dropout rate for secondary surgery such as palate repair (Hodges and Hodges, 2000, Gopalakrishna and Agrawal, 2010).

Compounding these reasons for not seeking intervention is the overriding issue of India’s vibrant but inequitable economy and imbalanced health care provision. India has a population of over 1.3 billion and in the last 15 years the Gross Domestic Profit (GDP) has risen from 500,00 billion to a staggering 1.85 trillion US$, making it the fourth largest economy in the world (World Bank, 2016). However, this rapid economic growth has not resulted in nationwide prosperity, rather the biased distribution of wealth has merely exaggerated the existing disparity between rich and poor (Wagstaff, 2002). India allocates only 4.0% of its GDP to health care (World Bank, 2016) and only 0.9% of those funds support the public health sector, the remainder being allocated to private health care, which does little to serve the poorest in the population (Kumar et al., 2011, Mishra et al., 2009, Sanneving et al., 2013).

Thus, despite India consistently featuring in the top 10% of economies worldwide for the last 20 years, a lack of infrastructure, restrictive labour regulation and fiscal debt has lead to an immense variation in the socio-economic profile (World Bank, 2016). Disproportionate wealth, in conjunction with a huge wave of globalization, as the population migrates towards towns and cities in search of work and opportunities, has widened the gap still further. According to the Multidimensional Poverty Index, based on indicators of educational, health and well-being, the percentage of the population living below the poverty line lies at approximately 45% (Alkire et al., 2011b). Such inequalities are particularly striking in relation to health care and clearly illustrated by the infant mortality figures where the discrepancy between rich and poor stand at 34 and 82 per 1000 respectively, a
figure comparable with some of the world’s poorest nations (Chatterjee, 2010, Wagstaff, 2002, Balarajan et al., 2011, World Bank, 2016).

In 2015 the Indian Government drafted its goal of providing "universal access to good quality health-care services without anyone having to face financial hardship as a consequence" (Ministry of health & family welfare, 2015, Patel et al., 2015). However, this ideal is not yet a reality and since only a small percentage of people in India can afford health insurance, the majority of individuals rely on Out Of Pocket Payments (OOP) to fund their healthcare; in 2000/2001 an estimated 70-80% of total health expenditure in India was raised directly from the population in the form of OOPs (Kumar et al., 2011, WHO, 2006, Patel et al., 2015). To the individual these expenses often represent a substantial proportion of household spending and become unmanageable if there is a sudden unexpected cost such as a difficult birth or a requirement for surgical intervention such as for CLP (Russell, 2004, Prinja et al., 2014). For an underprivileged family with a low or unreliable income these payments often escalate their poverty causing a downward spiral of events leading to catastrophic debt from which they may not recover (Van Doorslaer et al., 2006, Sanneving et al., 2013). In addition, increased poverty means a reduction in general household expenditure, poorer diets, restrictions on education and employment and an increasing likelihood of poor health and further poverty. This vicious circle widens the socio-economic gap still further and in the words of Wagstaff (2002) their 'poverty breeds ill health and ill health maintains poverty'. This demonstrates that, whilst India’s economy is flourishing the same cannot be said for the health of its less affluent population. In order to explore these concepts further, let’s consider a poor family living in rural Assam in India’s North East who experience a sudden and unexpected expenditure due to the birth of a child with CLP.
2.6. Benorika’s story: A child born with CLP in rural Assam

Assam sits south of the Himalaya and shares an International border with Bhutan and Bangladesh. It has a population of 31 million and is famous for its tea production and fine silks. The State relies heavily on agriculture and 87% of its population live in rural communities, 36% below the poverty line (Dutta and Bawari, 2010).

A population survey in 2005 revealed that 65% of the population are illiterate, 70% of the women in Assam are anaemic due to poor diet and multiple pregnancies, 72% chew tobacco, 60% use alcohol and consanguineous marriage is commonplace (Medhi et al., 2006). All these factors increase the risk of having a child with CLP/CP in a region where resources for obtaining treatment are restricted (Mossey and Little, 2009a). In addition, their low educational status and high illiteracy levels mean they are likely to have little knowledge regards health in pregnancy (Khan et al., 2004), be unlikely to seek or be offered antenatal checks and have little awareness of what management options are available for CLP (Pallikadavath et al., 2004).

When Benorika was born, her family had never seen a child with CLP before. They were from a remote village and both her parents were uneducated, surviving on the dollar a day her father earned as an agricultural labourer. Superstition was endemic and they believed that God was inflicting a punishment on them for previous wrongdoings (Weatherley-White et al., 2005) and they were scared that the family would be rejected and ostracised so were reluctant to take her out of their home (Weatherley-White et al., 2005, Loh and Ascoli, 2011, Ross, 2007, Reddy et al., 2009).

Their immediate problem was that Benorika was unable to suckle, when they tried using a bottle or spoon instead, the milk just poured from her nose (Reid, 2004). In contrast to babies born with CLP in wealthy Indian urban households, where outcomes are often over and above International standards, they did not have
access to state-of-the-art hospital facilities or a full complement of specialist care or any advice (Govindarajan and Ramamurti, 2013). They were never told about using specially adapted bottles and formula and, even if they had been, they could not have afforded such 'luxuries', even at the risk of their child failing to thrive (Schreiber Willcox, 1994, Pandya and Boorman, 2001, Lang et al., 1994).

Benorika was lucky and the persistence of her mother expressing milk and spending long hours helping her feed meant that she survived. The family’s worry over marginalisation were unfounded and, although Benorika was often teased for her appearance and poor speech, she was accepted into school and made friends. Nobody in the village was aware that treatment was a possibility and, as for many in the same situation, they accepted their fate for passively for fifteen years (Udwadia, 2003).

When they finally discovered that Benorika could be treated, the concept brought with it joy, trepidation and a series of additional worries; they lived many hundreds of kilometres from Guwahati which meant they would have to travel longer in order to reach treatment facilities than urban dwellers would (Akin and Hutchinson, 1999), treatment implied relatively high transportation expenses, lost wages, and accommodation costs which they could ill afford (Govindarajan and Ramamurti, 2013, Wagstaff, 2002). The family took the opportunity and attended for the first operation and were given travel expenses and accommodation at the hospital. Despite this assistance, every visit will cost her father a minimum of 3 days loss of work, so they remain uncertain as to whether they will return for further surgery after her lip is corrected.

**In summary**, a child with CLP/CP born into a wealthy urban family is most likely to flourish, have relatively normal speech and aesthetics, a good education and normal employment and marriage prospects. A comparable child born into an underprivileged rural family is less likely to receive intervention, therefore face a lifetime with a facial defect, compromised speech, nasal regurgitation, stigmatisation, marginalisation, low education, employment and marriage expectations and consequent poverty. The inequality in treatment outcome could
not be more striking and demonstrates that the disparity in health care is not entirely economic.

Creating equity in health care in India is clearly a complex task, since much of the inequality has its foundation in national policy, physical geography and cultural beliefs. However, with the help of state and community intervention and the assistance of global partners, great inroads are being made for change, particularly in relation to health education and surgical provision. Significant investment is being directed towards adult education concentrating on literacy and health awareness. Targeted grass roots education is also being introduced aimed at providing girls with a better understanding of how good nutrition and health during pregnancy can reduce perinatal morbidity (Gokhale et al., 2002). As a means of implementation the Indian Ministry of Health have commissioned 'Accredited Social Health Activists' (ASHA) to identify areas of need and offer health education at a community level thus empowering women in relation to maternal health (Gopalan et al., 2012). 'Mothers clubs' have also been introduced in parts of rural Assam to improve accessibility at a local level (Medhi et al., 2006).

In terms of medical facilities and expertise, despite almost 50,000 doctors graduating in India each year, demand still far outstrips supply, particularly in rural areas. Poor salaries, inadequate facilities, low prestige and underemployment diminish recruitment opportunities of skilled staff into rural and isolated locations and a desire to take opportunities abroad accentuates the problem (Adkoli, 2006, Purohit and Martineau, 2016, WHO, 2010). Consequently many rural services remain poorly covered with low quality of care, despite the introduction of telemedicine, improved salaries and international recruitment guidelines which is intended to improve the profile of many of these previously unmanned facilities (Taylor and Dhillon, 2011).

2.7. Management of CLP in India

The need for intervention for CLP has been recognised as a global problem by governments and funding agencies and cleft care has improved significantly over the last three decades. Initially the problem was tackled by local surgeons and Non-Government Organisations (NGOs) offering free “cleft camps” and “parachute
missions” where a surgical team set up temporary surgeries and operate intensively for a few weeks before leaving (Reddy et al., 2009). Although this system is still widely implemented, the process is significantly flawed since the services are short-lived, often unable to accommodate optimal timing for surgery and lack management strategies for complications and follow up care. In addition, provision of health care in such an informal manner is simply not sustainable nor cost effective and the lack of adjunct care such as speech therapy, physiotherapy and long term support can significantly compromise outcomes (Singh, 2009, Uetani et al., 2006, Shrime et al., 2015, Güneren, 2016)

To address these issues the Indian government, in conjunction with major NGOs such as Smile Train* and Operation Smile*, has introduced a scheme to provide funding for surgical treatment of CLP, assistance for travel to specialist centres and promote education programmes to improve attendance (Patil et al., 2011). In addition the 'Indian Collaboration on Craniofacial Anomalies' (INDIACRAN) has been developed to ensure quality of care, promote an MDT approach and provide a nationwide database for research and audit purposes (Mossey and Little, 2009a). A move away from satellite surgical missions towards supported comprehensive care in local hospitals has encouraged a more multidisciplinary approach, enhanced outcomes and a sustainable, cost effective service (Purnell et al., 2015). This has meant that thousands more people have access to treatment throughout India.

India faces huge challenges in providing comprehensive healthcare for its widely varied population of more than a billion people. For a sustainable service to be developed the government must make appropriate changes and find the political willpower to convince its population to embrace its rural and disadvantaged communities and services equitable nationwide. This will require comprehensive long term planning, strict co-ordination and accountability to pave the way for equitable health for all.

* Smile Train and Operation Smile are two global charities which play a key role in providing cleft care and surgical training in resource limited regions
CHAPTER 3: LATE INTERVENTION FOR CLP

This chapter will examine the literature pertaining to late intervention for CLP and discuss the limitations and consequences of these in relation to service provision for older patients presenting with untreated CLP.

It is well documented that timely closure of the lip and palate at a young age is associated with superior aesthetics and speech outcome (CSAG, 1998, Diego, 2002, Berkowitz, 2006). However, available evidence relating to intervention for CLP in adults and adolescents is less well established and results are often reported as disappointing, particularly in relation to speech. Unless evidence is available which indicates intervention for this cohort is beneficial and cost effective, it is difficult to justify offering treatment. (Sommerlad, 2014, Rohrich and Gosman, 2004). The following section outlines the history of assessing treatment for older patients and aims to identify why limitations in the studies might put these negative conclusions into question.

The first comprehensive cleft repair was reported by Von Graefe in 1816 (Gräfe, 1820) but a more descriptive account of congenital cleft care of the time is given by Snell (1828). In his book he details the issues of infant mortality associated with the inability to suckle and describes both surgical and prosthetic intervention for CLP in older patients as well as the emotional stress associated with inadequate speech.

It was not until the 1930s that anything was published regarding comparative outcomes of surgery and even in these early years non-standardisation of assessment tools and the lack of reliable data presented a problem: Ritchie (1937) pronounced;

"An extensive literature has accumulated since these early operations for cleft palate and but a small amount of it pertains to the functional results after treatment. Reports on the phonetic outcome in a group of cases may be satisfactory individually, but great difficulty is encountered in comparing the results in one group with those in another"
Ritchie goes on to describe his own subjective speech results, based on conversational speech, as 'excellent', 'good', 'fair' and 'bad', a system mirrored by Bentley and Watkins (1947) and Oldfield (1947). It is clear from their publications that their treatment cohort included a wide age range, including adolescents and adults, however, whilst speech outcomes are reported as excellent (42-52%) and good (29-43%), it is not possible to identify to which age ranges these results apply. Holdsworth (1954) was similarly vague in ascribing age ranges to his results but declared that faultless speech was not attainable if treatment was delayed beyond 3 years. Oldfield (1947) was one of the first authors to describe cleft speech characteristics and potential therapy and suggested that 'psychological causes' (shyness; ridicule at school, leading to an inferiority complex), 'deafness' and 'irregularity of the upper teeth' may prejudice intelligible speech.

The introduction of the term 'multidisciplinary team' in relation to CLP was first coined by Baker (1955), and embraced by Law and Fulton (1959) in Puerto Rico, in their quest to understand CLP more fully. Their goal was to identify the natural outcome of CLP if it was left untreated, and their research questions of 58 years ago show a remarkable parallel to those we are still asking today:

- What is the normal outcome of an oral cleft if allowed to mature without treatment?
- How does an adult with an untreated anomaly differ from his peers in physical function, social adjustment, and emotional health?
- How does such an adult compare with one who had received conventional treatment?

Law and Fulton (1959) engaged a team of 5 dentists, an SLT and an orthodontist to interview 124 Puerto Ricans, between 15 and 57 years of age: 59 with untreated CLP, 29 previously treated CLP and 36 control patients without CLP. The results included reports of emotional stress and restricted social inclusion among all CLP participants, individuals with untreated posterior cleft palate had 'almost normal voice quality' in comparison with those who had received surgical closure who demonstrated poor consonant production (Law and Fulton, 1959). A similar project by Landis and Cuc (1972), assessing consonant production and
intelligibility for 54 Vietnamese participants with untreated CLP, between 3 and 24 years old, was more promising: Scoring intelligibility on a 10 point scale (Normal 1-3, Passable 4-6, Poor 7-10), the untreated cohort scored between 6 and 10 whereas two thirds of the treated cohort scored 4-6 for intelligibility and only a third rated as poor, with the greatest success in patients with ICP. An important point which also emerged was the difficulty in assessing speech in a foreign language and the need to train native speaking therapists (Landis, 1973). Unfortunately they comment that they had 'no opportunity to see an ample sample' post-operatively, and since there are no defined parameters in their framework assessment, their results are not easily comparable with other studies.

Ortiz-Monasterio et al., (1966, 1974) reported consistently poor speech outcomes when assessing surgical repair of CP in Mexico using a broad four point scale (excellent, good, fair, poor). In particular they state that, whilst phoniatic training can be useful up to 12 years old, there is little advantage in applying it in older patients who have an established speech pattern. Unfortunately, they do not detail their methods, assessment parameters or therapy input, nor do they differentiate between age, development of fistulae or cleft type in relation to outcome, so the value of this study is limited (Sell and Grunwell, 1990).

Adekeye and Lavery (1985) assessed the outcomes of 160 Nigerian adults with CLP and although they refer to speech difficulties they do not describe post-operative speech assessment and only refer to better quality of life post surgery due to improved aesthetics. Finally, in describing his 33 year follow up of a 15 year old boy treated for his unrepaired BCLP in the UK in 1955, Rees (1991) describes the post-operative improvement in speech and psychological improvement as 'nothing short of extraordinary'. The boy apparently became instantly gregarious and intelligible, where before he had been a total recluse and impossible to understand, and 33 years later was leading a fulfilling and active life.

Whitehill et al. (1996) also report a single case study; an 18 year old Cantonese lady who's CLP had been treated by 13 years of age, then treated with an additional fistula repair and osteotomy. They reported excellent improvement in articulatory placement with electropalatography despite having velopharyngeal insufficiency.
Whilst interesting, these studies can only be considered anecdotal as they are have inadequate methodologies, are short term studies and lack necessary specifics. In order to prove categorically whether late intervention was a success or failure there was an obvious need for a more systematic approach to evaluation of outcomes following late intervention (Sell et al., 1994, Morris and Bardach, 1989). The following section describes seven of these in detail.

The first comprehensive longitudinal study in this field was undertaken by Sell and Grunwell in 1990, using detailed speech analysis to assess 18 adolescents, undergoing surgery and speech therapy in Sri Lanka, over a 12 month period. They reported severely disordered speech pre-operatively, only minimal spontaneous improvement following surgery alone and concluded that late repair for this cohort was of little value unless supported by intensive and on-going speech and language therapy. One of the problems they encountered was that traditional phonetic analysis did not work well with disordered cleft speech which prompted the need to device a unique method for analysing cleft speech.

Sell (1991) undertook a longitudinal study involving 88 patients undergoing palatal repair in Sri Lanka in a cohort comprising 67 individuals over 8 years old and 21 patients who were treated at a younger age. Speech was compared for 51 individuals from recordings made pre-operatively, early post operatively and at long term follow up 4-5 years post surgery recordings. She reports that near normal speech was only possible for younger individuals with less severe clefts and that palatal repair alone did not result in significant change in articulation in adults beyond 11 years. 76% of her cohort did not show improvement in speech even with speech therapy, only 20% of procedures resulted in velopharyngeal closure and fistulae were more likely to occur in adult repair than in adolescents. She also noted that, where the cleft was treated by palatoplasty alone, the best results with a single procedure occurred for posterior repairs and was not considered sufficient for others cleft types. She concluded that age alone is not a good predictor of outcome and it is important to consider many other aspects including cleft type.

Murthy et al., (2010) compared pre-operative and post-operative speech samples for 131 adolescents age 10 to 35 (mean 15.8) in Chennai, India. Despite the
persistence of some residual speech problems they reported a moderate improvement in articulation, hypernasality, nasal air-emission and intelligibility in most patients even without speech therapy. They concluded that whilst late repair did not result in normal speech it improved day to day communication and, anecdotally, they reported a reduction in regurgitation. These results are contrary to those of Sell (1991) however, the exclusion criteria mean that the two studies are not really comparable. Notably Murthy et al., excluded individuals with isolated secondary palate clefts owing to the reported variability in speech outcomes (Law and Fulton, 1959, Randall et al., 2000) and those with a residual palatal fistula. Sell (1991) reports better speech outcomes with late posterior repairs, so exclusion of this cohort might be expected to lead to a less positive outcome. Conversely, the presence of fistulae has a well-reported negative impact on speech (Persson et al., 2006, Morris, 1973) which might, at least in part, account for the more positive conclusions of the Murthy's et al. study, where this cohort were excluded.

Hayden et al., (2000) also claimed improvement post surgery, when they compared speech samples pre-operatively and one day post operatively in 10 Vietnamese patients aged 7 to 50yrs. Sixty percent of their late repairs demonstrated better syllable production however, the small sample, the wide age difference and the close proximity of the surgery to the assessment make the study somewhat inconclusive. A study by Schwarz (Schwarz, 2006) using a single stage procedure for lip and palate repair is similarly inconclusive since, although they claimed to assess speech and intelligibility in relation to late CLP repair, only four of the nine strong cohort had an assessment and it cannot be established from the paper whether assessments were pre-operative and/or post-operative.

Bruneel et al., (2017) discussed the speech outcome of 15 individuals in Uganda who had received intervention for CLP beyond 8 years. They concluded that speech deviation persisted post operatively but since there was no pre-operative data it was not possible to ascertain whether or not there had been change in speech so this study was also inconclusive.

Schonmeyr et al., (2015b) assessed the speech outcome of 31 patients in Assam who had received intervention beyond 7 years of age. Assessments were undertaken by 6 non-native SLTs and 4 local laymen using recorded speech
samples, categorising their sample according to the Veau Cleft classification (I- defects of soft palate only, II -hard and soft palate, not extending past incisive foramen, III - complete UCLP, IV- BCLP). Only 6 out of the 31 patients showed a discernible difference pre and post-operatively of which two thirds were Veau I and II and none Veau IV. In agreement with Sell (1991), they concluded that clefts of the posterior palate have a higher chance of a good speech outcome but there was "no convincing evidence that patients with complete clefts would benefit from repair with regards speech" and suggest that routine treatment is not recommended. There are limitations in this study however in that there was a wide variation in the surgery; 7 different techniques by 14 different surgeons; there were only 2 Veau IV patients included in the study, which makes the assessment of BCLP inconclusive. Also there was no acknowledgement that the development of fistulae in 50% of the Veau I & II cases, may been the reason behind the poor speech outcome (Sell, 1991). Murthy et al., (2010) excluded participants with fistulae from her study for this reason, which makes the studies difficult to compare.

Unfortunately Schonmeyr et al., (2015b) only considered speech to have improved if there was total agreement from all 10 assessors and did not differentiate between assessments conducted by the lay and the SLT assessors in this study. This is potentially important, since SLT analysis has been reported as having a tendency to be over critical, particularly in relation to VPI, (Havstam et al., 2011, Sell, 2005, Brunnegård et al., 2009) whereas an untrained listener adds a 'real life' significance to speech assessments (Sell, 2005). Since the use of lay listeners is intended to assess the acceptability of speech, in contrast to professional speech analysis, failure to differentiate the two compromises the benefit of their inclusion (Brunnegård et al., 2009, Whitehill, 2002, Havstam et al., 2011).

A further study by Schönmeyr et al., (2015a) surveyed patients who had received intervention beyond 7 years old, using QoL questions based on the Speech Handicap Index plus four questions on nasal regurgitation. 134 participants were interviewed pre-operatively and 98% of the participants reported that they were hoping for better speech and 35% felt their CLP had limited their opportunity to find work. Only 46 participants were identified for follow up, reflecting the
difficulty in assessing this cohort longitudinally. Although there was no significant increase in the speech index score, 71% of the participants perceived that their speech had improved, 82% felt their lives had improved post-operatively and all of the participants felt that the surgery had lifted their self esteem, commenting that they felt less left out of conversations and less handicapped by speech. Interestingly regurgitation was reported as increasing by 23% post-operatively, however it was not possible to ascertain from the paper whether or not this related to development of fistulae. Despite the perceived psychosocial improvements reported by the participants in this survey, Schönmeyr et al. concluded that, based on the lack of change in the speech index scores and the increased nasal regurgitation, their results do not support the assumption that patients will benefit from late palate repair. They suggest that the anecdotal reported improvement reflects a positive response bias and a placebo effect from surgery and recommend that late palate repair should not be considered in areas where resources are limited.

A similar level of patient satisfaction was reported by Sharp et al., (2008) following a patient-reported post-operative outcome survey of 54 individuals (average age 10.5) who had received intervention for CLP in the Philippines: 99% of their patients said they would encourage others to have surgery, 55% reported improved speech, there was a 30% reported reduction in regurgitation and many participants indicated that they were suffering less bullying and greater inclusion. These findings are supported by Dvivedi and Dvivedi (2012) who describe a 93% post-operative satisfaction with aesthetics and function immediately post-operatively in their survey of 4567 participants over 5 years old in Nepal, however there is scant methodology reported in their paper.

It has been argued that an individual’s own satisfaction with treatment is possibly the most important factor in evaluating their care (Havstam, 2010, Semb et al., 2005). Yet Schönmeyr et al., (2015a) pay little heed to the reported satisfaction of their participants, despite mirroring those of sharp et al., (2008) and Dvivedi and Dvivedi (2012), and prefer to make their judgement based on the speech index, concluding that delayed surgery is of no benefit and ill-advised. There is a great deal of research relating to CLP which indicates that personal and professional
assessment of satisfaction do not always correlate (Mani et al., 2010, Semb et al., 2005, Sinko et al., 2005, Turner et al., 1997, Bickham et al., 2017) and that the professional assessments might not always prove the most accurate (Havstam, 2010). Whilst there may well be an element of responder bias in the questionnaire as they reported, it is also possible that the differential lies in the validity of the assessment instrument which may not be accurately representing a holistic outcome.

There are other confounding factors which may be influencing the discrepancies in these studies including the level of experience of the operating surgeon which can compromise surgical outcome (Bearn et al., 2001). This is borne out in several studies; Schönmeyr et al., (2016) report a generally higher rate of complication, including bleeding and fistula formation, following surgery in adults but these occurred in 50% of the procedures undertaken by visiting surgeons in comparison with only 22.6% of resident surgeons procedures. Similarly Rossell-Perry et al., (2015) reported that fistula formation increased from 3.8% to 25% when the same surgeon worked in an unfamiliar theatre and Maine et al., (2012) confirmed a tenfold increase in fistulae when working on a mission in Ecuador rather than their regular US clinic. These differences have been attributed to lack of experience, operating in unfamiliar surroundings, fatigue associated with high surgical volume during satellite missions and limitations in post-operative care (Rossell-Perry et al., 2015, McQueen, 2015). Appreciating that the complication risk is known to be greater for older patients, better outcomes might be expected where this cohort is treated by an experienced local rather than a visiting surgeon.

A common factor which puts the conclusion of many of these papers into question is the heavy dependence of perceptual speech analysis as a measure of success with the expectation of normal speech in response to a single surgical procedure: Even when CLP surgery is optimally timed, VPI and fistulae are common complications causing hypernasality, nasal emission and turbulence. Even under optimal conditions, recent studies report the incidence of surgical breakdown and fistulae formation as between 5 and 27% of cases (Murthy, 2011, de Agostino Biella Passos et al., 2014, Hardwicke et al., 2014, Bykowski et al., 2015). The incidence may depend on the type of repair, the size of cleft and timing of surgery.
(Parwaz et al., 2009, Landheer et al., 2010, Losken et al., 2011) and experience of the surgeon (Andersson et al., 2008). Fistulae often require secondary surgery with patients being supported by regular SLT intervention to minimise the impact on speech and communication (Kummer et al., 2012, Britton et al., 2014, Harding and Grunwell, 1995, de Agostino Biella Passos et al., 2014). Even in absence of fistulae and VPI, distinctive compensatory speech characteristics regularly endure into adulthood (Britton et al., 2014, Peterson-Falzone et al., 2001) and are notoriously difficult to remediate (McWilliams et al., 1990, Bzoch, 1989). For individuals who have received late repair for CLP the problem is potentially accentuated owing to surgical scaring, established cleft speech characteristics and higher than average risks of developing fistulae and velopharyngeal incompetence (Whitehill et al., 1996).

In the UK the projected speech outcome standard for 5 year olds who have received full comprehensive cleft care is that 60-70% should have normal speech, 70% should have no structural problems without secondary repair and 50% should not demonstrate articulation difficulties (Kuehn and Moller, 2000, Kummer et al., 2012). However the literature reports that all three standards are only achieved at 50% meaning that secondary intervention will be advised for half of all individuals with CLP (Britton et al., 2014). Reported VPI rate for CLP repair is between 25 and 43% (Morris, 1973, Enderby and Emerson, 1996, Peterson-Falzone et al., 2001) but the question of whether cleft type is a predictor of outcome remains inconclusive: Pulkkinen et al., (2001), Timmons et al., (2001) and Brunnegard & Lohmander (2007) did not demonstrate a significant difference in cleft type and VPI related speech outcomes whereas Hardin-Jones et al., (1993), Persson et al.,(2002) and Nyberg et al., (2010) demonstrated the best speech outcomes in with Veau I clefts and more speech errors with larger clefts.

Britton et al., suggest that we are setting the standard bar too high and we should expect between 50-60% of cases to require additional intervention after their primary surgery (Britton et al., 2014). If this is the situation following optimally timed surgery in the UK, it is unsurprising that perceptual speech outcomes following late repair, as a single procedure, for adults in the developing world does not demonstrate particularly exceptional results, particularly where there is
negligible SLT support or scope for secondary surgery. Also, these studies are comprehensive and will include results from every case treated, good and bad, giving a balanced assessment of outcome. In contrast, studies relating to late intervention are only likely to comprise a cohort who are experiencing difficulties, as individuals who have acceptable speech are unlikely to seek intervention. Consequently, the cohort under investigation are potentially less likely to excel in speech post surgery than might be expected from a wider sample.

Long term psychological consequences of CLP are also well documented amongst individuals who have had optimally timed intervention for CLP (Richman and Eliason, 1982, McCormack et al., 2009, Chapman, 2011). Whilst the patients personal psychological well-being and patient perception of change is reported anecdotally throughout the literature discussed earlier, the majority of papers focus heavily on the more tangible outcomes such as surgical complications and speech analysis which demonstrate little change following intervention (Murthy et al., 2010). Sell (2005) acknowledges that reliance on speech analysis overlooks the psychosocial impact of improved communication for the individual and this may well be a factor which accounts for the dichotomy between perceived and measurable outcomes in this cohort.

**In summary**, it is clear from this review of the literature that we are far from reaching a conclusion regarding the efficacy and outcome of late intervention for CLP. Much of the evidence is conflicting and difficult to compare owing to the different parameters being measured, different perspectives of success and failure, different inclusion criteria, different procedural input and an underlying complexity of agreeing, defining and measuring assessment and outcome. In the main, research hereto undertaken in this field conclude that late intervention for CLP is ill-advised as it does not provide a positive outcome and is therefore not a cost-effective procedure where resources are limited (Schönmeyr et al., 2015b). These conclusions are potentially inhibiting the provision of funding for treatment of adults with CP yet there are limitations in their research strategy and choice of outcome parameters which put their conclusions into question.

This raises two issues: Firstly, is there an outcome measure which can reflect changes in social functioning and psychological well-being following late palate
repair, rather than focussing the impairment in isolation? Secondly, can the
outcome measure be administered successfully, without the need for highly
trained SLTs, in diverse cultural environments?

The pursuit of answers to these questions forms the basis for this thesis but first
requires an understanding of the instruments which are currently available for
measuring quality of health care and service outcome in this field.
CHAPTER 4: ASSESSING QUALITY OF HEALTHCARE

This chapter outlines the role of outcome measure in relation to ensuring quality and equity in health care provision. It will start with an introduction to the concept of evidence based medicine followed by a generic overview of assessment instruments for measuring quality in health care before discussing outcome measures applicable to CLP with particular focus on this research.

4.1. History of ensuring quality and equity in health care

Managing health care requires sustainable and cost effective strategies, which can be delivered within the boundaries of available resources (Lopez et al., 2006). It is therefore imperative to establish how to achieve the desired outcome at the best value (Salinas et al., 2016, Porter, 2010). However, whilst the intention is clear, agreement on which parameters and performance indicators are most appropriate to measure is not (Mainz, 2003, Arah et al., 2003, Donabedian, 1966, Tarlov et al., 1989). This has led to the development of an overabundance of clinical trials, case studies and performance indicators, each with a slightly different focus (Saver et al., 2015). Bastian, Glasziou et al. (2010) estimated that an average of 75 clinical trials and 11 systematic reviews were being produced on a daily basis, which is far more than is possible to assimilate and process let alone implement.

The concept of measuring quality in health care is actually as old as medicine itself: Hippocrates, 'The father of modern medicine', documented his recommendation of auditing and adjusting treatment strategies declaring to his students “rely on actual evidence rather than on conclusions resulting solely from reasoning, because arguments in the form of idle words are erroneous and can be easily refuted” (Mountokalakis, 2009). His inspiration, from almost two and half thousand years ago, represented not only the conception of modern medicine but also the commencement of the outcome movement and evidence based medicine (EBM) (Di Blasi et al., 2001, Revicki et al., 2008).

Although it was Gordon Guyatt, in 1990, who coined the term EBM, to illustrate the concept of employing good scientific reasoning when selecting medical interventions, the notion was evident decades earlier (Kowalski and Chung, 2013):
In the 1850's Florence Nightingale's endeavours as a statistician and auditor, led to the execution of health care reforms in the Crimean war (Aravind and Chung, 2010), in her words “The ultimate goal is to manage quality, but you cannot manage it until you have a way to measure it, and you cannot measure it until you can monitor it” (Eagle and Davies, 1993).

It was not until the 1950's and 1960's, due to the escalating costs of medical services and increased patient need, that monitoring quality became more mainstream. Two key factors fuelled this change: Patient demand for equity of care at lower prices and fund holders insistence in value for money to lessen the fiscal burden of service provision (Kowalski and Chung, 2013, Shaw and Miller, 2000). It is the continuing endeavour to reconcile these two dimensions which drives much EBM today and establishes how resources are directed.

Historically there was an overwhelming tendency to focus on objective measurements to assess quality in health care, such as the number of procedures undertaken, length of hospital stay, mortality rates or the results of biochemical tests and physiological changes, figures which are easy to capture (Porter, 2010). Although valuable, information gleaned purely from clinical assessment gives only a narrow perspective of health care, may not reflect change and rarely include the patient perspective of events (Glasziou et al., 2012, Saver et al., 2015). Since the aim of most healthcare is to minimise disability, reduce symptoms and maintain or improve quality of life (Black, 2013, Homer et al., 1998), the assessment of their impact can only be truly assessed by including the patients viewpoint (Rathert et al., 2011). Recognition of this led to a change in position and a move towards a people-centred and integrated approach to health services, reflected in the WHO definition of quality care “care that is safe, effective, people-centred, timely, efficient, equitable and integrated.” (World Health Organisation, 2015, WHO, 2015b, Villeneuve and Mildon, 2013). This has brought with it the inclusion of many patient reported assessment tools with a goal of providing a more holistic view of healthcare and increasing public accountability (Black, 2013).
4.2. Outcome measures

Outcome measures are instruments which monitor the impact of an intervention by means of comparing a situation at two or more points of time (Collis and Bloch, 2012). This allows stakeholders and clinicians to compare the efficacy of treatment modalities for individuals and between services, a process known as benchmarking (Pantall, 2001, Enderby and John, 2015). By centering the process on the client there is an additional advantage of engaging patients in their care package, which has proven to improve outcome (Coulter and Ellins, 2007, Stewart, 1995, Black, 2013).

The Scientific Advisory Committee Of Medical Outcomes suggested a number of attributes which should be sought in an outcome measure (Perrin, 1995):

- Defined concept; the concept to be measured should be clearly defined
- Reliability; the instrument should be free of random error
- Validity; it should measure the concept for which it is designed
- Responsiveness; it should detect change over time
- Interpretability; it should produce results which are easy to interpret
- Burden; it should not put undue demands on the administrator.
- Adaptable; it should be adaptable to different modes of data collection/different environments.
- Culture and language; it should be amenable to translation and culturally sensitive.

Essentially, an outcome measure must be appropriately designed for the task in hand, make an accurate representation of the clinical scenario and be practical for clinical setting (Dobrzykowski, 1997; Smith et al., 1997; Skeat & Perry, 2008). Critically, it must also be easily understood, straightforward and quick to apply so it will be accepted and adopted into regular practice (Hesketh & Sage, 1999; Lee & McCormick, 2002; Simmons-Mackie et al., 2005). In general it should not require high levels of expertise to administer so as not to restrict its use to a specialist environment, this is of particular importance in the developing world where resources are limited.
Where an outcome measure is to be used in a variety of different environments it must be flexible enough to account for the diverse array of scenarios which might be encountered (Westrin, 1999; John & Enderby, 2000a, 2000b; McHorney & Bricker, 2002). It must also be flexible enough to account for the dynamic nature of health as it varies with time. This is true not only of the effect of any intervention but also in relation to the natural progression of a disorder and fluctuations in personal perception which may alter with time (Mainz, 2003).

Incorporation of all these criteria is unlikely in any single system, which often makes it difficult to find a robust outcome measure for accurately quantifying and assessing health care holistically (Gilbody et al., 2002; Lee & McCormick, 2002). The problem of outcome measure selection is prevalent across all branches of healthcare as will be demonstrated in relation to CLP in the following section.

4.3. Outcome measures and CLP

As outlined in the previous chapter, CLP encompasses a complex interconnecting web of sequelae which are managed by multiple disciplines over an extensive time span. This has led to multifarious outcome measures being developed to assess its impact on speech, communication, eating, drinking, hearing, breathing, psychology, well-being, inclusion, marginalisation, affect on family members, aesthetics, education, finance and global burden to name a few (Allori et al., 2017, Auslander et al., 1993, Lohmander and Olsson, 2004, Endriga and Kapp-Simon, 1999, Kuehn and Moller, 2000, Henningsson et al., 2008, Shaw et al., 2005): A Medline search for "cleft lip and palate" and "outcomes" yields over 2400 results and a similar search in Google scholar gets 23,400 hits. The problem is further compounded by the fact that different outcome measures have been developed for different purposes: They can be generic or explicit; pertinent to different age groups and ethnicities; use qualitative, quantitative or mixed methodologies; use measurable parameters, interviews, patient reported measures (PROMS) and questionnaires; and/or measure both single events and changes over time.

Whilst the plethora of research in this field shows an encouraging desire for practitioners to critically evaluate their services, it is problematic when comparing different interventions and benchmarking services (Sell, 2005, Shaw et al., 1992,
Lohmander and Olsson, 2004, Sitzman et al., 2016). Although there is a consensus that standardization could be beneficial (World Health Organization, 2002a), the debate on what to assess, which instruments to measure it with and from whose perspective to measure is an on-going debate (Allori et al., 2017).

It is beyond the scope of this thesis to discuss the plethora of outcome measures available to assess CLP so the following will focus on the rationale behind the final selection of the adapted TOM and the WHOQoL-BREF for the purpose of this study.

4.4. Choosing an outcome measure to assess psychosocial change

The concept of this research project coincided with a review by Eckstein et al., (2011) of the available patient centred outcome measures which focussed on both functional and psychosocial aspects of CLP. They reported on five validated measures matching their criteria, of these, four were not specific to CLP and related explicitly to children: The Child Oral Health Impact profile (Broder et al., 2007), The Youth Quality of Life – Facial Difference questionnaire (Patrick et al., 2007), the Child Oral Health quality of Life survey (Gherunpong et al., 2004), Paediatric Voice-related Quality of Life Survey (Boseley et al., 2006). The fifth, the Cleft Audit Protocol for Speech-Augmented (CAPS-A) (Sell et al., 2009) was identified incorrectly, since it is actually a specialist framework for analysing speech production in CLP not, as described, a tool for addressing quality of life (Sell et al., 2012).

These findings were mirrored by Klassen et al (2012) in their search for a Patient reported outcome measure to assess QoL in children with CLP, and clarified the need for the development of such an instrument. Interestingly, a review of the current literature shows little has changed since 2011 and there remains a lack of consensus or availability for a holistic outcome measure designed specifically for CLP (Ranganathan et al., 2015, Queiroz Herkrath et al., 2015, Stock et al., 2016).

In addition to requiring an outcome measure which reflected the impact of CLP on social functioning, this project required it to be appropriate for use in the developing world. It needed to be culturally sensitive, not language specific, globally acceptable and possible to administer in areas of low resource and limited
specialist expertise. The lack of an available outcome measure led the Primary investigator (PI) to consider the TOM (Enderby & John, 1997) which was being used widely as a holistic tool in other fields such as speech and language therapy, physiotherapy and audiology and showed promise for use in this project. A provisional research project, focussing on adults with untreated CLP in India resulted in the development of and adapted version of the TOM, the TOM-clp and a pilot study indicated its potential as an instrument clinical use in this project (Rees et al., 2016b).

With the aim of using multiple methods to collect data for this research a decision was made to utilise three techniques: Semi-structured interviews, which will be discussed in more detail in the following chapter and two assessment instruments; the TOM-clp and the WHOQOL-bref. The TOM-clp was included as CLP specific outcome measure and the WHOQOL-bref questionnaire, not as an outcome measure but as a generic quality of life measure for purpose of comparison. These will be discussed in detail in the following section along with an additional outcome measure, the cleft-Q which was not available at the start of this project and is just entering its pilot stage in 2017. The Cleft-Q professes to be a multi-cultural instrument with a focus on quality of life for CLP (Riff et al., 2017) and is significant as it has recently been adopted by the International Consortium For Health Outcomes Measurement (ICHOM) as a key part of their standard set of outcome measures for CLP, although it is not currently available for general use (Kelley, 2015).

4.5. The TOM and the TOM-clp

The TOM was developed to engage with the psychosocial features of health care which are difficult to quantify reliably and consistently (Enderby and John, 2015). The importance of these aspects had been highlighted in the World Health Organisation (WHO) document, the International Classification of Impairments, Disabilities and Handicaps (ICDH) in 1980 (WHO, 1980), however, the notion of encapsulating these elements into a single clinical and social outcome tool had not previously been attempted.
The construct of the ICDH classified the health components of disability into three core domains: ‘Impairment’, ‘Disability’ and ‘Handicap’ which acted as a framework to reflect the impact of morbidity on peoples’ everyday lives. It was intended to support a 'social model’ of care and encourage inclusion and support in a range of public sectors including health, education, policy making and the financial sector. This was in contrast to the impairment focussed 'medical model' which was popular at that time (Stucki et al., 2002).

Although the TOM has its foundation in the ICDH, it pertains to the more 'neutral' terminology of the updated WHO document the International Classification of Functioning, Disability and Health (ICF) (WHO, 2001) and includes a fourth domain to reflect the personal and emotional aspects of health. The four domains; Impairment, Activity, Participation and Well-being are rated independently on an eleven point ordinal scale, each representing a different element of health:

- **The impairment domain** reflects the anatomical and physiological components of health and how it differs from the norm. This might include pain, visual and auditory acuity, sensory and motor function or physical differences. In the case of late intervention for CLP this includes the type and the size of the cleft lip and/or palate.

- **The activity domain** concerns the manner in which the individual functions and how this is limited by their ability/disability. This might include the degree to which pain affects activity or the mental and physical ability to perform tasks. In the adapted version for CLP activity is assessed in relation to the ability to speak and the presence or absence of regurgitation.

- **The participation domain** is more contextual and reflects the psychosocial aspect of the condition and level of autonomy It might include the levels of social support, attitudes of others, cultural beliefs which are beyond the control of the individual as well as decision making and willingness to participate daily activities. In relation to CLP this domain is assessed in relation to a participants level of confidence in social situations, their readiness to socialise and eat in public and
their willingness to undertake daily activities such as shopping, using a phone, going to school/work.

- **The well-being domain** is concerned with emotional status and how personal factors reflect the comfort and security of the individual irrespective of their condition (Threats and Worrall, 2004). For CLP this might focus on episodes of anxiety and depression as well as embarrassment and frustration associated with the condition.

This approach takes a holistic view of health and acknowledges that impairment and disability were not necessarily linked and should be considered contextually rather than in isolation (Power et al., 2011, Enderby and John, 2015). Although each of the four domains is scored independently, when considered together, they encapsulate an overall representation of the situation from a clinical, functional, social and personal perspective.

Figure 2 demonstrates how each of the domains of the ICF and TOM can be used to reflect CLP. A copy of the TOM scale for CLP, the TOM-clp can be viewed in appendix (Appendix 1) and Therapy outcome measures for rehabilitation professionals p122 (Enderby and John, 2015)
Mossey and Little (2009b) advocate that the goal of intervention for CLP is to ensure an individual’s integration and inclusion without the prejudice or discrimination which occurs when CLP is left untreated. It is probable therefore that psychosocial aspects of treatment will be of critical interest to the individual, so it is imperative that these factors form a core part of our thinking when considering treatment rationale and measuring outcome. The TOM approach is particularly well designed for this purpose since it supports the assessment of how communication relates to social functioning rather than focusing on the impairment, speech patterns and articulation alone as described in the previous chapter (McLeod, 2004, McLeod, 2006, Hickson et al., 2005).
4.6. Using the TOM

The TOM procedure involves the assessor obtaining enough pertinent information from the participant to score each of the domains. In comparison with an interview or questionnaire, this is an informal process and the information is gathered during regular clinical procedures such as examination, history taking, standardized tests, observation and general conversation with the participant and/or friends and family members accompanying them at the appointment (Enderby and John, 2015). The assessor processes this information and judges the appropriate rating to assign to each of the domains based on the TOM scale appropriate to the condition.

As the process is undertaken in accordance with regular clinical activity it requires little additional clinic time to administer and since the scores can be documented in the patient notes it involves no additional resources. Once the assessor has been calibrated the process can be undertaken using the particular language, means of communication and level of understanding most appropriate to the age and ethnicity of the individual. Since the process is part of a normal routine appointment the process is less likely to be associated with the response bias which is sometimes associated with a more formal questionnaire or interview (Broder et al., 2007, Bowling, 2005, Furnham, 1986).

Each domain is assessed individually using an ordinal scale where zero is profound and five is considered normal for an individual of that age, culture and gender. In order to increase sensitivity the scale is divided to include half points resulting in the eleven point scale (Figure 3).

**Figure 3: TOM ordinal scale (from Enderby and John, 2015 p23)**

<table>
<thead>
<tr>
<th>Profound</th>
<th>Severe</th>
<th>Severe/mild</th>
<th>Moderate</th>
<th>Mild</th>
<th>Normal</th>
</tr>
</thead>
<tbody>
<tr>
<td>0</td>
<td>0.5</td>
<td>1</td>
<td>1.5</td>
<td>2</td>
<td>2.5</td>
</tr>
<tr>
<td></td>
<td></td>
<td>2</td>
<td>2.5</td>
<td>3</td>
<td>3.5</td>
</tr>
<tr>
<td></td>
<td></td>
<td>4</td>
<td>4.5</td>
<td>5</td>
<td></td>
</tr>
</tbody>
</table>

To ensure that the definition of the terms used in the ordinal scale are interpreted consistently between users, a 'core scale' was developed for purposes of cross referencing. In 1992 this concept was expanded further to provide disorder-
specific core scales to facilitate increased reliability amongst different raters. There are now 47 adapted scales spanning a wide range of health specialties and including the TOM-clp (Enderby and John, 2015)p122.

A detailed explanation of the implementation of the TOM-clp in practice will be given in chapter 4.

4.7. Validity and Reliability

4.7.1. Validity

As mentioned in section 0, in order to determine the level of confidence that can be placed in a measure, it is essential to demonstrate that the clinical expectations match the theoretical concept by accurately measuring what is required of it (Hammersley, 1987, Perrin, 1995). Four different methods have been described for assessing validity: Criterion and construct validity, which demand the comparison of the test with an equal or comparable measure which can act as a gold standard, not currently available for the TOM; Face validity and Content validity are based on whether the instrument is truly representative and has been assured for the TOM based on a review by an expert panel following multiple and extensive clinical trials (Roulestone et al, 2004; Enderby et al., 2006).

4.7.2. Reliability

Reliability of the TOM has been extensively tested, with good inter-rater and intra-rater reliability demonstrated after only a short training period (John et al., 2002, John, 1993, Enderby and John, 2015). Reliability of the TOM-clp was established by comparing the inter-rater reliability of non-specialist participants’ assessment of hypothetical case scenarios in a research setting. The resulting Kappa scores and percentage reliability were determined as good to excellent but needs to be confirmed in a clinical setting (Rees et al., 2016b). This will be discussed further in chapter 8.
4.8. Features of the TOM/TOM-clp appropriate to this research

- Already validated to reflect clinical and psychosocial change for use in other fields; communication disorders, occupational therapy, audiology and physiotherapy.
- Is cross-cultural and is not language specific.
- Is simple to administer and shows equal reliability irrespective of the level of experience of the user.
- Incurs no cost to implement.
- Data is numerical thus good for data-bases and benchmarking.
- Data volume is small and direct collection into a secure database is straightforward using a hand held electronic device.
- Is based on the International Classification of Functional Disability and Health (WHO-ICF) so internationally acceptable.

4.9. The WHOQoL-BREF

The WHOQoL-BREF is a shortened version of the WHOQOL-100 questionnaire, which was produced in 1991 as a generic quality of life questionnaire which could be sensitive, cross-cultural, multi-lingual and suitable for use in both developed and the developing world (Hawthorne et al., 2006, Whoqol Group, 1994). Defining Quality of life (QoL) as an "individual's perception of their position in life in the context of the culture and value system in which they live and in relation to their goals, expectations, standards and concerns" (WHOQoL Group, 1993) the process involved the collaboration of 15 international sites and produced a broad ranging assessment tool comprising 100 items organised into six domains: Physical health, psychological status, personal relationships and beliefs, inclusion and impact of their environment (WHOQoL Group, 1993, Harper and Power, 1999, Whoqol Group, 1994).

International field tests of the WHOQoL-100 were completed by 1995 but proved too cumbersome for practical purposes and a shorter version, the WHOQoL-BREF was produced, comprising 26 standard questions distributed in four domains: Physical (7 items), psychological (6 items), social relationships (3 items) and
environmental (8 items) (Skevington et al., 2004, Whoqol, 1998). It was decided that two 'National items' which were pertinent to different cultures would also be included as overall makers of quality of life. For example "do you feel respected by others" and "are you able to get the things you like to eat" were introduced in the Taiwan version (Chen et al., 2006, Yao et al., 2002). Each item is scored on a Likert response scale, from 1-5 and the scores are aggregated, according to the standard procedure outlined in the WHOQoL manual (Harper and Power, 1999, World Health Organization, 1998), with the intention of producing a single global 'quality of life score'; a higher score indicating a better quality of life. Alternatively each of the domain profiles can be represented independently and a number of researchers have found this to give a better indication of life quality (Noerholm et al., 2004, Harper and Power, 1999, O'Carroll et al., 2000).

4.10. Features of the WHOQoL-BREF appropriate to this research

- It is well established and considered one of the best known generic instruments for measuring cross cultural QOL (Bowden and Fox-Rushby, 2003)
- Two field sites in India were included in the founding research during its development (Whoqol, 1998, Skevington et al., 2004, Saxena et al., 1998): It has been translated into more than 40 languages including five widely spoken in India (Bowden and Fox-Rushby, 2003):
- The generic nature of the tool enable it to be used for a wide variety of conditions and it has been validated and used successfully for a wide range of disorders (Skevington and McCrate, 2012, Skevington et al., 2004, Saxena et al., 1998, Singh and Junnarkar, 2014, Li et al., 2009, Fu et al., 2013):
- It accounts for social relationships and environmental issues which can play a significant role in QOL in relation to this study (Chen et al., 2006):
- The psychometric properties of both the Hindi and English versions have been rigorously evaluated and validated in India (Agnihotri et al., 2010, Bowden and Fox-Rushby, 2003).
- It has been used widely as an outcome measure in India for a wide range of ages including both children and adolescents (Agnihotri et al., 2010, Saxena
et al., 1998, Sreedevi et al., 2016, Chandran et al., 2016, Awasthi et al., 2012, Singh and Junnarkar, 2014):

- Questions which are considered inappropriate because of age of the cohort or cultural issues can be adapted, missed or replaced (Fu et al., 2013, Li et al., 2009, Chen et al., 2006, Ghoshal et al., 2012) for example following a validation project amongst adolescent group in Lucknow (Agnihotri et al., 2010) and Taiwan (Chen et al., 2006) a culturally sensitive change to the instrument by replacing the question "are you satisfied with your sex life" with "are you satisfied with the respect you receive from others".

In the absence of a more suitable validated QoL measure for CLP, it was felt that the Global acceptance of the WHOQoL-BREF and its endorsed credentials amongst different age groups in India, made it the most appropriate instrument for triangulation in this project. It was made available in Hindi and English with the intention of using the culturally sensitive version for adolescents as described above.

4.11. The Cleft-Q

Since the cleft-Q it is not currently available for general use only a brief outline can be given: It has been designed specifically for use with children and young adults with CLP, in line with other similar patient reported outcomes measures (PROMS) such as the FACE-Q (Longmire et al., 2017). Its basic construct is a patient reported 4 point scale questionnaire made up of 171 items in 13 scales which has been developed, using a mixed methods approach including literature searches and qualitative thematic analysis from transcribed interviews (Riff et al., 2017). Data is collected both via traditional pen and paper and also being piloted in an electronic format. It claims to be culturally sensitive and has been translated into a number of different languages, including Hindi and Assamese, and is currently being piloted extensively. Interestingly, although the intention is for it to be internationally acceptable in developed and developing countries, of the 12 proposed pilot countries, only India is considered less than upper middle income according to the World bank (2017).
The Cleft-Q is of particular interest in relation to global cleft research since it dominates the recent standards set by the ICHOM, as the only subjective measure of health and accounting for 9 of the 24 standards (Figure 4).

**Figure 4: Summary of standards set for CLP from ICHOM**

(Adapted from Allori, Kelley et al. 2017)

<table>
<thead>
<tr>
<th>Outcome Domain</th>
<th>Included Outcomes</th>
<th>Instruments Used</th>
<th>Data source</th>
</tr>
</thead>
<tbody>
<tr>
<td>Eating and drinking</td>
<td>Body weight Change in weight Eating and drinking</td>
<td>Growth chart Growth chart CLEFT-Q Eating-and-Drinking subscale</td>
<td>Paediatrician/Nurse Paediatrician/Nurse Patient</td>
</tr>
<tr>
<td>Dental and oral health</td>
<td>Dental health Oral health Occlusion Mastication</td>
<td>dmft and DMFT scores COHIP Oral Symptoms subscale† Overjet assessment Lateral Cephalogram tF CLEFT-Q Eating-and-Drinking subscale†</td>
<td>Dentist Patient Dentist Patient</td>
</tr>
<tr>
<td>Speech/Communication</td>
<td>Intelligibility Articulation Velopharyngeal competence Overall speech Documentation</td>
<td>Intelligibility-in-Context scale† Percent Consonants Correct scale† VPC graded rating scale† CLEFT-Q Speech and Speaking subscales† Standardized speech and language sample</td>
<td>Family SLP SLP Patient SLP</td>
</tr>
<tr>
<td>Otologic health</td>
<td>Hearing Otologic health</td>
<td>Puretone average Otologic health screening questions</td>
<td>Audiologist Audiologist/Otolaryng./Family</td>
</tr>
<tr>
<td>Breathing</td>
<td>Nasal breathing</td>
<td>NOSE questionnaire</td>
<td>Patient</td>
</tr>
<tr>
<td>Appearance</td>
<td>Nasolabial appearance Facial profile Smile Documentation</td>
<td>CLEFT-Q Face subscale† CLEFT-Q Jaw subscale† CLEFT-Q Dental subscale† Standardized series of facial photographs</td>
<td>Patient Patient Surgeon</td>
</tr>
<tr>
<td>Psychosocial development</td>
<td>Sociometrics Psychometrics</td>
<td>CLEFT-Q Social Life subscale† CLEFT-Q School Life subscale † CLEFT-Q Feelings subscale†</td>
<td>Patient Patient Patient</td>
</tr>
<tr>
<td>Burden of care</td>
<td>Total number of interventions requiring anaesthetic</td>
<td>Medical record</td>
<td>Administrative staff</td>
</tr>
</tbody>
</table>

† Validated instruments.
4.12. Limitations of PROMs questionnaires as outcome measures

Whilst some advocate that PROMs are the only true measures of well being and life satisfaction (Layard, 2011), they are not without limitation (Devlin and Appleby, 2010). A particular problem when relying on personal opinion of morbidity and welfare is the dissimilarity in people’s perception of health: It is quite possible that two individuals with the exactly the same level of health will report their status differently in a questionnaire (Bago d’Uva et al., 2011, Jürges, 2007, Baron-Epel et al., 2005).

It is suggested that responses will vary according to the responders personality, level of acceptance of their condition and their cultural or environmental viewpoint (Goldberg et al., 2001). Additionally, opinion might reflect a 'response shift' whereby individuals change their perception of their condition through accommodation over time (Sprangers and Schwartz, 1999). This concept has been alluded to in relation to several aspects of CLP such as quality of life (McDougall et al., 2013), malocclusion (Masood et al., 2013) and oral health (Ruff et al., 2016, Arrow and Klobas, 2016).

Variation in self perception has also been shown to be closely related to the responder’s level of education, the observed health of those around them, their concept of good health, expectations and knowledge of the disease and its potential management (Bago d’Uva et al., 2011): It is well reported that in industrialised countries poorer people report higher morbidity and have higher mortality than the rich (Idler and Benyamini, 1997, Kunst et al., 1995, Kaplan et al., 1996, Evans, 2001, Deaton, 2008). Conversely, it is commonly observed that this is reversed in developing countries, with the poor reporting lower levels of ill health than the rich, despite having higher rates of mortality possibly associated with lower expectations (Murray and Chen, 1992, Sen, 2002, Murray, 1996). This was clearly demonstrated by Sen (2002) in his comparison of self-reported health morbidity in different Indian states and the USA: The highest self-reported morbidity was in the USA, followed by Kerala then Bihar yet life expectancy, levels of education and availability of health services were completely the reverse. This information is critical in relation to this project since, as described in section 2.5, many of the participants are poorly educated and live in poor socio-economic conditions with
limited access to health care, so it could be supposed that a PROM would under-report the influence the impact of their CLP in comparison with an individual living in a more developed area.

Whist not disputing that the internal view of the individual is a valuable component when assessing outcome, these studies indicate that a total reliance on such information for informing policy change could be misleading (Kumar et al., 2011). It has been suggested that these biases make data from self-reported questionnaires difficult to compare across countries, populations and sub-populations unless some measure of correction is implemented, particularly where there are socio-economic, cultural and demographic variables (Murray et al., 2003). Jylhä (2009) observes that "hardly any other measure of health is more widely used and more poorly understood than self-rated health". Although under certain circumstances PROMS are reported to indicate a comparable health status as objective measures (Goldberg et al., 2001, Singh and Junnarkar, 2014), other researchers question whether they merely reflect differences in perception of health, rather than a true depiction of health status (Van Nes et al., 2010, Jylhä, 2009).

One avenue which has been suggested to overcome these issues is the incorporation of vignettes or hypothetical scenarios as a means of calibration (Dowd and Todd, 2011, Murray et al., 2003). An individual or rater is asked to rate a standard scenario alongside their personal assessment; since these descriptions are fixed, it removes the personal reporting bias and gives an indication of the threshold by which they make their own assessment (Bago d’Uva et al., 2008, d’Uva et al., 2011, Tandon et al., 2003, King and Wand, 2006). For this reason the TOM-clp incorporates hypothetical case scenarios as a benchmark when training assessors which increases the reliability when comparing data from different sites.

Other points to consider are that a PROM questionnaires can only be considered as an outcome measure if they are repeatable and used to compare a situation at different times, which can be problematic as they can be quite labour and resource intensive to implement (Black, 2013). They also require appropriate translation and appropriate levels of literacy in order that they can be completed accurately (Whoqol Group, 1994). Lack of health literacy is important as it can be a barrier
not only to completion of questionnaires, but also access to health information for marginalised communities (Wolf et al., 2006, Sentell and Halpin, 2006)

Whilst PROMS can be considered as a valuable adjunct to health assessment it is widely suggested that they are treated as complimentary to biomarkers and should not aim to replace them (Black et al., 2016, Weldring and Smith, 2013). The TOM has an advantage in this respect as, whilst the clinician bases their assessment on the subjective opinion of the client, its holistic nature also incorporates objective information acquired from the clinical history, interview and examination.

**4.13. Paper based and electronic data collection methods**

Although surveys are a common means of data collection in low and middle income countries, data collection is commonly compromised owing to difficulties with logistics and administration capacity of personnel (Tomlinson et al., 2009). Problems are compounded with paper based surveys owing to the cost of printing, delivery and collection, data entry errors, incomplete surveys, storage costs and the double entry nature of the data analysis (Shirima et al., 2007).

We now live in a technological age and the use of online surveys and data collection has become commonplace, increasing the capacity to collect large amounts of real time data over a large geographical area (Yu et al., 2009). It allows for data collection and data entry to be undertaken simultaneously which minimises transcription errors (Shirima et al., 2007, Juniper et al, 2009) and is generally considered more efficient and more accurate than traditional paper questionnaires (Juniper et al., 2008, Greenlaw and Brown-Welty, 2009, Schobel et al., 2017). However, there is a risk of targeting a different socio-economic group in terms of income, age, educational status and understanding and availability of technology (Horevoorts et al., 2015, Marcano-Belisario et al., 2014), which could potentially exclude the rural minority groups living in socioeconomic deprived (Barentsz et al., 2014), despite 88% of Indian households having a mobile phone (Indian Government, 2011),

Although proposing to be less time consuming and cheaper (Greenlaw and Brown-Welty, 2009, Schobel et al.), where technology is unpredictable, this advantage can
be lost owing to technical difficulties both with the hand held device and the download process (Zhang et al., 2012). Also, their reliance on internet technology and risk of theft, loss, damage or corruption means that they are not without complication (Yu et al., 2009, Tomlinson et al., 2009).

Privacy issues are also a consideration with online surveys, particularly when individuals are using their own devices to complete the survey (Zhou, 2012). Concerns have proved to restrict peoples willingness to participate in online surveys, particularly in relation to sharing sensitive heath information online (Atienza et al., 2015, Cocosila and Archer, 2010). Validity must also be confirmed for online surveys, although some researchers show good comparability between paper based and online surveys (Mulhern et al., 2015, Marcano-Belisario et al., 2014). Additionally, it has been shown to reflect a degree of response bias between the two methods, questioning the comparability of the two methods in all circumstances (Bowling, 2005, Juniper et al., 2009, Juniper et al., 2008).

4.14. Rationale behind this research

Appraisal of the literature highlights the need for an outcome measure which can reflect the impact of untreated CLP, and its subsequent intervention, on well-being and social functioning in the developing world. Such an instrument must be quick and straightforward to understand without the need of specialist qualification, internationally acceptable, culturally sensitive and administrable where resources are limited. The TOM-clp originates from the ICF which was designed specifically for use on a global scale, it is simple to use, is not language specific and does not require specialist equipment or professional staff to administer. It has been successfully piloted under research conditions and shown to have good reliability after only a short training period and shows excellent potential for use in addressing the impact of untreated CLP in the developing world. It is the aim of this project to assess whether the TOM-clp can be implemented for this purpose at four cleft centres in India.
4.15. Objective of this research

This research explores the viability of implementing the adapted version, the TOM-clp (Rees et al., 2016b), in four cleft centres in India. In order to assess whether the results of the TOM-clp represents a balanced and representative view of the impact of CLP and its subsequent intervention, a mixed method approach is employed using the WHOQoL-BREF and semi-structured interviews as a means of data comparison. The qualitative interview data is analysed using a thematic framework in order to achieve a descriptive overview of the impacts of CLP and gain insight into the subjective perceptions of the participants whilst also ensuring transparency.

4.16. AIMS

The research has three aims one of which is divided into multiple research questions as summarised below.

**AIM 1:** To identify the underlying reasons why CLP remains untreated in India.

- What services are currently available for CLP repair, how are these services promoted and what facilitates or limits their accessibility?

**AIM 2:** To explore the psychosocial impact of untreated CLP for individuals and investigate the impact of late repair on activity, participation and well-being from the perspective of the individual and close friends and family

- In what way does untreated CLP impact the activity, participation and well-being of the individual?
- In what way are an individuals’ activity, participation and well-being impacted by intervention?

**AIM 3:** To examine the value and viability of an adapted TOM to explore the impact of untreated CLP and evaluate how well the TOM-clp can be implemented as a culturally sensitive tool to demonstrate change in relation to intervention.
CHAPTER 5: RESEARCH METHODS

This section will outline the philosophy and design of the research and describe in detail why a mixed method pragmatic approach was employed to address the central research questions in this study.

5.1. Introduction to methodology

The literature review demonstrated that, whilst many questions regarding the outcomes of late intervention for CLP have been answered, there remain identifiable gaps in our knowledge: Specifically, why the customary practice of using phonetic speech analysis as a measure of success or failure, may be limiting our broader understanding of the impact of delayed surgical and rehabilitation intervention for CLP.

As outlined in chapter 2, society places a high value on outward appearance (Klein and Shiffman, 2006) and verbal fluency (Blood et al., 2003, Boyle et al., 2009). Consequently, anomalies linked with CLP such as; facial differences, speech disorders and nasal regurgitation, are commonly associated with stigmatisation and social discrimination (Klein and Shiffman, 2006, Masnari et al., 2013, Blood et al., 2003, Boyle, 2016, Kapp-Simon et al., 1992). This is linked to diminished self esteem and a reluctance to integrate socially leading to feelings of marginalisation and a compromised quality of life (Strauss et al., 2007, Topolski et al., 2005).

This study intends to ascertain the extent to which this is the case for individuals in India who had experience of untreated CLP late in life, and to assess the impact that intervention has on these issues. The research aims to redefine the question of outcome by exploring the experiences of individuals in terms of socialisation and well-being rather than focussing on speech as the pivotal source of information.
5.2. Methodological Background

The study analysed accounts from 85 individuals aged between 6 and 36, who had received corrective surgery for their cleft lip and palate later in life. Data was collected using a mixed methods approach over a two year period at four cleft centres in India details of which will be provided in CHAPTER 9:

5.2.1. Philosophy

This study is designed around the underlying research paradigm of pragmatism (Biesta, 2010, Yvonne Feilzer, 2010). In this section, four alternative research paradigms will be presented before making a justification of the suitability of pragmatism.

In contrast with the numerical and statistical focus of quantitative research, a qualitative approach focuses on the study of life experiences, centering the human participant at the heart of the research (Polkinghorne, 2005, Alasuutari, 2010, Berg, 2007). Consequently, qualitative data is usually gathered in the reflective and spoken form and requires innovative research methods to capture meaning from the less tangible and multilayered subjects than are generally associated with quantitative methods (Berg, 2007, Denzin and Lincoln, 2000).

Although anthropologists have been collecting qualitative data since the early 20th century, the emergence of qualitative methods in mainstream science has really only materialised in the last 40 years (Polkinghorne, 2005). As the research community has become come to value subjective experiential opinion to explain phenomena, enthusiasm for this branch of research has grown rapidly, particularly in the fields of education and health care (Alasuutari, 2010, Kidd, 2002, Lee et al., 1999, Krahn et al., 1995). However, as outlined below, interest in this technique has also brought with it an array of controversies surrounding definitions, terminology and research philosophy, which is reflected in the materialisation of a plethora of different approaches and methodologies (Bolderston, 2014).

Over the decades, purists considered qualitative and quantitative research as antagonistic, in that they are completely separate entities with no overlap between
the two (Denzin and Lincoln, 2000, Guba and Lincoln, 1994, Morgan, 2007). The concept of using multiple methods in research was first formalised by Campbell and Fiske in (1959) when they proposed that comparing multiple converging traits could enhance the understanding of the phenomenon under investigation (Johnson et al., 2007, Fiorini et al., 2016). This process, later coined as 'triangulation' by Webb et al. (1966) became the foundation of combining research methods as a means of augmenting rather than confounding research (Fiorini et al., 2016). Morse (1991) further elaborated on this concept endorsing the combination of qualitative and quantitative methods however, it was Tashakkori and Teddle (1998) who consolidated and defined the theory of mixed methods, with the publication of their textbook 'Mixed methodology: Combining qualitative and quantitative approaches'.

Over the last 20 years, the development of mixed methods as a third methodology has gained favour and has been acknowledged as a means of gaining insights beyond those attainable using simply qualitative of quantitative methods alone (Johnson et al., 2007, Creswell and Clark, 2007, Mertens, 2014). However, whilst some researchers embraced the concept and incorporated into their research philosophy (Morgan, 2007, Fox and Alldred, 2017) others still argued that the apparent incompatibility underpinning mixed-methods invalidated its practicability entirely (Guba and Lincoln, 1994, Sale et al., 2002). The crux of the debate surrounded the foundation of reasoning behind different methods of research (Crotty, 1998): Traditionally, quantitative research begins with a hypotheses and uses a deductive approach to explore the phenomenon and look for causality. Conversely, qualitative research is more open minded and commonly associated with an inductive approach, exploring a phenomena and generating a theory according to the information emerging from the data (Berg and Lune, 2004, Neuman, 2002). Bryman (2012) suggest that the deductive nature of a quantitative approach and the inductive nature of a qualitative approach make the two mutually exclusive. This apparent dichotomy, traditionally referred to as the 'paradigm wars', largely revolving around ideological convention and epistemological issues, has meant that finding a legitimate paradigm to justify its foundation is an on-going debate, (Bryman, 2006, Morgan, 2007, Hammersley, 1992, Gage, 1989, Hall, 2012).
The term Paradigm was introduced by Thomas Khun in 1962 in his book *The Structure of Scientific Revolutions*, as a means of researchers summarising their beliefs (Kuhn, 1962). The exact definition of the term has been disputed and redefined many times over the years, resulting in a confusion which has meant the word ‘paradigm’ is commonly used to mean different things in different contexts (Morgan, 2007). Kuhn himself regretted his choice of word and in the second edition of his book adds a post-script suggesting that the term ‘disciplinary matrix’ may have been more appropriate descriptor (Kuhn, 1970). The most widely accepted explanation of the term paradigm is to describe a framework of common beliefs which are laid down by a researcher to clarify how problems should be addressed (Gage, 1989, Scotland, 2012). In the words of Teddlie and Tashakkori (2009, p84) "a worldview, together with the various philosophical assumptions associated with that point of view". The five commonly accepted world views, listed by these authors, are identified as: Positivism, constructivism, transformative, postpositivism and pragmatism. This list assumes the division of postpositivism and positivism as two separate paradigms, although this distinction is not always made (Creswell and Poth, 2017).

It is difficult to define each of these philosophies succinctly, however, the following sections will attempt to describe their essence in relation to their relevance to the research described in this thesis and explain the reasons behind their rejection or acceptance to describe the methodology for the current project.

### 5.2.1.1. Positivism

The French philosopher, Auguste Comte first introduced positivism in 1830, when he declared that only knowledge based on quantifiable observation was considered reliable and trustworthy (Comte and Bridges, 1865, Cohen et al., 2013). The positivist principle rejects natural philosophy and metaphysics and views the world as being made up of discrete measurable elements that interact in a way known as atomistic ontology (Bernard and Bernard, 2012, Neuman, 2002). It relies on a neutral, objective researcher, developing and testing an hypothesis based on purely deductive reasoning, with little room for human interest or abstract thought. Since this paradigm is based entirely on logic and reasoning it lends itself to statistical analysis and quantitative research (Bernard and Bernard, 2012).
As this study has its foundation in exploring a phenomenon from human perspective and proposes a large qualitative component, positivism was seen as inappropriate and rejected.

**5.2.1.2. Constructivism**

A constructivist approach, as outlined by Cohen and Manion (2013), aims to evaluate a phenomenon based on human experience, supported by information relating to the personal perspective of participants. Sometimes known as the 'interpretive position', it is an encompassing term which includes various formulations influenced by a variety of different philosophical reflections such as phenomenology (Zuboff, 1988), hermeneutics (Boland and Day, 1989) and ethnography (Kenny, 2010, Walsham, 1995).

The underlying assumption of constructivism is that knowledge is a subjective product of social experience and that the participants view and impact of the researcher are integral to the situation being studied (Cohen et al., 2013). This reliance on individual viewpoints requires data to be collected by means of observations, direct interaction and exchanges between researcher and participant (Mackenzie and Knipe, 2006). By definition data is therefore interpretive in nature and will involve inherent subjectivity in its analysis. To reduce this bias, constructivist research requires that the researcher does not have an underlying theory about the area being studied, rather they allow the theory to develop inductively throughout the research process (Creswell, 1998).

A popular analytical framework of constructivism is grounded theory which was introduced by Glaser and Strauss in 1967, as a means of legitimising qualitative research (Glaser and Strauss, 1967). There are many strands of grounded theory, all of which have a commonality in that; data is collected by means of open questions; the researcher should enter the process without any preconceived ideas; data is collected and analysed concurrently so interviews continue only until the point of saturation; categories are constructed inductively during analysis using theoretical sampling and coding; the process looks to explain a behaviour rather than just describe it; and analysis is purely qualitative (Charmaz, 2014, Starks and Brown Trinidad, 2007).
Whilst the prime purpose of this study is to examine the impact of CLP from an experiential perspective of its participants, it is designed in relation to a theory developed in previous research (Rees et al., 2016b). The requirement of large numbers of interviews over an intense 2 week period would not have allowed for concurrent analysis and the semi-structured format was based on the researchers prior knowledge. For these reasons a constructivist approach cannot be considered appropriate and must be rejected.

5.2.1.3. Transformative

The transformative paradigm emerged as a world view in the 1980's, amid a belief that socio-political research, dealing with the impact of social injustice on marginalised communities, could not be addressed using a constructivist viewpoint (Mackenzie and Knipe, 2006). Transformative research has a political agenda and focuses on cultural complexities surrounding human rights and social reform (Mertens, 2014, Canales, 2013, Mertens, 2007). It relies on participatory and inclusive mixed methods to gain an understanding of participants' experiences of topics such as incarceration, marginalisation and emancipation at an individual, and community level (Romm, 2015, Trevors et al., 2012).

The core principles of a transformative approach relates to studying how community strengths and cultural values of marginalised communities form the basis of social injustice (Mertens, 2012, Mertens, 2007). In contrast, whilst the individuals in this study are often marginalised, they are not a homogenous community with common beliefs, they are a divergent cohort of individuals who, by chance, have a common barrier impacting their inclusion into society. For this reason the transformative paradigm was not considered appropriate and was rejected for the purposes of this study.

5.2.1.4. Post-positivism

Post-positivism moves away from positivism by accepting a level of realism, recognising that the world is driven by unpredictability and observations of it are fallible (Lincoln and Guba, 1985). Post-positivism does not reject the concept of positivism entirely but proposes that ‘ultimate truth’ is not accurately measurable therefore, truth is based on probability rather than certainty (Mertens, 2014).
Post-positivism also accepts that there is a relationship between the subject and researcher which may encourage innovative discovery of knowledge but may also introduce bias (Bryman, 1984). To overcome this, post-positivists encourage the use of multiple measures in order to establish triangulation and get a more thorough and balanced understanding of the phenomenon under scrutiny.

Although post-positivism is inclusive of mixed methods, it has its foundation in experimental methodology favouring a quantitative approach whilst including a qualitative component in terms of fallibility and to provide context (Cruickshank, 2011). Since the focus of this study is the experiences of the participants it is predominantly qualitative in nature the post-positive paradigm is rejected as inappropriate.

5.2.1.5. Pragmatism

Miles and Hauberman (1994) propose that research rarely conforms precisely to any one given world view and that researchers are often compelled to bend their methodology to fit the peculiarities of their study. This is reiterated by Fox and Alldred (2017), who propose that it is better to justify what a chosen research tool can do to enhance research, rather than describe how it exemplifies a particular philosophy. In contrast to the paradigms outlined above, pragmatism follows this reasoning and does not limit its philosophy to a single reality, it centres its premise around the research question, applying the most appropriate method to answer that particular problem (Creswell and Clark, 2007, Mays and Pope, 1995, Tashakkori and Teddlie, 2008). Since pragmatists do not generally profess to hold a world view, there is an argument that it should actually be regarded as a practical approach to research rather than a philosophical paradigm (Morgan, 2007).

As outlined earlier, a key obstacle in some researchers accepting a mixed method approach, is the apparent dichotomy between objectivity and subjectivity which some consider still exists between quantitative and qualitative methodologies (Mackenzie and Knipe, 2006). Hanson (2008) proposes that this partition is more reflective of the political divisions among researchers as it is a divergence in methodologies, suggesting that following a single paradigm is simply an historic convention rather than a productive and beneficial means of achieving a goal.
Pragmatists argue that methodologies should not be considered mutually exclusive and the focus should be on mixed methods, integrating different research methods to enrich our understanding of a phenomenon (Creswell, 1998, Patton, 1990, Patton, 2002). They aim to bridge the gulf between positivism and constructivism, accepting the advantages and disadvantages of each whilst taking meaning from both (Morgan, 2007, Biesta, 2010). Rather than treating qualitative and quantitative methodologies as divergent, pragmatists consider them as a continuum, from which a researcher seeks aspects which compliment their research to best effect (Onwuegbuzie and Leech, 2005, Newman and Benz, 1998). For instance; rather than choosing between either induction or deduction, a pragmatist might take an abductive approach, incorporating both to develop a theory based on all available information (Morgan, 2007). To do this the researcher must be subjective in their own thoughts and reflections whilst also being objective in their data collection and analysis. If during analysis qualitative data shows recurrence and stability, it might be appropriate to represent it graphically (Pope et al., 2000) or analyse it using quantitative techniques (Yvonne Feilzer, 2010).

A key advantage of pragmatism is that it accommodates the concept of transferability of knowledge into other settings: Unlike a quantitative approach, which requires a strictly homogeneous sample or a large random sample to be representative of the population, qualitative research accommodates more purposeful sampling, relying on a smaller study a population which is not strictly homogenous, but has some desired level of comparability (Marshall, 1996, Marshall and Rossman, 2014, Byrne, 2001). This facilitates repeatability with a different study cohort and encourages dissemination of the research in a wider context (Marshall, 1996). This was an important consideration in this study, as it involves the development of an outcome measure designed to be useable in diverse environments.

After consideration it was felt that this study did not fall comfortably within any of the traditional world views and, as a consequence, a pragmatic mixed methods approach was considered the most appropriate alternative framework for this research. The concept that qualitative and quantitative methods can be taken as
complementary rather than antagonistic and that both are necessary to glean a thorough understanding of a phenomenon, mirrors the view of the researcher and reflects the foundation of this study (Creswell and Clark, 2007, Johnson et al., 2007, Bowleg et al., 2016). For these reasons a pragmatic approach was taken in the design of this research.

It is acknowledged that conflicting views occur in qualitative research where the emic perspective; that of the participants vary with the etic perspective; that of the researcher (Morris et al., 1999, Maxwell, 1992). Attempts have been made in the design of this study to balance these tensions by including multiple sources of data to triangulate the results rather than rely on a single source of information.

5.3. Qualitative methods

5.3.1. Interviews

This research aims to understand the lived experiences of the participants and to get an insight into how their lives are impacted by their untreated CLP and subsequent intervention. It was felt that direct dialogue would be the main instrument of choice to achieve this objective, and interviews were selected as the principal means of data collection.

According to Kvale (2006) interviews offer a powerful means of eliciting detailed personal views about life experiences, which can then be described and interpreted. As well as being economical and cost effective (Neuman, 2002), this form of personal narrative allows individuals to express themselves using their own frame of reference and expose and explain perceptions and emotions in way that may not be achievable using other techniques such as questionnaires (Blaxter and Poland, 2002, Mason, 2006, Edwards and Holland, 2013, Green and Thorogood, 2013). Interviews can be approached in a plethora different manners but will be considered here in three styles; unstructured (UI), structured (SI) and semi-structured (SSI) (Gill et al., 2008, Edwards and Holland, 2013, Nelson, 2009, DiCicco-Bloom and Crabtree, 2006). Each style differ in the amount of control the interviewer has over the direction of the interview (Stuckey, 2013), and the
following will describe why it was decided that an informal SSI would be most appropriate for this study.

The concept of the SSI, formerly known as the focused interview, was conceived by Merton and Kendall (1946) as a means of managing large volumes of communication. A SSI follows a relatively open naturalistic framework which facilitates participants to have the trust and confidence to elaborate on potentially sensitive topics surrounding their life experiences (Berg, 2007, Ryan et al., 2009). This is in contrast to the SI which are more formal and comprise predetermined closed questions, designed to answer specific enquiry and inhibit extensive dialogue (Edwards and Holland, 2013, Denzin and Lincoln, 1994).

The questions in a SI are specifically worded and ordered which requires the participant to provide immediate answers at the time of asking, without the option of clarification or elucidation (Corbetta, 2003). In contrast, the flexibility of the SSI, means that questions can be readdressed, redelivered in a different context and rephrased or reworded to ensure the participant fully comprehends the meaning (Blaxter et al., 2006, Green and Thorogood, 2013). This was considered essential in this project in order to accommodate the wide range of ages, genders, divergent ethnicities and educational abilities of the participants and the interpreters (McIntosh and Morse, 2015). Unlike a SI the plasticity of the SSI also allows the interviewer the chance to expand on a theme or probe further into a topic of conversation of interest during the interview (Corbetta, 2003, Berg and Lune, 2004). This can enrich the data and clarify points of apparent conflict and ensures that the researcher has fully understood the point the participant is making, and that the resulting information is accurate (Dörnyei, 2007, Irvine et al., 2013).

As mentioned, flexibility and freedom in an interview promotes dialogue and allows both interviewer and interviewee the elaboration on topics of their choosing (Dörnyei 2007). This is the founding principle of the UI, which is designed to produce very rich data and great profundity which may increase validity, as the process often elucidates not only an answer to a question but also the reason behind that answer (DiCicco-Bloom and Crabtree, 2006). A disadvantage of UIs is that they are time consuming so, whilst they are excellent for detailed case studies, they were not considered suitable for the number of interviews in this project.
(Ryan et al., 2009). Whilst UIs are valuable in their richness, their open structure means that the analysis of UIs is heavily reliant on theoretical reasoning and the process is not reproducible because each interview, by its very nature, is unique, thus particular areas of interest may remain unaddressed. In contrast, the degree of consistency between semi-structured interviews means that there is an element of replication and the relevant area of the research is covered, allowing the responses to be compared and potentially quantified (McIntosh and Morse, 2015). This was considered important since it allowed contrasts and similarities between different participants to be evaluated, it meant that the process could be replicated reliably at different field sites and equally importantly improved the transparency and reproducibility of the research for future studies.

One advantage of an UI is that the interview can progress tangentially in accordance with the feelings of the participants so that they can elucidate new and unexpected information. By the same conjecture, the lack of direction means that important subjects may not be discussed and useful information may be missed. The advantage of a semi-structured technique is that it is executed around preset themes which allows liberty in the conversation whilst keeping the conversation within pre-determined parameters (Berg and Lune, 2004). This helps to direct the content of the interview to ensure that all the pre-requisite topics are covered but it also avoids the generation of excessive information which could lengthen the interview, transcription, coding and analysis process.

The advantages and disadvantages of each type of interview technique were summarised and their merits compared (table Table 5:1). It was decided that semi-structured interviews would be most appropriate to address the research questions in this study, since they would ensure that appropriate questions were asked whilst still allowing new concepts to emerge from the data (Glaser and Strauss, 1967). It was accepted that, whilst multiple sequential interviews with each participant produce maximum breadth and content (Seidman, 2013), this was not possible in the current study due to the remote location of data collection and restricted time on each site.
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5.3.2. Selecting an interpreter for the interview process

This study was cross-cultural and interviews were undertaken through an interpreter. It is acknowledged that the reliance on the use of an intermediary and introduction of a second language can mean that expressions of emotions and feelings may not be represented as richly and competently as would be expected in a native language, also nuances and meanings may become lost or misinterpreted in translation (Fernández Guerra, 2012, Squires, 2009, Van Nes et al., 2010). To overcome this, consideration was initially given to the employment of a professional interpreter to maximise the accuracy of the translation. However, not only would this have significantly increased the cost of the research, it would not have been possible to predict in advance which languages would be required for each interview. In addition, unless the interpreter was familiar with comprehending the speech of a cohort such as this, who potentially have profound difficulty in communication, fluency may not have improved the accuracy of the translation. Critically it was also felt that participants would be more comfortable and open if the interviews were undertaken by someone known to the participants which would not have been possible with a professional interpreter (Squires, 2009, Van Nes et al., 2010).

For these reasons, where the participant was not conversant in English, a member of the cleft team acted as the translator for the interview process.

5.4. Instruments

5.4.1. TOM-clp

The TOM-clp was implemented as described in section 4.6 at all four field sites. Prospective scores were recorded independently by trained staff at three centres, one in Assam and two in Hyderabad. Scores were taken prospectively and retrospectively by the PI, in conjunction with face to face interviews, at all three sites in Hyderabad. This process will be discussed in detail in section 7.2.1
5.4.2. WHOQoL-BREF questionnaire

It was decided to use the standardised, validated WHOQoL-BREF questionnaire as a means of triangulation and also to test the construct validity of the TOM-clp.

5.5. Ensuring reliability, validity and objectivity

Although the terms reliability and validity are generally applied to quantitative research they are essential strategies to adopt in order to demonstrate rigor and ensure the credibility of qualitative research (Noble and Smith, 2015). Lincoln and Guba suggest the replacement of these terms with ‘truth’ ‘consistency’ ‘neutrality’ and ‘applicability’ in relation to qualitative research (Lincoln and Guba, 1985) however the underlying concept of ensuring accuracy and trustworthiness remain the same (Mays and Pope, 2000). This research employs mixed methods which is reflected in the combined descriptive terminology used in this thesis.

The problem is compounded by the diversity of methodology associated with qualitative research so there is no singular agreed measure to confirm its quality and validity (Tracy, 2010, Caelli et al., 2003). Checklists have been produced with suggestions of how to increase confidence (Caelli et al., 2003, Tracy, 2010), however Barbour (2001) argues that merely adopting these criteria does not equate to quality particularly if they are not pertinent to the underlying philosophy of the research. The following section outlines how the PI has endeavoured to ensure quality within the remit of this research project.

5.5.1. Validity

In order to be credible it is essential that studies give an accurate and representation of the participants account of the social phenomena under scrutiny. This refers not only to the validity of the data but also to any inferences that the researcher draws from them (Hammersley and Atkinson, 2007). In contrast to quantitative research, which revolve around relationships and causality of specific variables, qualitative research is more naturalistic and relies more heavily on interpretation. This puts a greater responsibility on the researcher to establish the parameters of their research for example to determine how long data collection
should continue, at what point the data is saturated and how best to interpret and analyse that data. This freedom to define the parameters brings with it a need to authenticate the associated rational behind those decisions.

**5.5.1.1. Member checking**

Where the PI acts as data collector and analyst there is the potential for their own personal viewpoint and principles become imposed in dominance over those of the participant (Miles and Huberman, 1994, Jennifer, 2002). This risk can be reduced by confirming with the participants that the information they have provided is a true representation of their beliefs through the process of 'member checking' (Birt et al., 2016, Lincoln and Guba, 1985).

Member checking typically involves transcribing and analysing an interview and returning to clarify its accuracy with the participants (Vogt and Johnson, 2011). Whilst many consider this process as a means of improving validity and accuracy in the interview process (Lincoln and Guba, 1985, Byrne, 2001, Doyle, 2007, Morse et al., 2002) it is not without its critics in that it can be seen to cause confusion with participants not remembering or changing their story since the time of the first contact (Sandelowski, 1993, Harper and Cole, 2012, Mays and Pope, 2000).

The nature of this study did not allow for a second review date so member checking was undertaken as a contemporaneous process during the interviews (Harper and Cole, 2012, Brewer and Hunter, 2006). During the interview, where there was need for clarification, the PI summarised or restated information and questioned the participant, via the interpreter, to determine the accuracy of understanding. Participants were thus given the opportunity to disagree or affirm their position and add additional material if they felt this added to the completeness of their views. This process helped to establish credibility and improve validity by ensuring accuracy of the interview process (Creswell and Clark, 2007).
5.5.1.2. Triangulation

Triangulation engages multiple methods of data generation within one study. This can offer a number of benefits including enrichment of the data by elucidating complimentary information associated with the phenomena under investigation. It can be used to assess consistency in the findings but also may reveal unexpected divergences which may enhance the data interpretation (Tashakkori and Teddlie, 2008, Johnson et al., 2007). Conversely, triangulation is often used to corroborate findings and as a method of validation. However this assumes that the introduction of a second data source will compensate for weaknesses in the first which may not always correspond and could be considered controversial. With this in mind each technique of data collection must be vigorous in its own right however triangulation is still a useful means of ensuring the research is robust and well designed (Kuper et al., 2008, Sandelowski, 1993). Triangulation was employed in this research by engaging three different data collection techniques and comparing the information from each with the aim of identifying patterns of convergence and corroboration.

5.5.1.3. Reflexivity

However carefully a project is designed and implemented it must be assumed that there is an underlying influence caused by the researchers own beliefs, assumptions and biases which may shape the results (Mays and Pope, 2000). Since this research follows on from a previous pilot study it is fair to assume that there will be preconceptions associated with this. Whilst it is acknowledged that that prior information may lead to bias and prejudice in research the author has made every effort to remain open to new ideas and use this knowledge to enhance the understanding of the phenomenon and develop a deeper understanding of its complexity.

Attempts have been made to minimise the bias by maintaining an open dialogue with other researchers throughout the process and keeping a reflexive journal to record on-going methodological decisions (Sandelowski, 1993). Information from these commentaries were openly discussed with other clinicians involved in the research and fed into the analysis process as the research progressed.
5.5.1.4. Avoiding Bias and Peer review

A review by additional individuals familiar with the research provides support and can assist in challenging the views of the PI (Lincoln and Guba, 1985). This approach was used throughout the development and analysis of the project to provide feedback and critical review which to improve credibility (Creswell and Miller, 2000).

5.5.1.5. Record keeping

In order to present a clear account of the research process, define concepts and explain conclusions, every attempt was made to ensure meticulous record keeping. Care was also taken to document a clear decision trail and include outlying data to ensure consistency and transparency.

5.6. Ethical considerations

The focus of qualitative research is centred around a participants willingness to share their experiences and as such the ethical issues must often be considered differently from that of quantitative studies. Ethical considerations include how the potential participants are approached and the information accessed as well as the effect the research may affect the participants.

It was acknowledged that topics covered in this research may prove sensitive and upsetting for the participants involved. With this in mind ethical consideration was made in relation to the vulnerability of participants, issues of age, gender, language, culture and literacy, recruitment, location, anonymity and confidentiality. In addition care was taken to protect the participants and researcher from either physical or emotional harm throughout the process and conflicts of interest were accounted for specific ethical considerations relating to these matters will be discussed in more detail and in context throughout this section.
5.6.1. Ethics approval

Ethics approval was gained from the University of Sheffield and the Indian Council of Medical Research Institutional Review Board (IRB).

The University of Sheffield ‘Ethics Policy Governing Research Involving Human Participants, Personal Data and Human Tissue’ and the ethics policy of the Indian Council of Medical Research was adhered to throughout this research (Appendix 2 and Appendix 3)

Each field site was approached independently to confirm that the above mentioned ethics standards were appropriate and the no further application was necessary regarding regional, local or institutional ethics policy

In addition to the scrutiny of the ethics review boards the researcher accepted the personal responsibility of ensuring the protection of the participants involved in the study. Careful planning and a policy of openness and full disclosure was employed throughout to avoid misunderstandings and unanticipated ethical dilemmas arising during the research process.

5.6.2. Payment

Participants were interviewed only during their regular attendance at the clinic so as to avoid incurring additional travelling expenses. No financial input or incentive was offered.

5.6.3. Potential harm to participants

Interviews were handled in a relaxing manner so as not to be intimidating for those involved. The aims of the project were discussed fully with the participants and only those individuals who wished to participate were included in the study. Participants were assured that they were free to withdraw at any point during the proceedings without any form of repercussion or embarrassment. It was made clear to the participants, from the outset, that the information given would be confidential and would only be used for the purpose of this study. It was clearly explained that participation would not in any way influence their own case
management in order to avoid influencing their responses with regards expectations from the cleft service.

Health workers were fully informed as to the nature of the research and only those interested in becoming involved were asked to do so. It was made clear that they could withdraw at any point during the proceedings without repercussion or need for explanation.

All proceedings were carried out in a sympathetic manner to ensure that none of the participants were put at risk of any harm or distress. Appreciating that some of the information in this study is potentially of a sensitive nature, it was ensured that support was available in case of individuals becoming distressed during the process. However the experience of the staff members in handling the interviews in an empathetic manner ensured that this was not required.
This chapter will describe how the methodology described in the previous section were employed to answer the research questions relating to the three aims of this study. This section will begin with a review of the research questions including a brief outline how they were addressed. This is followed by a detailed description of the location of the project, participant recruitment specific procedures and analytical methods.

6.1. Research summary relating to each research aim

6.1.1. Aim 1: To identify the underlying reasons why CLP remains untreated in India.

What services are currently available for CLP repair, how are these services promoted and what facilitates or limits their accessibility?

In order to address aim 1 information was sought directly from individuals who had experienced untreated CLP in later life, clinical and non-clinical members of the cleft team and through a literature search. This provided different perspectives on availability of cleft care for this cohort and perceived barriers to its accessibility.

Target populations:

- Individuals with untreated CLP and their families
- Medical and non-medical members of cleft teams including medical personnel, allied health professionals and administrators
- NGOs responsible for funding cleft services
6.1.2. **Aim 2:** To explore the psychosocial impact of untreated CLP In what way does untreated CLP impact the activity, participation and well-being of the individual?

- In what way does untreated CLP impact the activity, participation and well-being of the individual?
- In what way are an individuals’ activity, participation and well-being impacted by intervention?

Information to address aim 2 was sought directly from individuals with experience of untreated CLP late in life along with their friends and family members. Data was collected by the PI and dedicated cleft team members using face to face informal semi-structured interviews and patient recorded questionnaires.

**Target populations:**

- Individuals with experience of untreated CP beyond six years of age.
- Family and friends of individuals with experience of untreated CP beyond six years of age.

6.1.3. **Aim 3:** To examine the value and viability of an adapted TOM to explore the impact of untreated CLP and evaluate how well the TOM-clp can be implemented as a culturally sensitive tool to demonstrate change in relation to intervention

Information to address aim 3 was sought through direct feedback from members of the cleft teams. Topics of discussion included; current departmental policy on audit and outcomes; personal experience of training and execution of the TOM-clp within normal time and staffing constraints; the potential of implementing TOM-clp as a research and auditing tool within their care setting and opinion as to whether the TOM-clp might prove valuable on a wider scale as an auditing and benchmarking tool.
Target populations:

- Medical and non-medical members of cleft teams including medical personnel, allied health professionals and administrators
- Participants involved in data collection

6.2. Location

Research was initiated in two geographical locations of India with data collection undertaken in four sites:

- A comprehensive cleft care centre in Assam, Northern India
- A Specialist Dental and Orthodontic practice in Hyderabad, Telangana, India
- A cleft centre and research institute in Hyderabad, Telangana, India
- A comprehensive cleft centre in Hyderabad, Telangana, India

6.2.1. Identification and recruitment of cleft centres

International cleft centres were identified through NGO websites, personal contacts and internet searches. A personal e-mail was sent directly to the clinical lead of each centre, providing details of the nature of the project and inviting them to get in touch directly with the PI if they were interested in participating. A summary of the proposal was also posted on a British Association of Oral and
Maxillo Facial Surgeons website forum by way of an invitation to get in touch for anyone interested in participating in the research.

Replies were received via e-mail and every response was followed up by the PI. In each case an e-mail dialogue was initiated with the clinical director to clarify the details of the project, what a commitment to participate would involve and to establish that suitable the inclusion criteria were met (section 6.2.1.1). It was explained that data collection would be an on-going process and that at least one individual from the team would need to be appointed and trained for this purpose. Where possible training and data collection would include a field visit by the PI however on-line training would be provided where this was not possible. It was established that there was not any additional funding available for the purpose of this study and that data collection would need to be incorporated into the daily running of their service. When suitable centres were engaged each clinical director proposed a key worker from their team to act as the link for all further correspondence and local recruitment.

One of the core purposes of this study was to identify the impact of intervention for CLP so it was important to identify cleft centres which provided comprehensive follow up care for their patients. Initially it was thought that data would also be collected from short term 'parachute' missions and hospital based centres in order to compare outcomes. However, co-ordination of appropriate quantities of recall data for parachute missions proved unattainable so the limited data from this source was not included in this thesis.

6.2.1.1. Inclusion criteria for participating cleft centres

- Well established cleft centres.
- Provision of late intervention for CLP irrespective of age.
- Regular cohort of patients seeking intervention beyond 6 years of age.
- Offer follow up care.
### 6.3. Timing of data collection

Data was collected over a two year period between March 2015 to March 2017 as outlined in Figure 5.

**Figure 5: Project overview and time-line**

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<th>Project overview and time line</th>
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| **Development phase**  
*Sept 2013 - Dec 2014* |
| • Literature searches and background reading  
• Development of aims and research questions  
• Networking period and development of working relationship with global cleft centres  
• Construction of training material for the TOM-clp |
| **Piloting the training manual**  
*December 2014* |
| **Phase 1:** Provisional analysis of case scenarios with dental nursing staff in Cumbria  
**Phase 2:** provisional pilot of training process in three rounds, updates to the material being made following each round in accordance with feedback.  
• *Round 1:* training of 3 native English speaking dental students at UCLAN, Cumbria  
• *Round 2:* Training of 3 PhD students who use English as a second language at the University of Sheffield human communication sciences department.  
• *Round 3:* Final training of 4 different dental students at UCLAN, Cumbria. |
| **Data collection Assam**  
*(Demographic and TOM-clp data collection only)*  
*March - May 2015* |
| **March:** Three week field trip to Assam by the PI for staff training and initiation of data collection  
**May:** End of data collection in Assam |
| **Data collection Hyderabad**  
*(TOM-clp data collection only)*  
*May 2015 - April 2016* |
| Networking and recruitment of three cleft centres in Hyderabad, including online training of staff members and initiation of data collection |
| **Hyderabad field trip**  
*(Interview data collection by PI)*  
*March 2017* |
| 2 week field trip by PI to undertake 40 semi-structured interviews for framework analysis and TOM-clp data. Period included implementation of WHOQoL-BREF and provisional transcription of interview |
| **All data collection ceases end of March 2017** |
| **Data analysis and write up**  
*March 2017 - Dec 2017* |
| • Analysis of TOM-clp data and framework analysis on interview transcripts  
• Secondary literature review, PhD write up and submission. |
6.4. MATERIALS

- **The TOM-clp. (Appendix 1)** The use of this tool was confirmed by Pam Enderby who currently owns the intellectual property rights for this product.

- **Training manual for the TOM-clp:** (Appendix 7) A purpose developed training manual including case scenarios produced for a prior project, the imagery for which has been approved by Operation Smile and ethics approval for its development and use procured through the University of Sheffield ethics application system (Appendix 2).

- **Literature:** Consent forms and information leaflets were available in English and Hindi (Appendices 4, 5, 6, 7, 8, 9)

- **WHOQoL-BREF. (section 5.4.2)** The use of this tool was confirmed with WHO (Appendix 10) who currently own the intellectual property rights for this product was available in English and Hindi.

- **Audio recorder:** For use in recording the interview sessions and transcription.

- **I-pad / mobile phone:** For recording data using collection app.

6.5. Sampling technique and sample size

For quantitative research, participants are best selected using probability sampling to generate a random, representative sample of a population, aimed at proving statistically that a phenomena is not occurring purely by chance (Polkinghorne, 2005, Greenhalgh and Taylor, 1997). In contrast, qualitative research is generally more interested in exploring depth and richness of opinion rather than counting responses (Gaskell, 2000, Kuzel, 1992, Burmeister and Aitken, 2012). Non-probability sampling is therefore considered more appropriate, targeting a specific population of participants with the supposition that they have relevant experience which can provide a substantial contribution towards understanding the subject under investigation (Berg, 2007, Byrne, 2001).

A plethora of different named strategies of non-random sampling have been identified designed according to different research requirements (Miles and Huberman, 1994, Onwuegbuzie and Leech, 2005, Onwuegbuzie and Leech, 2007).
This study relied on participants having experience with CLP in later life, so the locations and participants were purposefully selected to provide appropriate and 'information rich' cases (Patton, 1990, Merriam, 2002). Sampling was therefore based on five strategies: Homogeneous and criterion sampling, based on the participants having similar characteristic according to strict inclusion criteria: Opportunistic and convenience sampling, taking advantage of including appropriate participants available during the research period: Targeted sampling; Seeking out participants known to fit the inclusion criteria (Onwuegbuzie and Leech, 2007, Miles and Huberman, 1994). Details of how these methods were implemented will be described in section 0.

Qualitative researchers commonly recruit participants through an iterative process; analysing the data sequentially and incrementally, continuing until saturation point, when no new information is emerging from the data (Glaser and Strauss, 1967). Although the process of attaining saturation point, traditionally coupled to grounded theory, has been adopted by many as a 'gold standard' by which sample size is dictated, it is not always appropriate to the underlying philosophy of all qualitative research (Guest et al., 2006, O’reilly and Parker, 2013). Also there is significant subjectivity associated determining 'saturation point' as it will be dependent on the depth to which the data is analysed (Charmaz, 2014, Corbin and Strauss, 2008).

Ritchie et al. 2003 suggest that sample size will be dictated by the heterogeneity of the cohort, the extent to which the analysis will segregate the data into smaller groups, the data collection method and the and the resources available. These concepts are now taking favour (Mason, 2010, Green and Thorogood, 2013, Gaskell, 2000) and it is argued that a sample should reflect the research (Marshall, 1996). Morse and Field (1995) conclude that appropriateness is more important than convention and that focus should be on obtaining the information to answer the research question rather than constraining the sample numerically (Bowen, 2008).

To conclude, there is little consistence in the literature regarding sample size for qualitative research: In Mason’s (2010) literature review, sample sizes ranged from five to three hundred and fifty: Charmaz (2014) suggests twenty five
participants is appropriate for a small project, Creswell (1998) suggest between ten and thirty for phenomenological and grounded theory research. However, in 2002 Creswell then suggest 15 - 20 interviews as appropriate for research involving grounded theory yet Morse (1994) suggests between 30 and 200 encounters are required for meaningful ethnographic studies. Owing to the lack of agreement in the literature the sample size for this project was selected as suggested by O'Reilly and Parker (2013) in accordance with the requirements and philosophy of the research as outlined below.

This research project followed on from a pilot study led by the PI (2016b) which meant there was significant a priori. In view of this it was felt that saturation could have been achieved after only a relatively small number of interviews. However, the purpose of the study was not only to elicit information about participants' perceptions of late intervention for CLP, it was also intended to ascertain whether the TOM-clp could be used reliably as an outcome measure in a clinical setting and determine the barriers which caused the delay in receiving treatment. Also time limitation meant that the in depth interviews would all be undertaken in a two week period which would not have accommodated sequential analysis as required using an iterative process of data collection.

Accounting for this information, participants were selected by targeted, non-probability sampling. Accepting that there would be some redundancy a minimum target of 30 participants from each centre was selected, with a view to collecting larger volumes of TOM-clp data as part of the longitudinal process. Interviews were transcribed during the data collection period for the purpose of accuracy and familiarisation (Chapter 7), however, this also provided an opportunity to provisionally catalogue themes as the process progressed, which confirmed that it was unlikely that further interviews would have challenged or deepened the knowledge base.
6.6. Identification and recruitment of individual participants

6.6.1. Individuals with experience of CLP later in life

Inclusion criteria: Interested individuals who had experience of non-syndromic, untreated CLP beyond 6 years of age. Participants could choose whether or not to be accompanied by close friends or relations during the process for support and provision of additional information.

6.6.2. Identification

As mentioned in section 6.5 participants were identified through purposeful sampling. All participants were selected based on the criteria above ensuring a degree of homogeneity in the cohort and identified using opportunistic, convenience and targeted sampling as outlined below:

6.6.2.1. Opportunistic / convenience sampling

Participants potentially fitting the inclusion criteria were identified as they attended for routine appointments. These included individuals who had not yet undergone assessment or treatment for their CLP, individuals awaiting treatment and individuals who had already received late intervention.

6.6.2.2. Targeted sampling

This cohort included individuals fitting the inclusion criteria who were already known to the department and were awaiting for or had already received late intervention for CLP. Such patients were identified from clinical records, with the express permission from the organisation, and contacted by telephone by reception staff so that their follow up appointments coincided with periods of data collection.

6.6.2.3. Recruitment process

On first contact all potential participants were given a brief outline of the project and asked if they would like to consider becoming involved. It was explained that full clarification of the details would be given before making a final decision on
their involvement. It was also made clear that involvement was purely voluntary and that their decision, whether or not they chose to participate, would not affect their on-going treatment in any way and that they would be free to leave the project at any time without explanation or consequence.

Where possible patients were addressed in their native language however, in some circumstances, owing to the wide ranging ethnicity in some regions, it was necessary to employ an alternative language in which the patient was comfortable and reasonably fluent.

6.6.2.4. Consent

A member of the cleft team and/or the PI discussed the project in detail with all potential participants to ensure that they had a clear and comprehensive understanding of the nature of the research. Where possible, if there was an issue of language, an alternative member of the cleft team was recruited to act as an interpreter as outlined previously. Although consent forms and information leaflets had been produced in Hindi and English (Appendix 4, 5 and Appendix 6), it soon became apparent that the low levels of literacy and diverse ethnicity and spoken languages, did not allow sufficient understanding of the written form to ensure valid consent. This was consistent with the pilot research (Rees et al., 2016b) and a decision was made to provide the information in detail verbally for each participants through an interpreter, in order to maintain consistency and ensure fully informed verbal consent was achieved.

Participants were given the opportunity to discuss the implications of their participation prior to confirming consent to their involvement, a process similarly delivered verbally in the patients preferred language. The consent process for minors below 16 years of age was carried out in agreement with a parent/legal guardian.
6.6.3. Methods of recruitment for cleft team members for implementing the TOM-clp

As mentioned in section 6.2.1 a key worker was identified by the clinical director at each cleft centre for purposes of correspondence and co-ordination of the data collection. Initial contact was made with each key worker by the PI via e-mail, to discuss the projects aims and its implications in detail. Where multiple participants were to be involved the key worker then approached interested clinical and non-clinical members from within their cleft team and offered them the chance to participate. It was explained that their involvement would include instruction in the use the TOM-clp for use in data collection during regular cleft clinics. It was made clear that involvement was purely voluntary and that they would be free to leave the project at any time without explanation or consequence.
CHAPTER 7: PROCEDURE

7.1. Interviews with staff, health workers and administrators

Information relating to current departmental procedures was collected through direct face to face informal discussion with departmental managers, fund holders and members of the cleft team.

Where possible interviews were recorded with the express permission of the interviewee however, discussion was often developed spontaneously during clinic time for which written notes were taken during or immediately following the dialogue. Assurances was given that the information provided would be anonymised and used solely for the purposes of this study and related publications and that data would be securely stored and managed as outlined in section (7.2.3)

Interviews relating to aim 1 were semi-structured based on topics and questions derived from the literature review and previous research (Rees et al., 2016b) as outlined in Figure 6: Topic prompts relating to service provision. This format was selected in order to retain a level of consistency between interviews whilst not restricting information which would otherwise be missed in a structured questionnaire however free speech was encouraged and often provide additional information.
**Figure 6: Topic prompts relating to service provision**

<table>
<thead>
<tr>
<th>QUESTION PROMPTS FOR SERVICE PROVISION</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>How are the cleft services funded?</strong></td>
</tr>
<tr>
<td>▪ National government</td>
</tr>
<tr>
<td>▪ Local government</td>
</tr>
<tr>
<td>▪ NGO</td>
</tr>
<tr>
<td>▪ Private investors</td>
</tr>
<tr>
<td>▪ Out of pocket payments (OPPs)</td>
</tr>
<tr>
<td>▪ Other</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th><strong>What do you consider are the main reasons for late presentation of CLP?</strong></th>
</tr>
</thead>
</table>

<table>
<thead>
<tr>
<th><strong>What procedures are in place for education regarding CLP and the need for intervention?</strong></th>
</tr>
</thead>
<tbody>
<tr>
<td>▪ In schools</td>
</tr>
<tr>
<td>▪ Health care workers</td>
</tr>
<tr>
<td>▪ In the community</td>
</tr>
<tr>
<td>▪ Other</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th><strong>How are patients recruited?</strong></th>
</tr>
</thead>
<tbody>
<tr>
<td>▪ Referral by other hospitals / hospital departments</td>
</tr>
<tr>
<td>▪ Referral by community health workers</td>
</tr>
<tr>
<td>▪ Cleft camps</td>
</tr>
<tr>
<td>▪ Casual attendance</td>
</tr>
<tr>
<td>▪ Other</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th><strong>How are patients selected as appropriate for treatment?</strong></th>
</tr>
</thead>
<tbody>
<tr>
<td>▪ Age</td>
</tr>
<tr>
<td>▪ Health</td>
</tr>
<tr>
<td>▪ Type of cleft</td>
</tr>
<tr>
<td>▪ Available funding</td>
</tr>
<tr>
<td>▪ Other</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th><strong>Are there restrictions for people being able to receive intervention other than being medically unfit for surgery?</strong></th>
</tr>
</thead>
<tbody>
<tr>
<td>▪ Age</td>
</tr>
<tr>
<td>▪ Financial</td>
</tr>
<tr>
<td>▪ Social</td>
</tr>
<tr>
<td>▪ Complexity</td>
</tr>
</tbody>
</table>
What funds are available to assist in patients accessing treatment of clefts?
- Surgery
- Medications/dressings
- Transport
- Accommodation / Subsistence costs
- Follow up care
- Other

What allied services are available and are they funded / supported?
- Speech and language therapy
- Dental treatment
- Orthodontic therapy
- Psychology
- Other

How are services and outcomes audited?

What improvements would you strive to implement?

7.1.1. **Interviewing individuals with Cleft lip and/or palate, friends and family**

Interviews were facilitated in a quiet private area away from the surrounding activity of the regular clinic by a trained clinical or non clinical team member and/or the PI. Seating arrangements were informal and the atmosphere kept as relaxed as possible in order to avoid intimidation and encourage free speech. Where necessary interpreters, fluent in the most appropriate language of the participants' choosing, were selected from the clinical team ensuring that the interviews were conducted to maximise communication.

All interviews undertaken for the purpose of framework analysis were audio recorded, with the express consent of the interviewee, for the purpose of transcription and analysis. It was explained that the information would be used and stored solely for use of this study and that any direct quotes used in future publications would be anonymised. Recordings were carried out using an unobtrusive hand held digital recording device with an internal microphone rather than using a standalone microphone. It was accepted that whilst this may compromise the clarity of the recordings it would be less formal and less
intimidating than asking an individual to speak directly into a stand-alone microphone which may in turn have restricted free flow in conversation.

Recorded sessions started with introductions and an explanation of the purpose and proposed format of proceedings. Individuals were reminded that the process was being carried out for research purposes only, there was no obligation to participate and they could withdraw from the process at any time during the process.

Participants’ were encouraged to lead with open spontaneous discussion aimed at gathering qualitative data from participants and their families on the topics relating to their CLP. In order to overcome shyness and maintain direction, a list of prompts was used taking care to encourage free discussion whist not influencing proceedings with leading questions. This ensured that sessions were consistent, that all desired areas important to the research were covered and that the information was adequate to make an assessment for the TOM-clp. Following the initial pilot stage a number of additional demographic and topic questions were included in the prompts as they were considered appropriate and informative.

Previous work led by the PI (Rees et al., 2016b) had shown that recording the entire session for transcription proved ineffective. This was partly due to the number of different languages requiring translation but also due to the shyness and unintelligibility of many of the participants so a decision was made to record the English translations of the interpreter throughout the proceedings. Comments were therefore translated after each question and recorded by the PI making careful note of who had been speaking. This also had the advantage of allowing interjection and clarification of points by the PI as the session progressed.

At the close of conversation there was a short break whilst a summary of the interview data was undertaken by the PI. The pertinent points were then presented verbally to the participants so that they could confirm that it was an accurate picture of their views and opinions. They were then asked to contribute any further information they feel appropriate, a technique which frequently prompted further information that would not have been gleaned from a questionnaire or formal interview. This process continued to saturation point
providing confidence that further themes would be unlikely to emerge (Blackburn & Stokes, 2000). Despite this careful procedure participants often provided additional information as they were leaving and after the session was complete and recording had ceased. This information was documented in writing and stored in a field journal then later added to the transcription of the recorded session.

The initial intention was to ask all participants to complete a WHOQoL-BREF outcome questionnaire following the interview. This was intended to compare with the TOM-clp scores allocated by the PI based on the content of the interviews as a means of validation. However although the WHOQoL-BREF has many translations the only languages appropriate to India are English, Hindi and Tamil. Since these are not the primary languages spoken in Telangana it was not possible for the participants to complete questionnaires without the assistance of a translator. Time constraints within a busy clinical setting therefore restricted the use of the questionnaire to unpressured periods limiting its use for only a small number of individuals.

7.1.2. Guide for topics of discussion (Figure 7)

The listed areas of discussion were based on components from the WHO Classification of functioning, Disability and Health (ICF) and cover topics highlighted from previous research during the development of the TOM-clp (Rees et al., 2016a). This list acted as a guide only and was not considered fully comprehensive and limiting since individuals often had experiences which were not listed but none the less pertinent to the research.
Figure 7: Guide for topics of discussion based on the WHO-ICF

<table>
<thead>
<tr>
<th>ACTIVITIES AND PARTICIPATION</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Learning and applying knowledge</td>
<td>Access to education</td>
</tr>
<tr>
<td>General tasks and demands</td>
<td>General autonomy</td>
</tr>
<tr>
<td>Communication</td>
<td>Needs assistance communicating, using phone</td>
</tr>
<tr>
<td>Domestic life</td>
<td>Need of help shopping, catching local transport</td>
</tr>
<tr>
<td>Interpersonal relationships</td>
<td>Ability to make friends, marriage prospects</td>
</tr>
<tr>
<td>Self care</td>
<td>Eating and drinking, dental pain, appearance of teeth, ear infections, deafness</td>
</tr>
<tr>
<td>Major life areas</td>
<td>Employment, economic status</td>
</tr>
<tr>
<td>Community, social and civic life</td>
<td>Involvement in community activity, recreation and leisure, human rights, citizenship</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>WELL-BEING</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Attitudes of local community, educational establishments, employers, other employees</td>
<td></td>
</tr>
<tr>
<td>Attitudes of local community</td>
<td></td>
</tr>
<tr>
<td>Ability to make friends, family support</td>
<td></td>
</tr>
<tr>
<td>Level of anxiety, frustration, pleasure, happiness, sadness, loneliness, isolation, shyness</td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>QUESTIONS RELATING TO CLP</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Which aspects of the CLP is seen as the influencing factors</td>
<td></td>
</tr>
<tr>
<td>Experiences of cleft care including knowledge of service availability, funding possibilities and follow up care.</td>
<td></td>
</tr>
<tr>
<td>Experience of receiving cleft care - complications, access to services, time commitment, financial implications</td>
<td></td>
</tr>
<tr>
<td>Personal expectations of cleft services</td>
<td></td>
</tr>
<tr>
<td>Expectations following surgical intervention including functional, social financial and psychological factors</td>
<td></td>
</tr>
<tr>
<td>Overall satisfaction with intervention - positive or negative experiences</td>
<td></td>
</tr>
</tbody>
</table>
7.2. Data storage

The PI is sole custodian of the data

7.2.1. TOMs data

Numerical data from the TOM-clp has been stored in a secure password protected on-line data base. Individual identification could only be made from this data during the active research process in order to ensure that individuals could be contacted for purposes of follow up. Participants were fully informed of this and reassured that the information would be removed from the completed data set at which point data would be anonymous owing to use of allocated numerical identifiers for each individual.

Access to the data for analysis was undertaken by the PI using password protected electronic devices.

7.2.2. Data collection app

As mentioned in section 4.13, there are advantages and disadvantages of both paper and electronic data collection systems. For the purposes of this study, since data was to be collected in busy clinics with minimal resources, it was important to keep costs of data collection to a minimum both in terms of time and money.

Since all individuals collecting the data had access to mobile devices, phone, tablet or PC, a decision was made to collect data electronically. This avoided the expense of printing material and allowed demographic data to be collected alongside the TOMs data. Each question in the app was designed using a combination of dropdown lists, calendars, free text and a specifically designed TOM-clp score sheet. In order to ensure completeness, it was essential to complete each question before the next question could be accessed. The final page displayed a 'complete' icon which, once pressed, meant that the data was no longer accessible. This system was incorporated to ensure that every assessment was made on its own merit without comparison with previous recordings.
An important concern when downloading data is the risk of losing information when the internet connection is intermittent and unreliable. To overcome this, the app was designed to work offline and automatically test the strength of the signal before attempting to download. This allowed data to be collected in any area of the clinic, and would make it suitable for remote cleft camps, without risk of losing data. Once the app detected an adequate signal strength data would automatically downloaded into a password protected secure data base only accessible by the PI. Data was presented in the form of an spreadsheet to simplify management and analysis.

7.2.3. Storage, usage and disposal recorded media

It was clearly explained that any audio recordings and photographs would be used mainly as an aide memoire for the researcher. Where material was transcribed it was done so in an anonymous form and direct quotations are in used only with the express wishes of the participants. All participants were offered the chance to participate without being recorded if they wished to do so, however none declined.

Specific consent was taken from some individuals to allow the use of photographic images for the purposes of education and publication.

Recorded and electronic data was stored securely in encrypted files and that which was no longer required destroyed/deleted following completion of the research. Only material which is required for possible use in further research and publications related to this project and teaching purposes has been retained with the express consent of the participants concerned.
7.3. Implementing the TOM-clp

7.3.1. TOM-clp methodology

Training in the use of the Tom-clp was undertaken in accordance with Enderby et al. (2015) as outlined in chapter 8. In Assam two assessors from the centre were trained face-to-face by the PI during a field visit: A local SLT and an American nurse, who was working full time at the centre. The local dentist, who was also based full time at the centre, was trained later by the American nurse. The three assessors in Hyderabad were trained through email correspondence using the training manual.

Inter-rater reliability was confirmed by the PI based on the test case scenarios and ICC as outlined in the following chapter. Agreement was considered acceptable where the ICC was above .75 (Cicchetti, 1994) and the raters TOMs scores fell within one unit of the original test-scores documented in the pilot study (Enderby and John, 2015). Where the level of reliability fell below this a second training session was offered, had the participants not achieved they would not have been included but would have been offered another role in the study if they wished to continue their participation in the research. However in this case appropriate inter-rater reliability was achieved after a single training session for all participants trained by the PI, without the need for additional training and the participant trained by the local nurse only required one further training session before a suitable level of reliability was achieved.

Participant identification, recruitment and consultation process were undertaken using the same protocol as for the SSI outlined in chapters 5 and 6. Where the participant was attending a routine appointment with the SLT, or dentist the process was undertaken during the regular session in their particular clinic room, alternatively a quiet consulting room away from the busy clinic was suggested as a location for collecting data.

The TOM-clp scores and demographic data were not documented in the patient records but were automatically downloaded into a secure database via a specially designed app. This ensured data security as only the PI had access to the data once
it had been downloaded which guaranteed confidentiality and data protection. It also assured that results could not be altered nor could they be seen during subsequent appointments: This element of blinding was aimed at reducing empirical bias which may have occurred if an assessor doing a repeat measure or a different assessor at follow was influenced by the earlier score (Jüni et al., 2001, Schulz et al., 1995, Treweek et al., 2015). The app had been designed to work off-line and on-line so as to avoid losing data where internet connections were unpredictable and could be used easily on any hand held device.

7.3.2. Analysis of the TOM-clp data

7.3.2.1. Analysis of the TOM-clp data collected through via the app

The original intention had been to undertake longitudinal research in Assam, collecting pre-operative and post-operative information over an extended period prospectively. Despite excellent initial progress data collection ceased after only 10 weeks owing to the development of a political situation associated with the cleft centre and one of its founding NGOs. Although the clinical director was keen to continue participating in the research, circumstances dictated that the primary project lead could no longer work at the centre. Simultaneously another key practitioner withdrew from the project for personal reasons rendering the continuation of the project impracticable.

Whilst this period of data collection proved extremely useful in terms of assessing the viability of staff training, piloting the data collection app and implementation of the TOM-clp in a clinical setting, the resulting data for this period was all pre-operative. The data collected by the clinicians in Hyderabad was also prospective however, owing to a delay in initiating the project at this location, there was a limit to the number of cases recruited in the given time frame. A decision was therefore made to use the data collected via the app as a means of exploring the psychosocial impact of untreated CLP only so post-operative data was excluded at this stage. The pre-operative data from all four sites was amalgamated and analysed using descriptive and statistical methods.
7.3.2.2. Analysis of the TOM-clp data collected through interviews

In order to establish retrospective TOM-clp scores, the PI used information drawn from the interviews and case notes: Pre-operative impairment scores were allocated based on information from the patient surgical records. Post-operative impairment scores were based on the clinical opinion of the PI at the time of interview.

Scores for the activity, participation and well-being domains were based on information from the interviews which represented each participants personal perception of the impact of their CLP before and after intervention and were produced as follows: Short case scenarios were written for each participant based on the interview data; one pre-operative, one post-operative and, where participants had already undergone at least one secondary intervention at the centre, a mid-term scenario was also created. Examples of these can be seen in (Figure 8).

**Figure 8: Case scenarios for TOM-clp analysis**

<table>
<thead>
<tr>
<th>ID</th>
<th>Age</th>
<th>Scenario</th>
</tr>
</thead>
<tbody>
<tr>
<td>0.51272501</td>
<td>45</td>
<td>His family and friends can understand him because they are used to it but nobody else can really. He went to a special school when he was younger but even now he doesn't really get teased because people accept it and are happy to be friends with him. He does worry that he won't be able to get a job or get married and become independent though.</td>
</tr>
<tr>
<td>0.98812545</td>
<td>69</td>
<td>She can be understood most of the time and does not have a problem with regurgitation. She is generally confident, has friends and socialises well.</td>
</tr>
<tr>
<td>0.53311596</td>
<td>44</td>
<td>He has to repeat himself often but most people can understand what he is trying to say. He is reasonably confident, goes out with friends and has a job. He occasionally has problems with regurgitation.</td>
</tr>
</tbody>
</table>

Each scenario was anonymised and allocated an identifying number before being entered into a Microsoft® (MS)Excel 2007 spread sheet. The order of these were then mixed using the randomised function in excel so that no perceptible link could be made between any of the scenarios. Each scenario was then rated independently by three of the assessors who had previously been calibrated during the training manual pilot study. Since impairment was judged as an
objective score based on information seen only by the PI, this domain was not included in this process.

Once completed an average of the three assessors scores was taken for each scenario in each of the individual domains and corrected to one decimal point. These were then re-connected to the original participant testimony for analysis.

The randomisation process was introduced to reduce empirical bias which might have been apparent if the pre-operative and post-operative scenarios had been scored sequentially. The process of averaging means that the TOM-clp scores are represented by fractions rather than whole and half numbers as would be the case in a normal clinical situation.

The resulting data was analysed using descriptive and statistical analysis.

7.4. WHOQoL-Bref

It had been intended to implement the WHOQoL-Bref, with the participants in Hyderabad, as a comparative patient reported outcome measure. Unfortunately the questionnaire was only available in English and Hindi, neither of which are native to the Telangana region, so whilst both languages are commonly understood, the fluency of the participants was not adequate to complete the survey independently. Seven forms were completed with the assistance of an interpreter, however, this technique proved non viable owing to the excessive clinical time required to perform the translation task and was discontinued.

In addition, although many of the questions in the WHOQoL-BREF related for quality of life assessment for CLP; such as questions on bodily appearance, capacity to find work and access to health services, many of the questions were considered irrelevant despite using a culturally sensitive version. For example, questions such as 'Are you currently ill?', 'To what extent do you feel that pain prevents you from doing what you need to do?' 'do you have enough energy for everyday life?' may reflect how illness and physical disability could impede mobility and cause physical restrictions in performing tasks, but they do not represent difficulties associated with CLP, such as limitation in communication. Although it is acceptable
to leave irrelevant questions blank (World Health Organization, 1998), doing so left too few items in some domains to be considered comprehensive.

For these reasons a decision was made to exclude this element of the research and no further data was collected using the WHOQoL-BREF.

7.5. Choice of analysis for the semi-structured interviews

The purposes of the interviews in this study were threefold: Firstly, to discover the participants personal perspective of their experiences of CLP and impact of subsequent intervention without the constraints of rigid diagnostic categories. Secondly, to ascertain the reasons behind the delay in receiving treatment; Thirdly to elicit enough information to complete current and retrospective TOM-clp scores; The data for each of these three aspects were managed differently and the analysis process and results will be described in detail in the following chapters.

7.5.1. Patients perspectives

Qualitative interviews produce large volumes of textual data which, although rich and descriptive, cannot provide explanation without appropriate management (Pope et al., 2000, Miles and Huberman, 1994). The following section reflects on why framework analysis was selected as the most appropriate method for this study followed by a detailed description of how the technique was implemented.

Numerous diverse approaches have been developed for analysing qualitative interview data, many of which involve the process of thematic coding; identifying commonalities and differences in the data as a means of condensing the raw data into a more manageable format for analysis (Morse and Richards, 2002, Smith and Firth, 2011, Gibbs, 2007, Gale et al., 2013). Techniques vary from descriptive analysis reports which document an individual’s experience to interpretive analysis which seeks to generate an explanation behind the phenomena under investigation (Attride-Stirling, 2001, Braun and Clarke, 2006, Tesch, 2013, Sandelowski and Barroso, 2002, Sandelowski and Barroso, 2003).
Thematic coding forms the basis of methods and methodologies which can broadly be divided into two groups; those pertaining to a particular epistemological stance and those which are applicable across a range of methodologies independent of specific philosophical theories (Braun and Clarke, 2006, Crotty, 1998). Selecting the most appropriate method depends on the aims and requirements of each individual study, however in general thematic coding techniques share a common set of guiding principles:

- Collection of data through interviews and/or observations
- Transcription of the interviews and documentation of observations
- Familiarisation with the data through immersion to gain an insight into the phenomena
- Categorisation and coding to link overarching themes

As this research takes a pragmatic approach, analytical methods which stem from a particular paradigm were not considered appropriate. However, the rational for rejection of some methods will be summarised briefly although the list is far from exhaustive. Owing to the confusion in the use of the term thematic analysis as either a specific methodology or as a general term for theme based analysis (Hancock et al., 1998) the term will not be employed in this discussion.

Grounded theory relies on thematic coding but was rejected as it takes a constructivist approach and its application is limited to research without pre-existing knowledge or supposition (Glaser and Strauss, 1967, Straus and Corbin, 1998). Interpretive phenomenological analysis (IPA) had potential as it aims to understand the connotations of individuals experiences to a particular phenomenon, however the idiographic requires in depth analysis of interviews taken from homogeneous samples (Smith et al., 2009, Larkin and Thompson, 2012, Biggerstaff and Thompson, 2008). Although there was a level of uniformity in the population studied in this research, in that all the participants have experience of untreated CLP, the mixed demographic in age, ethnicity and geographic location meant the data set was not truly homogeneous. Also, since the study involved 49 interviews, these would have been too numerous to have applied the depth of analysis required for IPA. By a similar token the underpinning philosophy and number of interviews precluded the implementation of narrative analysis which is
biographical and aimed at analysis of individual case studies rather than multiple interviews (Crossley, 2000, Murray, 2015, Riessman, 1993, Greenhalgh et al., 2005).

Framework analysis provides a means of systematically mapping, reducing and categorising large quantities of qualitative data but is not restricted by any specific epistemological position (Gale et al., 2013, Ward et al., 2013, Swallow et al., 2011). It was originally designed for use for large scale policy research where the object for investigation was typically set in advance of the research (Gale et al., 2013, Ritchie et al., 2013, Ritchie, 1994) which makes it particularly suited for this project where there is a priori and where the aim is to obtain a descriptive overview of large but relatively homogeneous data set (Gale et al., 2013).

A number of thematic techniques have been criticised for fragmenting the data and reducing transparency which diminishes vigour as the original context often becomes lost in the analysis (Attride-Stirling, 2001, Furber et al., 2009, Dixon-Woods, 2011). The matrix construct of framework analysis avoids fragmentation and ensures that the links between the original data are represented throughout the analysis process giving the researcher the opportunity to re-evaluate and refine themes repeatedly by means of cross-referencing (Furber et al., 2009). In this way data can be examined both by case (seeing what each person did) and theme (relating what people did) but also in combination whilst maintaining a connection with each participant’s individual perspectives. This format not only facilitates the researcher in recognising differences and commonalities within the data set and identifying relationships which run through it, but also offers a strong level of transparency and ensures that it is the participants’ experiences which ultimately drive the research (Ritchie et al., 2013, Braun and Clarke, 2006, Pope et al., 2000).

For qualitative projects where there is a priori it may be possible to anticipate core themes from existing literature and experiences of the researcher prior to data collection and analysis. This can be valuable in structuring the analysis, ensuring that aims of the study remain embedded in the framework so reducing the risk of losing sight of the original research question (Miles and Huberman, 1994). However, it is important to maintain an open mind whilst reviewing the data and
allow new themes to emerge which may reflect the participants’ account more accurately (Sokolowski, 2000, Starks and Brown Trinidad, 2007). Framework analysis promotes diversity by encouraging the emergence of new ideas which reflect the views of the participants but also embraces the incorporation of pre-determined codes as a malleable starting point.

The open matrix format of framework analysis means that information can be visualised easily which allows systematic and comprehensive question-focussed analysis whilst keeping the data ordered and unambiguous to avoid bias (Dixon-Woods, 2011). This level of clarity, transparency and flexibility advocated framework analysis as the preferred model to answer the research questions in this study.
7.6. FRAMEWORK ANALYSIS PROCESS

Ritchie and Spencer (1994) outline five stages in framework analysis: Transcription and familiarisation, identifying a thematic framework, indexing, charting and mapping and interpretation. Although described sequentially the process implemented was dynamic and the researchers moved back and forth between stages as a means of clarification and incorporation of new information as the analysis progressed (Green et al., 2007).

7.6.1. Transcription and familiarisation

7.6.1.1. Familiarisation

Braun and Clarke (2006) describe the familiarisation process as 'the bedrock of the analysis' and it is considered an essential aspect of qualitative research for developing an initial understanding of the data and creating provisional ideas for future coding (Ritchie, 1994, Ward et al., 2013, Hunter et al., 2002). Since the primary investigator (PI) had been personally involved in all the interviews there was already a degree of prior knowledge before the transcription process began, however familiarisation continued through a process of transcription, re-reading, re-listening, note taking and correction (Braun and Clarke, 2006, Riessman, 1993).

7.6.1.2. Transcription

Good transcription demands that interviews are accurately represented and recorded consistently to the level appropriate to answer the research questions (McLellan et al., 2003, Braun and Clarke, 2006). Methods vary in precision levels along a continuum of two extremes: naturalised transcription, which aims to recreate the dialogue as accurately as possible using verbatim transcription, intonation, grammar, non verbal sounds and even non-linguistic observations such as facial expressions and mannerisms (Schegloff, 1997, McLellan et al., 2003); de-naturalised transcription which focuses on the sense of the dialogue with greater emphasis on the overall meaning that the detail of the structure and manner in which the interview ensued (Oliver et al., 2005). The former is intended for studies involving an in depth understanding of the beliefs and attitudes of an individual or small group exploring the content, terminology and manner in which
participants’ express their views (Oliver et al., 2005). The latter was considered more appropriate for this study which was more interested in identifying generalised patterns and salient themes of a larger population (Boyatzis, 1998, Braun and Clarke, 2006).

Since the recording for transcription in this study generally comprised an overview of the conversation provided by the interpreter, rather than the spoken words of the interviewee, any associated nuances and inflections would not have related to the participant directly thus not relevant in the analysis. However it is acknowledged that the manner in which something is said and a person’s demeanour may impart as much information as the words used to say it particularly in relation to their emotional state (Poland and Pederson, 1998). To retain this component of the conversation for analysis, field notes on the subject were taken by the PI at the time of the interview, written up and linked to the individuals’ transcriptions for cross-reference in the analysis (Arthur and Nazroo, 2003, Ward et al., 2013).

Kvale (2006) recommends that it is important to have multiple transcribers and compare the transcripts for accuracy however to ensure consistency others recommend that accuracy of audiotape transcription is increased using a single researcher who is familiar with the research topic, the transcription protocol and the terminology (McLellan et al., 2003, MacQueen and Milstein, 1999). For the purpose of this study all the transcription was undertaken by the PI using Microsoft® (MS) Word, the first draft of each being completed within 24 hours of the interview to retain a level of familiarity. This proved particularly valuable where the interviews had been quiet, lacked clarity or had been backed up by numerous field notes to support less intelligible interviewees.

Each recording was linked to a patient identifier which logged the participant’s profile (age, gender, demographics, cleft type, surgeries), field observations and any additional documented comments which had been made by the participants outside of the recording process. Re-listening was undertaken whilst proof reading the transcription to correct any inaccuracies. During the initial transcription headphones were not available so the recordings were revisited later and further
corrections made with the benefit and clarity of headphones. Where sections still remained inaudible or unclear this was recorded in the transcription notes.

The transcription protocol selected chose to exclude lengths of pauses, inflections as in discourse or conversational analysis however, punctuation was implemented to help retain meaning (Hancock et al., 1998). The PI concentrated on content of what was said but included all audible words including 'ums' and 'ers' so as to remain faithful to the original recording. Although Sandelowski (1994) suggest that transcribing fillers is not greatly beneficial regarding content, they were included in this study as indicators of the fluency or difficulty in finding the appropriate word by the translator which could possibly have affected the meaning of the phrase. Potential ambiguities were clarified in brackets when the subject of the phrase was only inferred rather than specified in the conversation. Data was transcribed verbatim but all identifying information such as names of hospitals and professionals was omitted from the transcripts.

Gale et al. (2013) suggest that during the familiarisation period it can be useful to make analytical notes which document thoughts and impressions emerging during the process. In this case a short profile was written summarising the points made during the interview and the demographic information relating to each participant. This enhanced familiarisation and also facilitated the provision of current and retrospective TOM-clp scores.

Once the transcriptions were complete the interviews were examined line by line and each pertinent sentence, phrase or paraphrase was cut and pasted into a MS Excel spreadsheet. Only comments which did not relate to a patients perspective such as "They have only had x-ray now the treatment is not beginning yet" were excluded, although the information was incorporated into the profile summary. If the authentic significance of each phrase was not perceivable whilst being read out of context bracketed clarifiers were included for example "[before treatment]..whatever they eaten they get regurgitation [after treatment] now it has improved ma'am". By remaining faithful to the transcript and using the participants own words it ensured that the coding remained true to the original data reducing the risk of subjective interpretation being introduced by the researcher (Ritchie et al., 2013).
A total of 381 participant phrases were documented and additional two comments were included marked 'other'. These were comments made by one of the key workers during an interview and included as they corroborated and emphasised the importance of a statement made by a participant. This initially resulted in a list of 383 phrases entered into a MS Excel spreadsheet for the coding process, however late on in the process it was realised that two of the phrases were actually repeated in the list. Since numerical identifiers had already been allocated to each line in the spreadsheet the replicated terms were replaced by the term "repeat phrase" to avoid confusion in the analysis. The final analysis was therefore undertaken based on 381 phrases.

### 7.6.2. Developing a working analytical framework

The next stage was a taxonomic process comprising reading the transcribed phrases in detail and assigning a label or 'code' which summarise important elements of the transcript (Gale et al., 2013, Green et al., 2007, Miles and Huberman, 1994, Cope, 2009). Saldaña defines a code in qualitative research as ‘a word or short phrase that symbolically assigns a summative, salient, essence-capturing, and/or evocative attribute for a portion of language-based or visual data’ (Saldaña, 2015)p3. Allocating such codes is a subjective process and will vary according to the requirements of the particular study and the preference of the researchers.

Researchers can employ 'open coding' where codes emerge from the information using an inductive process (Glaser and Strauss, 1967) or codes may be pre-defined so the process is deductive in nature based on a pre-existing theory according to the subject under investigation (Miles and Huberman, 1994). Since the researcher is already assimilating the information they are exposed to, whether consciously or sub-consciously, during the development of the research, a truly inductive process is rarely possible (Pope et al., 2000). An alternative is an integrated approach using both inductive and deductive thinking to develop a code list or 'code dictionary' (Bradley et al., 2007).

Selection of codes will depend on the aims of the study; they may relate to tangible processes such as specific activities or behaviours or they might relate to
indications of emotions, specific values which a participant might attribute to an incident or representations of particular settings or environments associated with the script (Gale et al., 2013, Pope et al., 2000, Patton, 1990, Lofland and Lofland, 2006). What is important is that a code is assigned to all aspects of the information facilitating the systematic comparison of the entire data set and where appropriate more than one code may apply to an individual phrase (Green et al., 2007).

Some experts suggest that it is sufficient for a single researcher to develop all the codes from the data in a given project (Morse and Richards, 2002) however it is more commonly recommended that this part of the coding process is undertaken independently by at least two researchers to avoid the opinions and idiosyncrasies of one individual dominating the outcome (Green et al., 2007). Looking at the data from more than one perspective also increases the chance of identifying unexpected outlooks which may be missed particularly where there is a priori (Gale et al., 2013). If multiple coders are employed it is important that codes are used consistently and that their attributed meaning is agreed and applied appropriately throughout (Green et al., 2007). The involvement of researchers with a different range of knowledge and experience also introduces a level or richness and rigor to the process which can be particularly useful when considering outliers and scrutinizing apparent deviation within the data (Bloor, 1978).

The coding process for this study was undertaken by two independent researchers collaborating to produce a preliminary coding protocol and codebook defining each of the codes. The PI acted as one coder, bringing a significant amount of a priori from a previous pilot study, a detailed literature review, designing the research project and implementing and transcribing the interviews. The second coder was an academic colleague without previous experience of cleft or TOMs, recruited in order to maximise the inductive element of the process and counteract unintentional bias from the PI. With this intention, the second coder was not privy to potential pre-assigned codes or themes and was unaware of the structure of the prompt sheet until after the first phase of the coding process.
7.6.3. Initial cycle of coding

The first fifty phrases were cut and pasted into a MS Word document and scrutinised by the two coders independently. Each coder worked alone and attributed a descriptor or series or descriptors to each phrase in adjacent columns. The documents were then compared and discussed to identify differences and commonalities which could lead to the construction of a preliminary coding protocol.

The results of the initial coding cycle, outlined in Table 7:1 (details of all 50 phrases can be seen in Appendix 11) displayed good general agreement with only one of the phrases (49) lacking any consistency. All other phrases demonstrated agreement on at least one of the descriptive labels (Saldaña, 2015) with disagreement relating largely to terminology rather than overall concepts.

Percentage agreement was 84% for individual codes, using the total number of agreements as a percentage of all suggested codes (Miles and Huberman, 1994). However, looking at the codes, this does not represent a disagreement of 16% rather it indicates 16% of additional ideas and new concepts.

Both reviewers had used simultaneous coding in all but five cases, finding that more than one code was attributable to a particular phrase which held a number of inferences (Saldaña, 2015). This meant that as well as the points of agreement both reviewers independently introduced new concepts: The 2nd reviewer highlighted the element of parental concern and suggested ADL could be used as an overarching theme to cover topics such as shopping, travel and using the phone. The PI recognised elements which related to medical or surgical aspects of CLP and introduced the concept of ‘well-being’ and ‘participation’ as pre-determined themes. These terms are two of the four domains of the TOM and WHO-ICF which had shown to be useful overarching expressions which represented many of the topics covered in the pilot project (Rees et al., 2016b). A third domain ‘activity’ was also discussed but did not appear to add value at this stage of coding but was kept in mind for introduction at the next stage in the cycle.
<table>
<thead>
<tr>
<th>ID</th>
<th>Quote</th>
<th>Coding from 2nd reviewer</th>
<th>Coding from PI</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Ch01 [father] then can’t understand [but] now it is better</td>
<td>Being understood</td>
<td>Parental concern</td>
</tr>
<tr>
<td>2</td>
<td>Ch01 [whether it could be fixed] that they didn’t know, the parents are uneducated we don’t know</td>
<td>Understanding of CLP</td>
<td>Family social circumstances</td>
</tr>
<tr>
<td>3</td>
<td>Ch01 [finding out about repair] i was in Hyderabad and i heard about it with advertisement</td>
<td>Understanding of CLP</td>
<td>Learning about CLP</td>
</tr>
<tr>
<td>4</td>
<td>Ch01 She had some small of holes there in palate so now it was clear [speech]</td>
<td>Understanding of CLP</td>
<td>Being understood</td>
</tr>
<tr>
<td>5</td>
<td>Ch01 [father] she has faced a lot of problems in the communication</td>
<td>Being understood</td>
<td>Socialising</td>
</tr>
<tr>
<td>6</td>
<td>Ch01 They won’t be able to understand and make with other friends so she will face that problem</td>
<td>Being understood</td>
<td>Marginalisation/socialising</td>
</tr>
<tr>
<td>7</td>
<td>Ch01 [father] now is ok, speech is much better</td>
<td>Speech</td>
<td>Being understood</td>
</tr>
<tr>
<td>8</td>
<td>Ch01 Now all are able to understand what she is speaking</td>
<td>Being understood</td>
<td>Socialising</td>
</tr>
<tr>
<td>9</td>
<td>Ch01 So now shopping and things it’s made life easier</td>
<td>ADL [activity of daily living]</td>
<td>Life getting better</td>
</tr>
<tr>
<td>10</td>
<td>Ch01 Did you know anyone with CLP before? No ma’am</td>
<td>Understanding of CLP</td>
<td>Life getting better</td>
</tr>
<tr>
<td>11</td>
<td>Ch01 So now shopping and things it’s made life easier</td>
<td>ADL</td>
<td>Life getting better</td>
</tr>
<tr>
<td>12</td>
<td>Ch01 They won’t allow her go on her own, not yet [cultural not because of cleft]</td>
<td>Socialising but ? Relevant as not cleft but cultural</td>
<td>Cultural</td>
</tr>
</tbody>
</table>
The pilot project and the content of the prompt sheet had led the PI to expect that individuals would comment on the reasons for not having received earlier intervention and this was corroborated in the second phrase. However, since the specific reasons behind treatment being delayed had already been addressed separately, a decision was made to merge these codes under 'knowledge and awareness'.

It was agreed that phrase 49 was slightly ambiguous in nature and both coders admitted to applying a bit of speculation as to the exact meaning, which may have accounted for the divergent coding. However since it did not introduce any unexpected or outlying codes it was not considered further at this stage.

It was noticeable that the second reviewer implemented in vivo coding (Saldaña, 2015), using the exact words in the text, when describing different emotions for example: I can't talk in any language, and fear and hesitation, fear, fear, very much fear. It was agreed that this system should be employed throughout as a means of sub-coding so as not to lose meaning from the original text.

A list of proposed codes was drawn up and grouped into categories using a tree diagram which acted as a provisional working framework (Gale et al., 2013), The terms were compared and discussed until agreement was achieved on the terminology and coding protocol to employ. A draft coding dictionary was then created listing the definitive codes and their defined meanings to help ensure that a consistent coding protocol was used for the remainder of the process (Table 7:2) The use of a codebook was considered valuable not only as a means of improving reliability by reducing subjectivity in the coding but also to make the process replicable with a greater level of accuracy (Rourke et al., 2007, Coffey and Atkinson, 1996, DeCuir-Gunby et al., 2011).
<table>
<thead>
<tr>
<th>Codes from 2nd reviewer</th>
<th>Codes from PI</th>
<th>Provisional coding</th>
<th>Sub-codes</th>
<th>Provisional codebook</th>
</tr>
</thead>
<tbody>
<tr>
<td>Being understood</td>
<td>Communication</td>
<td>Communication</td>
<td>-ve</td>
<td>Not being understood, unable to pronounce words properly, unwilling to speak,</td>
</tr>
<tr>
<td>Speech</td>
<td></td>
<td></td>
<td>+ve</td>
<td>Being understood, able to pronounce words properly, willing to speak,</td>
</tr>
<tr>
<td>Parental concern</td>
<td>Affect of CLP on family</td>
<td>Family impact (emotional and financial)</td>
<td>Financial</td>
<td>Costs involved in travel to appointments, loss of earnings, cost of surgery</td>
</tr>
<tr>
<td>Family finance</td>
<td>Financial</td>
<td></td>
<td></td>
<td>Family emotions relating to child’s CLP</td>
</tr>
<tr>
<td>Family social circumstances</td>
<td>Low educational status</td>
<td>Family background</td>
<td></td>
<td>Social status and family circumstances</td>
</tr>
<tr>
<td>Learning about CLP</td>
<td>(No) Knowledge of CLP</td>
<td>Knowledge about CLP</td>
<td></td>
<td>Why intervention was not sought earlier</td>
</tr>
<tr>
<td>Understanding CLP</td>
<td>(not) Getting CLP repaired</td>
<td></td>
<td></td>
<td>Parents understanding of why the CLP developed</td>
</tr>
<tr>
<td>Socialising</td>
<td>Participation</td>
<td>Participation (theme)</td>
<td>Inclusion</td>
<td>Being accepted and included into society (or not)</td>
</tr>
<tr>
<td>Marginalisation</td>
<td></td>
<td></td>
<td>Making friends</td>
<td>Ability to make friends or not</td>
</tr>
<tr>
<td>Making friends</td>
<td>Ability to make friends or not</td>
<td></td>
<td>ADL</td>
<td>Comments relating to activities such as Using the phone, shopping, travelling</td>
</tr>
<tr>
<td>Activity of daily living (ADL)</td>
<td>Reaction of outsiders</td>
<td>Teasing</td>
<td>Reaction of others</td>
<td>Ridiculing</td>
</tr>
<tr>
<td>Reaction of outsiders</td>
<td></td>
<td></td>
<td>Teasing</td>
<td>Examples of emotional responses due to CLP</td>
</tr>
<tr>
<td>Attitude of others</td>
<td>Daily life improved</td>
<td>Quality of life</td>
<td></td>
<td>Indications of fulfilment and contentment with life (or not)</td>
</tr>
<tr>
<td>Life getting better</td>
<td>Autonomy</td>
<td>Autonomy</td>
<td></td>
<td>Indicates independent living</td>
</tr>
<tr>
<td>Category</td>
<td>Well-being as a theme not code</td>
<td>Information regarding work or ability (inability) to find work</td>
<td></td>
<td></td>
</tr>
<tr>
<td>-------------------------</td>
<td>-------------------------------</td>
<td>---------------------------------------------------------------</td>
<td></td>
<td></td>
</tr>
<tr>
<td>(not) Finding work</td>
<td>Employment</td>
<td>Indicates involvement in school or higher education</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Education</td>
<td>Education</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Emotion</td>
<td>Emotions</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Emotion</td>
<td>Emotions</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Frustration</td>
<td>Frustration</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Shy</td>
<td>Shy</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Confidence</td>
<td>Self confidence</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Happy</td>
<td>Happy</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Expectation</td>
<td>Expectation</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Fear</td>
<td>Fear</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Self respect</td>
<td>Self respect</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hesitance</td>
<td>Hesitance</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Regurgitation</td>
<td>Regurgitation</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Surgical</td>
<td>Surgical</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Well-being as a theme not code

- Finding work: Information regarding work or ability (inability) to find work
- Education: Indicates involvement in school or higher education

Emotions

- Frustration
- Shy
- Confidence
- Happy
- Expectation
- Fear
- Self respect
- Hesitance

Regurgitation

- Nasal regurgitation, food packing, difficulty eating and drinking

Surgical

- Comments relating to surgical aspects of cleft
Once coding consensus had been reached for the first 50 phrases the process was repeated for the remaining 333 phrases with potential codes once again being compared using a tree diagram. During this phase coding was fully inclusive and a new code was introduced for each particular topic or significant word identified (Gale et al., 2013) which generated a long list which would ultimately be clustered together into defined categories and then further grouped into still broader themes. This process continued until both reviewers were satisfied that they had reached theoretical saturation and no new concepts were likely to emerge (Glaser and Strauss, 1967, Patton, 2002), at which point the final working code framework was constructed.

Deciding when saturation point is reached is not always straightforward (Fusch and Ness, 2015) however in this case all the codes appeared numerous times in the data and appeared to cover all aspects of the predetermined themes. This led the PI to the conclude that it was unlikely there were any clear conceptual gaps in the data (Bradley et al., 2007) and it was improbable that any new information would have come to light by increasing the number of interviews (Guest et al., 2006). It was considered important to reach saturation not only for completeness of analysis but also to ensure replicability (Walker, 2012, O'reilly and Parker, 2013).

An open coding technique was used in order not to miss meanings which might have become lost if the group had been clustered into a single group too soon. For example, 25 different in vivo sub-codes were identified signifying emotions all of which potentially held different connotations. As a second cycle coding exercise, these sub-codes were reconsidered in context by cross-referencing the original script allowing terms with similar connotations or synonyms to be merged. This condensation process is summarised below (Table 7:3) and resulted in 12 final sub-codes nine with negative connotations and three positive.
Table 7:3 Condensation of sub-codes for emotion

<table>
<thead>
<tr>
<th>Categories</th>
<th>Initial coding</th>
<th>Final coding</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Negative emotions</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Embarrassing</td>
<td>Embarrassment</td>
<td></td>
</tr>
<tr>
<td>Unhappy</td>
<td>Upset</td>
<td></td>
</tr>
<tr>
<td>Upset</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Shy</td>
<td>Shy</td>
<td></td>
</tr>
<tr>
<td>Self-conscious</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hiding</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Reluctant</td>
<td>Tentative</td>
<td></td>
</tr>
<tr>
<td>Hesitance</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Afraid</td>
<td>Fear</td>
<td></td>
</tr>
<tr>
<td>Fear</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Shame</td>
<td>Humiliation</td>
<td></td>
</tr>
<tr>
<td>Low self esteem</td>
<td>Low self esteem</td>
<td></td>
</tr>
<tr>
<td>Concern</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Not comfortable</td>
<td>Worry</td>
<td></td>
</tr>
<tr>
<td>Worry</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Troubled</td>
<td>Aggression</td>
<td></td>
</tr>
<tr>
<td>Aggression</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Positive emotions</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Confident</td>
<td>Confidence</td>
<td></td>
</tr>
<tr>
<td>Boldness</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Comfortable</td>
<td></td>
<td></td>
</tr>
<tr>
<td>No fear</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Happy</td>
<td>Happy</td>
<td></td>
</tr>
<tr>
<td>Good</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Accepting</td>
<td>Expectation</td>
<td></td>
</tr>
<tr>
<td>Expectation</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

A similar system was used for the 'opinions of others' and in doing this it was identified that the term 'Teasing' was used both in a negative context and also as a positive where teasing had either never occurred or reduced. Had this second cycle been omitted this may have been overlooked resulting in the coding system misrepresenting the participant’s original meaning. As a result the code was split to represent either negative or positive reactions from others.
Apart from the additional in vivo emotional codes, 17 entirely new codes emerged from assessment of the additional phrases:

- It was decided that 'Knowledge about CLP' was too broad a term so the team introduced the term Gaining CLP knowledge as a code.
- The impact of time to travel to appointments was noted and included as a code under Family background, as was a reference to having a relative with CLP.
- Ear infections, dental problems and difficulty breathing were introduced under the surgical category but a decision was made to rename it 'medical' and include the surgical aspect under failed procedures.
- Marriage was introduced and linked with making friends under the category of interpersonal relationships.
- Change in role was added with autonomy along with 2 sub-codes; reliance on others, independence, inclusion and living life to the full.
- 4 new sub-codes were introduced under reaction of others; criticism, acceptance, questioning and support.
- Appearance was identified by both reviewers and introduced as a theme

Having made the decision to subdivide 'emotions' and 'influence of others' into negative and positive connotations other themes were then looked at and it was decided that meaning would only be retained if a similar division was made.

Consequently the categories of 'appearance', 'communication', 'regurgitation', 'ADL', 'mixing with friends' and 'work and education' were all partitioned to demonstrate negative and positive aspects. It was felt that these subdivisions would give more insight into how the CLP impacted the lives of the participants.

All of the code words which had been generated in the first and second cycle were listed and those showing similarities were grouped together in categories which were then grouped into overarching themes (Iliffe et al., 2015, Gale et al., 2013).

During this phase additional changes were made in relation to terminology:

- The term Access to work and education and domestic life were selected from the interview guide as preferred terms to replace employment, education and ADL.
• Interpersonal relationships was introduced as an overarching term for mixing with friends and marriage:
• The phrase Self care was taken from the WHO-ICF to encompass medical and regurgitation.

Refinements continued until a consensus was reached as to the most fitting analytical framework to best represent the core concepts within the data and address the research questions. By using an integrated approach the code structure was developed with a combination of bottom up and top down approach therefore minimising the risk of the PI subconsciously forcing the data into predetermined categories (Bradley et al., 2007). Reference was made back to the original transcripts throughout this process to ensure that the true meanings and implications were accurately represented.

Finally two of the domains of the TOM’s 'activity' and 'participation' were discussed and introduced as themes in conjunction with family background and identity. 'Well-being', a third domain of the TOMs, was considered to have a significant overlap across the other themes apart from 'knowledge about CLP'. Therefore the working framework was completed by encompassing 95% of the other themes under this heading as all were considered as components of 'well-being' as summarised in Table:7:4.

The terms in the framework were then added to the code book and allocated abbreviations so they could be used easily in the indexing process as a catalogue of salient terms to preserve the context of the transcripts without needing to write out the code in its entirety (Bradley et al., 2007).

Reflective practices such as consultation with other colleagues and writing memos were employed throughout the coding process as a means of engaging with the data and tracking how ideas evolved (Cutcliffe, 2003, Starks and Brown Trinidad, 2007). This process was considered essential in providing transparency by means of an audit trail to record how the analytical process moulded the understanding of the initial research question (Cutcliffe, 2003). This was considered particularly important as the PI was acutely aware of the possibility that prior knowledge of the subject and intense involvement in the data collection and analysis would

7.6.4. Confirming reliability of the code structure

Bradley et al. (2007) suggest that using a single researcher can be a more time efficient and consistent method of applying the finalized code structure than using multiple coders so it was decided that the indexing process would be undertaken by the PI alone. However for purposes of testing the code structure it was decided to test the inter-reliability using a small sample of the transcript texts.

A selection of twenty phrases were selected using the randomisation feature in MS Excel and coded independently by both the PI and a reviewer fresh to the initial coding process. For this purpose a percentage agreement of over 80% has been deemed as acceptable as a rule of thumb (Bradley et al., 2007, Miles and Huberman, 1994) in this case Inter-rater reliability was established at 86%. It was interesting to note that there was no disagreement in the codes selected however the PI included a greater number of codes which relied on inference from the text. For example in the phrase 'now everybody getting understanding [speech] and he is good life, big differences are there' There is an inference that the participant's speech had been bad before but now it was better which was utilised by the PI. On discussion it was also felt that the level of prior immersion the PI had experienced with the transcripts made these inferences more noticeable owing to a prior knowledge of the conversations in context.

As a result of this process it was concluded that the coding framework was reliable and that coding based on inference would be included in the protocol.
Table:7.4: Final working code framework

<table>
<thead>
<tr>
<th>Themes</th>
<th>Categories</th>
<th>Codes</th>
<th>Sub codes</th>
</tr>
</thead>
<tbody>
<tr>
<td>Family Background</td>
<td>Knowledge about CLP</td>
<td>Why treatment was late</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>Why clefting occurred</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>Gaining CLP knowledge</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>Other relative with CLP</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Family impact</td>
<td>Financial commitment</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>Time commitment</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>Emotion about CLP</td>
<td></td>
</tr>
<tr>
<td>Activity</td>
<td>Communication</td>
<td></td>
<td>Communication -ve</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Communication +ve</td>
</tr>
<tr>
<td>Self care</td>
<td>Medical</td>
<td>Ear problems</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>Difficulty breathing</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>Failed procedures</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>Dental problems</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Regurgitation</td>
<td>Regurgitation -ve</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>Regurgitation +ve</td>
<td></td>
</tr>
<tr>
<td>Well Being</td>
<td>Domestic life</td>
<td>Shopping, Using a phone.</td>
<td>Negative</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Travelling</td>
<td>Positive</td>
</tr>
<tr>
<td></td>
<td>Influence of society /</td>
<td>Negative impact</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Opinion of others</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>Positive impact</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Interpersonal relationships</td>
<td>Mixing with friends</td>
<td>Negative</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Marriage</td>
<td>Positive</td>
</tr>
<tr>
<td></td>
<td>Role in society</td>
<td>Access to work and education</td>
<td>Negative</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Positive</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Autonomy / Change in role</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Emotions</td>
<td>Negative emotions</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>Positive emotions</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Appearance</td>
<td>Appearance -ve</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>Appearance +ve</td>
<td></td>
</tr>
</tbody>
</table>
7.7. Applying the Analytical framework

The analytical framework was applied to all the transcripts as a process of indexing; applying one or more codes to all the phrases listed from the transcripts and annotating the information in a spreadsheet (Gale et al., 2013, Ward et al., 2013). Decisions were made based on significance to individual codes in the text or inference to their relevance as mentioned previously. Since the code framework had been developed using representation from the entire data set and discussed previously no changes in the coding system were needed during this stage.

Since the dataset was considered of a manageable size the process was undertaken using MS Excel rather than implementing a computer aided qualitative software package. It is recognised that software packages can be extremely beneficial in the storage and management of large and unwieldy datasets and are particularly useful in cross indexing the information (Pope et al., 2000, Leech and Onwuegbuzie, 2007), however, although some qualitative studies claim that the implementation of software increase the ‘power’ of their research they have the disadvantage of increased cost and can lead the researcher to lose contact and intimacy with their data. In addition, as with many 'time saving’ computer systems, software analysis packages may not always prove as judicious and efficient as anticipated (Lee and Fielding, 1995). In addition, since MS Excel is commonly available it made the sharing of information more straightforward for all involved (Swallow et al., 2003).

The simplicity of the process was enhanced using multiple linked screens, split screens and cut and paste facilities in conjunction with hand written notes and analytical memos.

7.7.1. Charting and mapping

The creation of a thematic matrix was also undertaken using MS Excel: The process involved allocating an individual row to each of the participants involved in the interview process. Participants were identifiable only by their unique ID number not including individuals who had not fitted the inclusion criteria. Associated
demographic data and summarised coded data were then logged in corresponding columns using the pre-assigned codes.

Indexing was completed for all the transcribed data, allowing information to be analysed across the entire data set without losing the connection or context of individual participants’ viewpoints. In order to simplify the task of cross-referencing back to the original text each annotated code was linked to a pop-up window showing its associated phrase (Figure 9).

The content was then scrutinised to identify patterns and connections within the data. Properties of MS Excel such as randomisation and filtering were used to enhance this mapping process: Randomisation was used to mix the order of the participants to be sure that information was not masked by the ordering process. This was considered important as the initial order divided the participants into different centres which may have influenced the results. Filtering and re-ordering was useful for example comparing different components against age or gender.

After the data had been assessed in its entirety different techniques were applied to the framework in order to view the data with a different perspective: In order to visualise demographic data within the framework different colours and shading options were used to differentiate age and gender brackets. Finally it was realised that inference could be different depending on whether comments related to a pre-operative or post operative state. In order to differentiate the two states different coloured text was used as a visual cue during analysis; blue to represent pre-operative remarks and orange for post-operative.
**Figure 9: Section of the thematic matrix**

<table>
<thead>
<tr>
<th>ID</th>
<th>Gender</th>
<th>Age</th>
<th>Home</th>
<th>Knowledge</th>
<th>Family Impact</th>
<th>Communication</th>
<th>Self Care</th>
<th>Domestic Life</th>
<th>Medical</th>
<th>Regurgitation</th>
<th>Activities of Daily Living</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>-ve</td>
<td>+ve</td>
<td></td>
</tr>
<tr>
<td>CH01</td>
<td>F</td>
<td>19</td>
<td>rural</td>
<td>3 4 10</td>
<td>2 5 6</td>
<td>2 7 8 15</td>
<td>14</td>
<td>16</td>
<td>14 16</td>
<td>SG9 Ph15 Tr15</td>
<td></td>
</tr>
<tr>
<td>CH02</td>
<td>M</td>
<td>21</td>
<td>rural</td>
<td>19 20 25</td>
<td>FC19 FC20</td>
<td>21 22 23 24</td>
<td>17</td>
<td>18</td>
<td>18</td>
<td></td>
<td></td>
</tr>
<tr>
<td>CH04</td>
<td>M</td>
<td>36</td>
<td>rural</td>
<td>31 34 45</td>
<td>EC30</td>
<td>36 37 38 33</td>
<td>33</td>
<td></td>
<td>35</td>
<td></td>
<td></td>
</tr>
<tr>
<td>CH06</td>
<td>F</td>
<td>12</td>
<td>urban</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>52</td>
<td></td>
<td>51 53</td>
<td>51</td>
</tr>
<tr>
<td>CH07</td>
<td>M</td>
<td>6</td>
<td>rural</td>
<td>57 59</td>
<td></td>
<td>56</td>
<td>56 60 61</td>
<td>55 63</td>
<td>63</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

CH04 (31). Now my family feel upset that that they didn’t know to come for surgery.
CHAPTER 8: DEVELOPING A TRAINING PROGRAMME FOR THE TOM-clp

8.1. Instruction and training

The strength of research is dependent on data being collected in a robust and consistent manner. This demands not only a stable and reliable measuring tool but also one that has good sensitivity and can be implemented accurately and reliably. The reliability and sensitivity of the TOM-clp has been detailed in previous research (Rees et al, 2012) so the main focus of this aspect of the research was directed at ensuring accurate implementation.

A major source of inaccuracy in data collection is observer error, particularly where there is an element of subjectivity in the assessment and where multiple observers are involved. In order to allow for inter-observer bias it is essential to ensure that training is comprehensive and consistent and that an evaluation of observer agreement is undertaken. For the purposes of this study there were two elements of data collection and training which needed to be addressed: firstly the teaching and application of the TOM-clp, and secondly the use of the data collection tool. Each of these elements was considered to ensure that they were taught and used in a dependable and reliable manner which was consistent with that expected for use in the field.

8.1.1. TEACHING AND APPLICATION OF THE TOM-clp

The TOM-clp is a simple and straightforward outcome measure developed to be used by non specialist staff in the field. Its clinical application involves informal interviews which provide information from which a subjective assessment as to the level of activity, participation and well-being of each participant can be made. Prior to application it is essential that each observer is trained to a set standard in order to maximise the consistency of the raw data. The chosen method of teaching for this purpose was simulation based learning using hypothetical case scenarios which was considered to replicate real life experience whilst not exposing patients to unnecessary risk or inconvenience.
Simulated learning allows students to become familiar with new information in a realistic situation without the pressure of clinical stress (Gaba 2004). It has been shown that this style of inductive education enhances students’ learning through scenario analysis as well as giving them a good conceptual understanding of additional background information provided in parallel. The result is a deeper and longer lasting understanding than occurs with more conventional forms of directional instruction (Ramsden, 2003; Norman and Schmidt, 1992).

Case based simulated learning has the advantage of being reproducible and ensures that all students are exposed to a wider range of situations in a short time frame than would be possible using a real time clinical situation (Lateef, 2010). This encourages a broad knowledge base whilst providing consistency of training between different groups. Such equity in exposure is particularly beneficial when using a case based technique for testing performance and reliability when applying the new skill.

### 8.2. Creating a training manual

The manual (Appendix 7) was designed to provide a basic introduction to the project and to be used alongside the training workshop rather than a fully comprehensive stand alone tool. Care was taken to ensure that the manual was easy to read, visually appealing and that the language was straightforward avoiding complex terminology. Where new concepts or terms were used they were accompanied by an explanation using more basic vocabulary.

The manual included a brief introduction to cleft palate and an explanation of why it can be a problem and how it can be managed. This was followed by a brief introduction to the research concept, the proposed role the participants and a sample of how the assessments would be made. Care was taken to explain that a fuller explanation of the process would be provided during the workshop session. A copy of each or the case scenarios was included as an appendix along with a copy of the TOM-clp in order that the participants could familiarize themselves with the information prior to the training workshop.
8.2.1. Developing reliable case scenarios

There are three basic elements required in a case scenario; it should be based on a real life situations; it requires the learner to study and understand the scenario; it has multiple perspectives and does not lead the learner to a particular conclusion.

Using the conceptual framework outlined in Figure 10 (Kim et al., 2006) the case scenarios were developed based on real life testimonials kindly shared for this purpose by Operation Smile. Each aspect of the framework was carefully considered as outlined below and incorporated as appropriate.

---

**Figure 10: Conceptual framework of teaching case development**

<table>
<thead>
<tr>
<th>CONTENT</th>
<th>Level of learner</th>
<th>Objective</th>
<th>Case setting</th>
<th>Distracters</th>
<th>Authenticity</th>
<th>Multiple perspectives</th>
<th>Rich</th>
<th>Unusual cases</th>
<th>Varying difficulty</th>
</tr>
</thead>
<tbody>
<tr>
<td>STRUCTURE</td>
<td></td>
<td></td>
<td>Gradual disclosure</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Multiple cases</td>
</tr>
<tr>
<td>ATTRIBUTE</td>
<td>RELEVANT</td>
<td>REALISTIC</td>
<td>ENGAGING</td>
<td>CHALLENGING</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>PROCESS</td>
<td>Build on prior knowledge</td>
<td>Assessment</td>
<td>Feedback</td>
<td></td>
<td>Teaching aid</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

From A conceptual framework for developing teaching cases: a review and synthesis of the literature across disciplines (Kim et al., 2006)
8.2.2. Content, Structure and Attributes

8.2.2.1. Level of learner

The TOM-clp is appropriate for use by non specialist staff in developing countries so scenarios were written using non-medical terminology and in straightforward language to allow good comprehension where participants use English as a second language or where scenarios require translation.

8.2.2.2. Objectives

The objective of the case scenarios was to represent a true clinical picture simulating the person’s circumstances before and after intervention for untreated CP. For this reason a 'before' and 'after' scenario was included for each hypothetical case to be considered and scored independently.

Care was taken to include enough information to allow students to make a subjective evaluation of each of the domains of the TOM-clp; activity, participation and well-being. Assessment of the impairment was not included in the scenario since this element is measured clinically using a technique taught as a direct skill using the data collection tool.

8.2.2.3. Case setting

Cases were composed to reflect activities of daily living in a community setting similar to those likely to present in a field situation. Since the participants potentially represent a wide range of social and ethnic backgrounds attempts were made to include an equal diversity within the case scenarios in order that students could identify with the teaching resource.

8.2.2.4. Authenticity and distracters

Case scenarios were based on true testimonials to keep them authentic, however additional content was added to ensure that adequate information was available for participants to make a considered assessment. Cases were also expanded to include tangential information as distracters in order to encourage participants to sift out non-pertinent information when making their assessments. For some scenarios some information was deliberately omitted in order to challenge
participants in representation of real life situations where relevant data may well not be available (Thomas, 1992; Eshach, 1992)

8.2.2.5. Rich and engaging

Every effort was made to make the hypothetical characters in the scenarios as engaging as possible in order that the students could empathise with their situation. A degree of layering and embellishment was used to expand the life stories for this purpose since it has been shown that if students can identify well with the cases they are more likely to analyse and reflect on the information provided (Lunderberg, 1997).

8.2.2.6. Multiple cases

Ten cases were developed to include wide ranging ages and different ethnic and socio-economic backgrounds. They were designed to reflect real life and encourage comparison of individuals’ circumstances by showing the contrasting reactions and attitudes of typical and less predictable individuals. Different extremes were included so assessments could be made which utilized a full range of scales within the TOM-clp.

8.3. Piloting the training manual

8.3.1. Process

Although the TOM-clp is designed for use by individuals with or without a clinical background a decision was made to pilot the training system with participants with at least some degree of medical knowledge. It was felt that this would elicit a better level of feedback than from individuals without any clinical background. However none of the participants approached had any direct experience of working specifically with CLP.
8.3.1.1. Phase 1: Analysis of resource content

Before piloting the entire training process for the TOM-clp an initial assessment was carried out to assess the appropriateness of the information content of the case scenarios. This was undertaken by means of an informal 90 minutes session training session and discussion group.

8.3.1.2. Participants

The project was discussed with a local principal of a dental practice who approached her dental nurses regarding their potential interest in participating. Each individual was provided with an information sheet (appendix 8) which included an outline of the project, what to expect from the participation process, how the information provided would be utilised and a sample of the data collection sheet. Individuals were reassured that they could withdraw from the process at any time without giving reason and without consequence.

Three dental nurses were recruited through this process and asked to read and sign a consent form (Appendix 9) verifying their willingness to participate and confirming their full understanding of the information provided. An informal meeting was arranged with the three participants and each was provided with a copy of the scenarios and an outline of the proposed session process.

8.3.2. Process

The session was undertaken in a small informal group using a quiet office at the dental practice away from general disturbance using the programme outlined in table 3.2 The session was lead by the primary researcher and participants were encouraged to interrupt and ask questions freely throughout the meeting.
Table 3.2: Session programme for phase 1

<table>
<thead>
<tr>
<th>DURATION</th>
<th>FUNCTION</th>
</tr>
</thead>
<tbody>
<tr>
<td>Introductions and Familiarisation Session (15 minute)</td>
<td>Group introductions and familiarising the participants with the case scenarios and the purpose of the study</td>
</tr>
<tr>
<td>Introduction to the TOM-clp (15 minute)</td>
<td>Explaining the rationale and application of the TOM-clp model</td>
</tr>
<tr>
<td>Practical group workshop (15 minute)</td>
<td>Practical session on the technique for assessing the case scenarios using the data collection sheet provided</td>
</tr>
<tr>
<td>Break (15 minute)</td>
<td></td>
</tr>
<tr>
<td>Practical session (30 minute)</td>
<td>Using the TOM-clp on six case scenarios</td>
</tr>
</tbody>
</table>

After introductions there was a period of familiarisation outlining the purpose of the session and an explanation of the role of the TOM-clp in the process. The scenarios were then discussed and the first example considered as a group training exercise in order to establish the scoring requirements and use of the data collection sheet (Table 8:1)
Table 8.1: Participant score sheet

Please read the case scenarios in the research manual provided. Using the scoring system outlined below mark the appropriate box in the score sheet to indicate how easily are you able to assess the following from the information provided.

<table>
<thead>
<tr>
<th>SCORING SYSTEM GRADED FROM 1 TO 5</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 = All the information is available and assessment is made easily</td>
</tr>
<tr>
<td>5 = None of the information is available and assessment is not possible</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>BEFORE SURGERY</th>
<th>ACTIVITY</th>
<th></th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>Not at all</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Ability to be understood</td>
<td>easily</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
<td>Problems eating and/or drinking</td>
<td>easily</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
<td>PARTICIPATION</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Willingness to socialise and join in activities</td>
<td>easily</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
<td>Willingness to speak to strangers</td>
<td>easily</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
<td>Willingness to eat in public</td>
<td>easily</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
<td>WELL-BEING</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>level of anxiety / happiness / embarrassment</td>
<td>easily</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
<td>General feeling of knowing the subject</td>
<td>easily</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>Not at all</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>AFTER SURGERY</th>
<th>ACTIVITY</th>
<th></th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>Not at all</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Ability to be understood</td>
<td>easily</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
<td>Problems eating and/or drinking</td>
<td>easily</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
<td>PARTICIPATION</td>
<td></td>
<td></td>
<td></td>
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<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Willingness to socialise and join in activities</td>
<td>easily</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
<td>Willingness to speak to strangers</td>
<td>easily</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
<td>Willingness to eat in public</td>
<td>easily</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
<td>WELL-BEING</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>level of anxiety / happiness / embarrassment</td>
<td>easily</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
<td>General feeling of knowing the subject</td>
<td>easily</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>Not at all</td>
</tr>
</tbody>
</table>
Once it had been established that the process was fully understood the dental nurses were requested to assess the content of each of the remaining scenarios independently and asked to provide self-reported feedback indicating the usefulness and limitations of individual cases.

On completion of the exercise the scenarios and scores were discussed in more detail in relation to each assessment in order to ascertain which of the individual components contributed to ease of decision making process, which were confusing, and where they thought additional information would benefit the process.

Participants were thanked for their contribution and informed that they could be kept up to date with the progression of the research via email if they would like.

8.3.3. Results

Overall the case scenarios were considered appropriate and easy to understand cases 2, 3, 8 and 9 requiring little change whereas it was felt that improved clarity would have made assessment easier in the other cases. There was some confusion for scenarios 1 and 7, where information was included regarding the persons past and assessors were unclear whether the assessment should be made based on the current situation or from an historic point of view. Comments were also made that scenarios 1 and 6 lacked information about eating and drinking which would have made assessment easier. Scenarios 5 and 7 were thought to be confusing owing to the amount of information giving something of a mixed message whereas cases 4 and 10 were felt to be lacking in information although they were easy to assess. Details of each scenario are detailed below.
<table>
<thead>
<tr>
<th>Participant 1</th>
<th>Participant 2</th>
<th>Participant 3</th>
</tr>
</thead>
</table>

**Case 1: Juana pre-op**

![Graph showing data for Juana pre-op](image)

**Comments**

No information regarding eating and drinking. Some confusion over whether to answer as she was at presentation or retrospectively as a child. Unsure of rating speech.

**Case 2: Averoot pre-op**

![Graph showing data for Averoot pre-op](image)

**Comments**

Straightforward to assess.

**Juana post-op**

![Graph showing data for Juana post-op](image)

**Case 2: Averoot post-op**

![Graph showing data for Averoot post-op](image)

**Comments**

Straightforward to assess.
## Case 3: Nosantu pre-op vs. Nosantu post-op

<table>
<thead>
<tr>
<th>Ability to be understood</th>
<th>Problems eating and drinking</th>
<th>Willingness to socialise</th>
<th>Willingness to speak to strangers</th>
<th>Level of anxiety</th>
<th>General feeling of knowing the person</th>
</tr>
</thead>
</table>

### Comments

**Straightforward to assess**

**Very straightforward to assess**

## Case 4: Lokhi pre-op vs. Lokhi post-op

<table>
<thead>
<tr>
<th>Ability to be understood</th>
<th>Problems eating and drinking</th>
<th>Willingness to socialise</th>
<th>Willingness to speak to strangers</th>
<th>Level of anxiety</th>
<th>General feeling of knowing the person</th>
</tr>
</thead>
</table>

### Comments

**Generally straightforward to assess.**

Participant 3 unsure regarding willingness to talk because he wrote things down rather than speaking.

**Straightforward to assess.**

No specific information regarding speaking to strangers or eating in public but assumed by participant 1 from information about willingness to socialise.
### Case 5: Kiran pre-op

<table>
<thead>
<tr>
<th>Ability to be understood</th>
<th>Problems eating and drinking</th>
<th>Willingness to socialize</th>
<th>Willingness to eat in public</th>
<th>General feeling of knowing the...</th>
</tr>
</thead>
<tbody>
<tr>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
</tbody>
</table>

**Comments**

Although there was a lot of information about Kiran it was generally thought difficult to assess and would be better if it was worded more clearly.

### Kiran post-op

<table>
<thead>
<tr>
<th>Ability to be understood</th>
<th>Problems eating and drinking</th>
<th>Willingness to socialize</th>
<th>Willingness to eat in public</th>
<th>General feeling of knowing the...</th>
</tr>
</thead>
<tbody>
<tr>
<td>0</td>
<td>2</td>
<td>1</td>
<td>3</td>
<td>4</td>
</tr>
</tbody>
</table>

**Reasonably straightforward to assess, confusion whether being too shy to eat in public counted under 'problems with eating and drinking'**

### Case 6: Keo Neang pre-op

<table>
<thead>
<tr>
<th>Ability to be understood</th>
<th>Problems eating and drinking</th>
<th>Willingness to socialize</th>
<th>Willingness to eat in public</th>
<th>General feeling of knowing the...</th>
</tr>
</thead>
<tbody>
<tr>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
</tbody>
</table>

**Comments**

Straightforward to assess, felt there wasn't much specific information regarding anxiety and eating in public so difficult to assess these points.

### Keo Neang post op

<table>
<thead>
<tr>
<th>Ability to be understood</th>
<th>Problems eating and drinking</th>
<th>Willingness to socialize</th>
<th>Willingness to eat in public</th>
<th>General feeling of knowing the...</th>
</tr>
</thead>
<tbody>
<tr>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
</tbody>
</table>

**Straightforward to assess**
### Case 7: Jacqueline pre-op vs. Jacqueline post-op

<table>
<thead>
<tr>
<th>Ability to be understood</th>
<th>Problems eating and drinking</th>
<th>Willingness to socialize</th>
<th>Willingness to eat in public</th>
<th>Level of anxiety</th>
<th>General feeling of knowing the...</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>6</td>
</tr>
</tbody>
</table>

**Comments**

Felt the scenario was confusing with mixed messages making it difficult to assess, difficult to know which information was current and which historic.

Not really enough information to make a clear assessment, too much about her dream for the future rather than how she is now.

### Case 8: Demitria vs. Demitria post-op

<table>
<thead>
<tr>
<th>Ability to be understood</th>
<th>Problems eating and drinking</th>
<th>Willingness to socialize</th>
<th>Willingness to eat in public</th>
<th>Level of anxiety</th>
<th>General feeling of knowing the...</th>
</tr>
</thead>
<tbody>
<tr>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
</tbody>
</table>

**Comments**

Straightforward to assess wording not very clear regarding willingness to socialize caused some confusion

Generally straightforward to assess
Case 9: Kaza pre-op

| Comments | Straightforward to assess | Not enough information and a bit vague so difficult to assess |

Case 10: Issac pre-op

| Comments | Information about activity in public was a bit difficult as scenario focused on home life | Reasonably straightforward to assess but would have liked a bit more information. |
Each of the comments was considered and the case scenarios adjusted accordingly as summarised in table 4. At this point it was concluded that each of the scenarios was straightforward to assess and could be used in a training manual as a teaching aid for the TOM-clp.

<table>
<thead>
<tr>
<th>Table 4: Summary of changes made to case scenarios</th>
</tr>
</thead>
<tbody>
<tr>
<td>Juana Pre-op</td>
</tr>
<tr>
<td>Juana post op</td>
</tr>
<tr>
<td>Averoot pre-op</td>
</tr>
<tr>
<td>Averoot post-op</td>
</tr>
<tr>
<td>Nosantu pre-op</td>
</tr>
<tr>
<td>Nosantu post-op</td>
</tr>
<tr>
<td>Lokhi pre-op</td>
</tr>
<tr>
<td>Lokhi post-op</td>
</tr>
<tr>
<td>Kiren pre-op</td>
</tr>
<tr>
<td>Kiren post-op</td>
</tr>
<tr>
<td>Keo Neang pre-op</td>
</tr>
<tr>
<td>Keo Neang post-op</td>
</tr>
<tr>
<td>Jacqueline pre-op</td>
</tr>
<tr>
<td>Jacqueline post-op</td>
</tr>
<tr>
<td>Demitria pre-op</td>
</tr>
<tr>
<td>Demitria post-op</td>
</tr>
<tr>
<td>Kaza pre-op</td>
</tr>
<tr>
<td>Kaza post-op</td>
</tr>
<tr>
<td>Issac pre-op</td>
</tr>
<tr>
<td>Issac post-op</td>
</tr>
</tbody>
</table>
8.3.4. Discussion

Initially the participants were concerned that there was a lot of information missing in the scenarios so they had to score 5 for many of the domains. However it was explained that some information had deliberately been omitted to be more representative of a real life case where a judgement may need to be made based on limited information. It was important however that every scenario could be assessed consistently in relation to each of the TOM-clp domains.

There were many useful comment relating to wording of the scenarios which were utilised to improve their readability and comprehensiveness, however many comments were particularly pertinent to the need for more background information in the teaching process. Participants were particularly unsure when they were provided with historic information yet it was unclear if this should be used in the assessment. Knowing this ensured that it could be included in the pre-training process to avoid discrepancies in the assessment process. This was particularly valuable and would have been missed if this pilot process had not been arranged.

8.3.5. Conclusion

Developing good case scenarios was considered fundamental to the training process. Through the process of content analysis it was possible to upgrade the case scenarios to ensure that they could be more accurately and consistently used as an exercise in a training manual designed to teach the understanding and implementation of the TOM-clp in the field.
8.4. Phase 2: Piloting the training process

8.4.1. Participants

Participants were approached from two sources for this phase.

1. The project was discussed with a senior lecturer at a dental training institute in a city close to the researcher. He then approached his dental students regarding their potential interest in participating.

2. The project was discussed with two post graduate supervisors at the University of Sheffield who in turn invited post graduate students to participate. This source was considered important since it was directed at overseas students who used English as a second language which was important in ensuring that the terminology in the material was clear and free from jargon which may prove difficult to understand for a non native speaker.

Interested individuals were provided with an information sheet (Appendix 8). In addition they were given a copy of the training manual to peruse in order that they would be familiar with the material prior to the training session.

These processes generated three dental students and three PhD students in the first instance then a further four dental students indicated their interest for the final pilot.

8.4.2. Process

Three sessions were undertaken in total; the first involving the three initial dental students at the university in Cumbria; the second involving the three PhD students at the University of Sheffield; and the third involving the four additional dental students at the dental school in Cumbria.

Sessions were facilitated in a quiet office lead by the primary researcher. A restful quiet environment was prepared and terminology kept within the ability and language skills of the participants since it has been shown to keep observer rating bias to a minimum. Sessions were kept relaxed so that individuals felt comfortable
interjecting and asking questions to clarify uncertainties (Bailey, 1997; John et al., 2002).

Training in the use of the TOM-clp was administered in accordance with the methodology for use of the TOM as described in the original TOMs manual (Enderby et al. 2006). Several aspects were considered to increase reliability of the test session: John and Enderby (2000) recognized that two and a half hours was the most effective length of session since it is long enough to impart adequate information whilst not risking fatigue and loss of interest. However, training for the original pilot of the TOM-clp (Rees, 2012) was limited to two hours whilst still resulting in good inter-rater reliability so a similar format was used for these sessions as outlined in table 3.

Table 3: session programme for phase 1

<table>
<thead>
<tr>
<th>DURATION</th>
<th>FUNCTION</th>
</tr>
</thead>
<tbody>
<tr>
<td>Introduction and familiarisation (30 mins)</td>
<td>To familiarise the students with the nature and impact of CLP and introduce them to way in which it can affect the lives of individuals living with an untreated cleft and implications of intervention</td>
</tr>
<tr>
<td>Introduction to TOM-clp (15 mins)</td>
<td>To explain the underlying principle and application of the TOM-clp model</td>
</tr>
<tr>
<td>Practical group workshop (15 mins)</td>
<td>Practical workshop session to demonstrate the technique for implementation of the TOM-clp using a hypothetical case scenario</td>
</tr>
<tr>
<td>Break (15 mins)</td>
<td></td>
</tr>
<tr>
<td>Practical session (45 mins)</td>
<td>Testing the use of the TOM-clp using the remaining nine case scenarios</td>
</tr>
</tbody>
</table>
Participants were also asked for feedback on the format and usability of the teaching manual and asked for their consideration on how it could be improved. Improvements were made after each session until no further potential improvements were highlighted.

8.4.3. **Statistics and analysis**

Inter-rater reliability was assessed using the Intra-Class Correlation Coefficient two-way random, average consistency, random model in IBM SPSS Statistics 21. The Intra-class correlation (ICC) was chosen to assess inter-rater reliability since it accommodates multiple assessors and is valid where certain subsets are rated by multiple assessors and the rest by a single assessor. Unlike Kappa, ICCs incorporate the magnitude of any disagreement where a larger magnitude of disagreement results in a lower ICC. High ICC indicates good agreement with perfect agreement equalling 1 and chance agreement equalling zero using the cut offs as cited according to Cicchetti (1994):

<table>
<thead>
<tr>
<th>Level</th>
<th>ICC Range</th>
</tr>
</thead>
<tbody>
<tr>
<td>Poor</td>
<td>&lt; .40</td>
</tr>
<tr>
<td>Fair</td>
<td>.40 - .59</td>
</tr>
<tr>
<td>Good</td>
<td>.60 - .74</td>
</tr>
<tr>
<td>Excellent / Near perfect</td>
<td>.75 - 1.0</td>
</tr>
</tbody>
</table>

Rater agreement was also compared according to the difference in maximum and minimum scores given for each domain (table 5). Agreement is considered good for the TOM if raters agree within one unit as required for the original TOM (Enderby & John, 2015).
### 8.4.4. RESULTS

Table 4: TOM-clp scores for session 1, 2 and 3

<table>
<thead>
<tr>
<th></th>
<th>SH</th>
<th>SJ</th>
<th>AK</th>
<th>NF</th>
<th>RB</th>
<th>RN</th>
<th>JR</th>
<th>MS</th>
<th>AC</th>
<th>NS</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Session 1</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
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<td></td>
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</tr>
<tr>
<td>Averoot</td>
<td></td>
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<td></td>
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<tr>
<td>Pre-op &amp; post</td>
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<tr>
<td>Nosantui</td>
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<td>Pre-op &amp; post</td>
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<tr>
<td><strong>Session 2</strong></td>
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<td>Activity</td>
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<td>Participation</td>
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<td>well-being</td>
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<td><strong>Session 3</strong></td>
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<tr>
<td>well-being</td>
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<td></td>
<td></td>
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</tr>
</tbody>
</table>

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152
Two-way random effects model where both people effects and measures effects are random.

a. The estimator is the same, whether the interaction effect is present or not.

b. Type C intraclass correlation coefficients using a consistency definition-the between-measure variance is excluded from the denominator variance.
<table>
<thead>
<tr>
<th>Case Scenario</th>
<th>Domain</th>
<th>Range (difference between highest and lowest rating)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Difference in agreement &gt; 1</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Averoot pre-op</td>
<td>Activity</td>
<td>1</td>
</tr>
<tr>
<td></td>
<td>Participation</td>
<td>1</td>
</tr>
<tr>
<td></td>
<td>Well being</td>
<td>0.5</td>
</tr>
<tr>
<td>Averoot post-op</td>
<td>Activity</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>Participation</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>Well being</td>
<td>0.5</td>
</tr>
<tr>
<td>Nosantui pre-op</td>
<td>Activity</td>
<td>0.5</td>
</tr>
<tr>
<td></td>
<td>Participation</td>
<td>0.5</td>
</tr>
<tr>
<td></td>
<td>Well being</td>
<td>0.5</td>
</tr>
<tr>
<td>Nosantui post-op</td>
<td>Activity</td>
<td>1.5</td>
</tr>
<tr>
<td></td>
<td>Participation</td>
<td>0.5</td>
</tr>
<tr>
<td></td>
<td>Well being</td>
<td>0.5</td>
</tr>
<tr>
<td>Lokhi pre-op</td>
<td>Activity</td>
<td>0.5</td>
</tr>
<tr>
<td></td>
<td>Participation</td>
<td>0.5</td>
</tr>
<tr>
<td></td>
<td>Well being</td>
<td>0.5</td>
</tr>
<tr>
<td>Lokhi post-op</td>
<td>Activity</td>
<td>1.5</td>
</tr>
<tr>
<td></td>
<td>Participation</td>
<td>0.5</td>
</tr>
<tr>
<td></td>
<td>Well being</td>
<td>0.5</td>
</tr>
<tr>
<td>Kiran pre-op</td>
<td>Activity</td>
<td>0.5</td>
</tr>
<tr>
<td></td>
<td>Participation</td>
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<td>Kiran post-op</td>
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<td>Well being</td>
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<td>Keo Neang pre-op</td>
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<td>Jacqueline post-op</td>
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<td>Well being</td>
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<td>Demitria pre-op</td>
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<td>Participation</td>
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<tr>
<td></td>
<td>Well being</td>
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8.5. DISCUSSION:

For each of the sessions all the participants were able to rate the case scenarios independently after the training session. Using standard agreement convention there is an excellent agreement between observers both before and after modifications to the case scenarios indicating that the TOM-clp can be used to assess the case scenarios with a near perfect level of consistency after only an hour of training. This is important since, with only a small sample size and few observers a high level of reliability is required to minimise the effect of random error. Thorndike and Hagen (1969) suggest a reliability of at least .75 under such circumstances. The scores of .9 and above in all the sessions indicate a very low random error and a good confidence in the reliability of the TOM-clp under these conditions. Since all the sessions produced a high ICC there is no indication from these values as to whether the alterations in the scenarios influenced the assessments.

Whilst the ICC did not show a significant difference between the three sessions the difference in the range improved after the final adjustments from session 2 with all aspects of the scenarios being judged within 1 unit by the third session. Six of the seven case domains showing discrepancies greater than 1 in the first session were scored accurately in the second session indicating that the changes in the scenarios may have improved the participants’ ability to gauge
the TOMs scores. The fact that eight different case domains in the second session showed discrepancy greater than 1 may have been indicative of miscomprehension due to language issues by the non native English speakers in session 2. From the graphical data it is clear that the discrepancies did not relate to any one individual over or under estimating consistently indicating that the underlying problem lay with the scenarios rather than the participants.

8.6. CONCLUSION

Although there is not a statistical difference in reliability between the original and the modified text the near perfect ICC indicate that the assessors had a high rate of agreement suggesting that the TOM-clp ratings were similar across all participants. This high agreement indicates that the case scenarios can be considered suitably accurately for training purposes. In addition the range of scores was reduced to below 1 point in all of the domain ratings by session 3 suggest that the modifications improved the comprehensibility for each of the domains.

It is appreciated that there is a bias introduced as a result of the small number of observers and the fact that the observers and text were altered as the process progressed and ideally each of the participant groups would have been asked to re-rate the scenarios after the text modifications had been made. However the results suggests that minimal measurement error has been introduced and as such the statistical power of the TOM-clp as an outcome measure assessing the given scenarios is considered suitable for training, data collection and subsequent analysis.
CHAPTER 9: RESULTS

SECTION 1: OVERALL DEMOGRAPHICS, AVAILABILITY OF SERVICES AND FACTORS IMPEDING ACCESS TO INTERVENTION.

9.1. Data collection

Data was collected as described using two techniques; either downloaded electronically via a specifically designed app or direct annotation by hand and transcription from recorded face to face interviews. The first of these techniques was used longitudinally by clinical leads and/or the PI to collect the data for all for 27 participants in Assam over a 10 week period, and 32 sets of data in Hyderabad over a 25 week period. The data included demographic information, a series of simple response questions, as outlined previously and TOM-clp scores. All the data in Assam was pre-operative whereas in Hyderabad both pre-operative and post-operative data was collected.

The remainder of data was collected by the PI at all three centres in Hyderabad, using face to face interviews over a 2 week period during February 2017. Data comprised current and retrospective information regarding demographics, TOM-clp scores and qualitative information for use in Framework analysis. This was important in order to examine the qualities of the TOM-clp in its ability to demonstrate change over time and perform as an outcome measure. Therefore, for the purpose of analysis, the TOM-clp scores collected in the interview process have been treated independently from that collected via the app.
9.1.1. Reason for excluding data

9.1.1.1. Participant recruited in Assam

All 27 participants recruited in Assam fitted the inclusion criteria and were retained in the study.

9.1.1.2. Participant recruited in Hyderabad - app data collection

32 participants were recruited initially however eleven were excluded as not fitting the inclusion criteria; either having received surgery at an optimal time and/or were under the age of six years. An additional three cases were excluded as they presented with untreated soft palate cleft only which left a total cohort of 18 participants in the final data set.

9.1.2. Participant recruited for interview in Hyderabad

The total number of participants recruited for interview was 48, however eight were excluded as they did not fit the inclusion criteria: One child was severely syndromic, two were under the age of 6 and four had received treatment at an appropriate time.

The eighth exclusion was a twelve year old boy who had received CLP surgery at one year old but the entire pre-maxillary segment had been removed, rather than repaired. This had resulted in the complete absence of the anterior teeth, lip and palatal segment giving exceedingly poor function, incomprehensible speech and considerably reduced aesthetics. According to the clinical lead, such examples of inappropriate surgery are all too common and significantly increase the complexity of corrective surgery at a later date. Although this case was excluded, as there was no residual palatal fistula, a decision was made to include cases where ineffectual or inappropriate surgery had relapsed and left a residual palatal defect.

Following the exclusion of these eight cases the final dataset comprised 40 participants for analysis in the framework matrix and comparative TOM-clp scores.
9.2. Demographic information for Assam and Hyderabad

Table 9:1: Demographic summary for Assam and Hyderabad

<table>
<thead>
<tr>
<th></th>
<th>Assam</th>
<th>Hyderabad</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total number of participants recruited</td>
<td>27</td>
<td>80</td>
</tr>
<tr>
<td>Number of participants after exclusions</td>
<td>27</td>
<td>58</td>
</tr>
<tr>
<td>Method of collection</td>
<td>100% app data</td>
<td>39% app data: 61% interview</td>
</tr>
<tr>
<td>Languages spoken</td>
<td>Assamese, Garo, Bengali, Nepalese</td>
<td>Telugu, Urdu, English</td>
</tr>
<tr>
<td>Male: Female</td>
<td>11:16</td>
<td>30:28</td>
</tr>
<tr>
<td>Age range (Figure 11)</td>
<td>6 - 23</td>
<td>6 - 37</td>
</tr>
<tr>
<td>Place of residence</td>
<td>100% rural</td>
<td>76% rural, 14% urban</td>
</tr>
<tr>
<td>Time to reach cleft centre (Figure 12)</td>
<td>1 - 14 hours</td>
<td>1 - 14 hours</td>
</tr>
<tr>
<td>Ratio CLP: ICP: Surgical breakdown (SB)</td>
<td>88% CLP: 22% ICP: 0% SB</td>
<td>81% CLP: 9% ICP: 10% SB</td>
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</table>

9.2.1. Age range (Figure 11)

The age ranges show a normal distribution for the Hyderabad with 75% of the participants falling in the mid range; between ages 11 and 25. The Assam data is heavily skewed towards younger participants with over 75% presenting under 15 years old.
Figure 11: Age ranges for participants in Assam and Hyderabad (n=85)

9.2.2. Place of residence and distance from cleft centres

All of the participants from Assam were recruited from rural locations in comparison with only 76% of the Hyderabad cohort. Interestingly the overall times taken to reach the cleft centre were greater in Hyderabad than Assam, with nine individuals travelling for over 15 hours for their appointments (Figure 12).

Figure 12: Time to get to cleft centre in hours

(Concentric rings = miles from cleft centre)
9.3. Degree of nasal regurgitation

In addition to demographic data and TOM-clp scores participants were asked how often they suffered from nasal regurgitation. Pre-operatively more than two thirds suffered from this problem to some degree and for 22% of the participants regurgitation occurred every time they ate or drank anything (Figure 13). The impact of regurgitation and the affect of surgical intervention will be discussed more fully using framework analysis in the following section.

Figure 13: Number of participant reporting nasal regurgitation pre-operatively (n=58)
9.4. Reason for not seeking intervention sooner (Figure 14)

It had been planned to ask all participants what factors they felt had limited their access to more timely treatment. Unfortunately this information was not collected from the Assam cohort but was available for all except four of the Hyderabad participants.

The majority of individuals had a general lack of awareness that treatment was possible or did not know that it could be accessed in their area. A quarter of the participants had already had treatment elsewhere but the surgery had failed, leaving a significant palatal defect: One individual had undergone five previous surgeries, none of which had been successful, all bar one of the remainder of the participants had undergone lip repair before attending these clinics, six had attended for lip surgery at an optimal time but through miscommunication had not returned for palate surgery.

One of the two mothers, who believed that the CLP was the fate of God, also thought the defect had resulted from seeing an eclipse whilst she was pregnant. Another couple, whose only child was born with severe bilateral CLP, were so terrified she might die during surgery that they did not go through with the operation and she remained completely untreated until the age of 33.

Only three participants said that a lack of finance was the problem, four families lived too far away and one had not thought the CP to be important until they realised the impact it had on speech.

Details relating to individual cases will be discussed in more detail in the following chapter using framework analysis.
9.5. Information from interviews with cleft care providers

The clinics attended were often extremely busy and time constraints disallowed the implementation of formal interviews with all the service providers. Two informal interviews were completed with clinical directors and further information was obtained during ad-hoc discussions with staff members throughout the field trips and via e-mail correspondence. Five different NGOs were approached for information, however only one responded and agreed to share information regarding their general policies and outlook.

The following information outlines the contents of these discussions in relation to the research questions pertinent to this study. It comprises direct quotes where they are available and additional anecdotal information. The use of all the material, including direct quotes, has been approved by each of the providers in person. Some of the information is cross-referenced to previous audits and studies undertaken at the centres which were referred to by the clinical directors during the interviews. These will be discussed further in relation to this study and other comparative literature in the discussion.
Question 1: What services are currently available for CLP repair, how are they promoted and what facilitates or limits their accessibility?

There was agreement in all the centres in Hyderabad and Assam, that there was a high demand for cleft services and many individuals were not attending for treatment at an optimal time. A variety of explanations were presented for the delay in accessing treatment, all of which matched those which had been reported in the literature (section 4.2), most commonly; lack of awareness, poor education and superstition. Naram, Makhijani et al. (2013) had undertaken a survey at one of the cleft centres, specifically aimed at identifying the barriers which prevented their patients from seeking more timely treatment. Their results also echoed those of the literature and have directed their thinking on how best to develop an education strategy.

The teams in Hyderabad believed that the majority of CLP patients, even those born at home, were brought to the cleft centre within 36 months of birth, mirroring the results of El-Shazly, Bakry et al. (2010) from their study in the same region. They felt that most of the patients attended their clinics with the expectation of improving aesthetics and/or quality of life but few were aware of impact of CLP on speech. However, despite their relatively timely presentation, the teams reflected that most patients had not been offered a scientific explanation for their CLP by the referring practitioner. They also commented that this lack of understanding was the most likely reason for patients returning for palate repair.

All the cleft teams concluded that there was a need to develop a collaboration with primary health care providers, schools and communities and disseminate information about treatment for CLP. They also recognised that people would be more receptive to advice if it was delivered in a culturally sensitive and empathetic manner.
In summary the teams outlined the following key points:

- Understanding the culture of a region is essential in tailoring the education according to beliefs and superstition.
- Education must be delivered in a format suitable for the community it serves: Examples include the production of a video in Telugu, the most commonly spoken local language of Telangana and Andhra Pradesh, which dispels the myths surrounding CLP, and short video clips which can be accessed by educators to show to parents to give them a better understanding of CLP (Singh and Das, 2010).
- The afore mentioned study by Naram et al. (2013) had highlighted the need to direct education towards a more scientific explanation for CLP and discourage them from seeking treatment from traditional healers and non-specialist surgeons. They were aware that care must be taken not to transfer a feeling of blame onto the parents as parents sometimes interpreted intended advice as a criticism that they did not manage their child's CLP appropriately.
- A recurring theme amongst the cleft care providers was the notion that mixing with other families with CLP does much to reduce personal feelings of isolation, lessen social stigma and improve general well-being. They felt that mingling also allows families to exchange stories, develop an understanding of what to expect from treatment and help to dispel their misconceptions about CLP. In particular they had noted that when parents are confronted by large numbers of cleft patients, spanning a wide age range, they came to realise that seeing an eclipse during pregnancy could not have been responsible.

All the cleft centres arranged education events; some organised cleft camps, sending a trained educator into schools and villages to identify individuals with CLP and disseminate accurate information about treatment options. Clinicians also mentioned that it should be part of the remit of trained ASHAs (see section 4.2) to identify CLP and refer cases for treatment, however, they were not convinced that this always happened. Additionally, patients with CLP and their families are encouraged to pass on the information in their own towns and villages.
One centre has an active partnership with a local hostel, which accommodates and supports 80 children with CLP from low income families around the region. Living in Hyderabad allows them to attend an inclusive local school and gives them access to comprehensive cleft care, including regular speech and language therapy, which would not have been possible if they were having to travel long distances for appointments.

The cleft teams reported that people are often unaware of free specialist cleft services and sometimes pay significant amounts for poor or inappropriate treatment. As mentioned, it is not uncommon to encounter patients who have had the premaxilla removed or poor techniques have resulted in such bad scarring that their ultimate outcome is severely compromised.

There was a perception amongst the teams that operational expertise varied between individual surgeons and different cleft centres: There are four care providers in Hyderabad supporting CLP, all funded by the same provider, yet offering a different quality and quantity of care. They were aware of a number of Ear nose and throat (ENT) surgeons, maxilo-facial surgeons, and paediatric surgeons doing occasional cleft repairs, which consequently required significant corrective surgery at the specialist centres. Even where surgeons have training in CLP, there was a general feeling that inexperience was the underlying reason for poor outcome in some centres. All the clinical directors felt that this disparity was inappropriate and that services should be more closely regulated to ensure parity and quality of care.

All the centres included in this study were run by resident multidisciplinary teams with high levels of experience, allowing them to provide comprehensive and cohesive care (Reddy et al., 2009). This was considered universally important as such services, as discussed in chapter 5, are reported as being associated with superior outcomes than those provided by more transient services. Two of the centres provided internships for local and visiting surgeons but only on placements of six months or more. They held concerns in relation to short-term parachute missions and brief visits from overseas surgeons and associated them with poor outcomes.
In terms of patient selection, none of the centres discriminated by age in terms of service provision and undertook treatment based solely on clinical need. The degree of external agency funding was very inconsistent between centres; At one extreme, a centre was fully funded for the provision of comprehensive cleft care including primary and secondary surgery, alveolar bone grafting, orthodontics, dental care, speech and language therapy. In addition, all costs for medication and dressing were included and patients were offered transport to the hospital, accommodation and subsistence, for one additional family member, for the duration of their stay. Two of the centres received external funding for certain aspects of this care and minimised costs by offering elements of secondary surgery, such as rhinoplasty and VPI repairs, in conjunction with other procedures, to reduce the cost of theatre time, travel and loss of earnings for the family. The fourth centre, which offered specialist dental and orthodontic care, received the least funding so, not only did they give their free time, they also funded much of the treatment from their own pockets. Unfortunately, since the patients get their surgery free of charge they have come to believe that the dental and orthodontic care should also be available free of charge, and the clinicians found it difficult to explain these disparities in the service.

The requirement for secondary surgery was a common topic of conversation at all the centres in Hyderabad. Apart from the aforementioned need for revision of failed procedures, the teams felt strongly that a more comprehensive care package should be offered to patients from lower socio-economic backgrounds. All the teams in Hyderabad commented that, whilst the gold standard for treatment of CLP often involves multiple surgical procedures, orthodontic intervention, dental care and SLT input, the global tendency was to believe that this cohort could be managed using only one or two procedures and minimal adjunct therapy. Whilst they appreciated that not all individuals would travel long distances to undertake extensive treatment, many would, and did, particularly when they realised what a lifelong improvements which could be achieved.

Discussions around audit revealed that, whilst standardised comprehensive medical records are kept at all of the centres, there is not currently any means of benchmarking or measuring outcomes, particularly in relation to less tangible
measures such as communication, patient satisfaction and well-being. The directors of one of the centres which offered secondary repairs, felt that there was too much emphasis on counting numbers of individual patients treated and not enough on monitoring quality. He considered that his centre was being penalised, as they did not get funding for 'putting right other surgeons mistakes', only for treating new cases. He was concerned that this was potentially promoting greater activity at centres with a high turnover, even if they did not necessarily have good outcomes. He commented that finding an appropriate measure of outcome for audit and benchmarking of services was the only way to expose and overcome these discrepancies. There was a general feeling, certainly in the Hyderabad centres, that the TOM-clp could prove a valuable adjunct to service provision in this capacity, particularly as it was linked to the WHO-ICF which they were aware was already being used as an outcome measure for other services.

A personal correspondence with the Vice president for Strategy and Evaluation for Smile Train (October, 2017) confirmed that, whilst the organisation has supported comprehensive multidisciplinary care since their inception, including secondary cleft surgery, speech therapy orthodontics and dental care, this has not been achievable universally. More recently, the organisation has emphasised the need to scale up these services and enhance its staffing levels to offer these levels of comprehensive care to a wider global population. Concurrently, they aim to complement their already comprehensive surgical record keeping, with an outcome measure which can reflect the quality of life and psychosocial gain associated with intervention, which could in turn be used for auditing and benchmarking these important elements of service provision.

9.6. Participants perception of communication and regurgitation before and after intervention

Although the psychosocial impact of regurgitation is investigated in the interviews and reviewed more fully in the framework analysis, each participant was asked to rate the level of regurgitation before and after surgery using a five point descriptive scale (Figure 15). Over three quarters of the participants complained of at least some degree of nasal regurgitation pre-operatively, and for almost one
third this occurred every time they ate or drank. Post-operatively, only 28% of participants reported regurgitation and then only on rare occasions. Whilst the data is not adequate for statistical analysis, the indicates a positive impact from intervention in relation to preventing regurgitation.

**Figure 15: Pre-operative and post-operative regurgitation (n=40)**

A similar trend can be seen with regards participants perception of their ability to communicate (Figure 16): Six participants could not be understood at all pre-operatively, and a further eight could only be understood by family members. Fifteen participants could converse with friends which means 70% of the participants could not be understood by strangers before treatment. Post surgery, this figure fell to a quarter with the majority being understood even by strangers most or all of the time. Communication will be discussed in more detail through framework analysis.

**Figure 16: Perception of communication before and after surgery**
9.7. Background information for interview participants (n=40)

The following section summarises the framework analysis carried out on the interview data from Hyderabad. The section begins with a short summary of the demographics for this cohort and a description of individuals perceived change in nasal regurgitation and communication skills following intervention. This is followed by a descriptive account of the interview data analysis, including direct quotes. The results are further illustrated by a number of short vignettes which reflect the broad spectrum of participant profiles and give a personal perspective to the data.

It is important to note that all the quotes represent the sentiments of the participants but are transcribed verbatim as they were spoken by the translator. This causes some grammatical confusion as the information was occasionally delivered in the form of a summary in the form of an explanation of what the participant had said or relayed in the third person rather than in the exact words of the participant. Attempts have been made to clarify these elements using bracketed explanation in the text.

9.7.1. Demographics

Forty participants fitting the inclusion criteria were recruited for interview at three centres in Hyderabad, resulting in 40 interviews for participants: Seventeen interviews were carried out at each of two centres and six interviews at the third.

Only two of the participants presented with ICP, the remainder had unilateral or bilateral CLP. Apart from one exception all had received lip surgery before the age of six.

There was an even gender distribution with ages ranging from 6 to 37 following a normal distribution with 75% being in the mid range of 11 to 25 (Figure 17)
77% of the participants came from rural rather than urban communities, some travelling up to 18 hours to reach the cleft centre, often overnight. Only seven participants had come from suburbs of Hyderabad, one of whom was a student at a local hostel but whose home town was several hundred kilometres away.

**QUESTION 1: WHAT FACILITATES OR LIMITS ACCESSIBILITY OF CLEFT CARE, WHAT SERVICES ARE CURRENTLY AVAILABLE AND HOW ARE THEY PROMOTED?**

### 9.8. Family background: Knowledge and impact of CLP

#### 9.8.1. Reasons for late intervention and understanding of CLP

##### 9.8.1.1. Lack of access to education about the condition

The previous section described the principle reason for the majority of families not seeking more timely treatment was a lack of awareness about CLP and the treatment opportunities. Three families had encountered a relative or another child with CLP previously but for most families the birth of their child was their first experience of the condition:

- *They have 2 male and 1 female child the male child both the male child are facing the same problem (CH07)*
ii. They didn’t know before the children were born what a cleft looked like (TB02)

A number of participants indicated that their families low educational status was the key reason which had limited their knowledge of treatment opportunities:

iii. [whether it could be fixed] that they didn’t know...... parents are uneducated we don’t know (CH01).

For others it was geographical isolation which impeded their access to information resulting in an assumption that nothing could be done to manage the situation:

iv. We were in a really rural area and my parents don’t know what to do or how to do (TB19).

Superstition

There was a general naivety amongst the families with regards to why the CLP had occurred but most families resigned themselves to the situation as simple misfortune. However, for a few families it was considered an act of fate or a divine punishment which they were destined to endure:

v. She had no idea that this was something that they can do thought it was a curse from god (TB15)

vi. His palate was operated when he was fourteen years old ..... it was taken as whatever was given by god (TB10)

9.8.1.2. Miscommunication

When a baby suffered from regurgitation during feeding, most families realised that there was a problem and sought intervention within a reasonable timeframe:

vii. She had nasal regurgitation whilst feeding so they got to know that there was a cleft in the palate so as soon as she was born they observed (GSR02)
The reasons for not seeking treatment for the CP were more complex, possibly because it was not visible and they didn’t realise treatment was necessary:

\[\text{viii. His family didn’t know ma’am they went for lip but don’t remember that more surgeries were needed (CH23)}\]

\[\text{ix. Were not aware that cleft [palate] can be treated (TB16).}\]

Some families were misinformed or misunderstood about when to return for the next phase of surgery: The parents of two participants were told to wait for nine months before palate surgery but misheard and returned nine years later. Another family understood they were to come back when their child was 10.

\[\text{x. Doctor said when he becomes the age of 10 years or so then bring him (CH20).}\]

The parents of two of the participants had been told that their infant was underweight and that the surgery could not be carried out at that time, not realising that the operation could be performed as soon as the child was fit enough, they never returned:

\[\text{xi. Delay in getting the palate repaired was because the child was underweight initially and they didn’t go back for surgery (CH07)}\]

\[\text{xii. They went to hospital but they [were] told that this is not the right time for treatment so they were waiting for the treatment to be done (GSR02)}\]

Another child had been taken for his lip surgery by his grandfather who later died without imparting the information to the parents that a further operation was needed for his CP. Consequently, it was only when they came across a cleft camp run by the hospital, when the child was already ten years old, that they realised that treatment was an option.

\[\text{xiii. his Grandfather took him for the treatment So after that his grandfather died and these people [his parents] they didn’t}\]
9.8.1.3. Inadequate and Inappropriate surgery

Unfortunately, for a significant cohort, when surgery was offered it was either unsuccessful or inappropriate: Two children had been operated on by general surgeons who had wrongly removed, rather than repaired, the premaxillary segment (ref back to introduction for explanation). For a few, the palatal repair had broken down completely leaving severe scarring and a fistula as large as the original cleft.

Xiv. He had a large palatal fistula which they attempted to fix several times age 17/18 but it broke down every time (TB19)

9.8.2. Finding out about treatment options for CLP

9.8.2.1. Chance meeting

For every interviewee, the discovery that appropriate surgery was available for their CLP was largely a result of fortuity: For a few participants a chance meeting provided the information which guided them to seek treatment:

Xv. They didn’t know about CLP and they met someone who said they could have something done in their own hometown actually (TB10).

9.8.2.2. Targeted advertising

For others the opportunity to seek help came about through targeted educational material produced by the cleft units: A few individuals spotted advertising notices or banners displayed in their own town or whilst travelling to Hyderabad for other business;

Xvi. [finding out about repair] I was in Hyderabad and I heard about it with advertisement (CH01)
Cleft camps

Others attended temporary educational camps organised and directed by the cleft centres specifically for this purpose. Such events involve a trained member of the team travelling to distant towns and remote villages with the sole purpose of educating schools, health workers and members of the community with regards to the availability of treatment for CLP. In due course transport was arranged to collect the patients and family members to bring them to Hyderabad for treatment.

xvii. Until 17 years she never had any treatment because they had no idea what could be done. at 17 years when hospital had conducted a camp this is when Sir first saw her and told whatever best can be done. (TB15)

xviii. after some time through a hospital mobile resident who goes for camps, through that person they got to know they realised that this problem could be treated and he got into this hospital last year (GSR02)

9.8.2.3. Spreading the word

A recurrent theme which emerged, was the participants’ active personal commitment to promote information about the availability of treatment in their own regions. They spoke quite passionately about this as a moral duty to help others with CLP and ensure that they would not miss out on receiving care at an earlier age.

xix. he tells them that they have to come to hospital and get it treated and the right place where to come (TB05).

xx. he makes sure he gets people in the village to the hospital to make sure they get operated in the correct place because he’s not faced many problems since the surgeries (TB18)

xxi. they make sure that they direct other people from their village to come across and get their surgeries done (TB07)
Such sentiments confirmed the degree of importance they gave to educational promotion as a key element in improving access to timely intervention for CLP.

9.8.2.4. Attitude towards treatment

Frustration

Parents often expressed frustration at not having being able to get good quality treatment for their child and parents spoke of their aggravation and disappointment about surgeries having needed to be repeated:

\[ xxii. \text{It was first repaired at some other hospital locally but it split open later so they went and asked why is this happening and all that they had a fight in the hospital and moved to another place, that was again not done properly and there was a fistula that formed (TB13) } \]

Uncertainty

Expressions of apprehension about the surgical options were particularly evident amongst those who had received treatment which had failed previously and there was an impression that they felt their experience was going to be a repeat of the last time

\[ xxiii. \text{So earlier when they came for treatment they were very sceptical... how much of improvement is going to happen...but now she is happy that at least things are improving (TB10) } \]

Hope

Once participants had become more informed, there appeared to be a more general sense of confidence in the treatment they were currently receiving:

\[ xxiv. \text{[She's] is happy coming to treatment knowing that she will get better with time (TB01) } \]
Regret

It was clear that the lack of understanding and the assumption that nothing could be done to manage the situation had fuelled a strong feeling of anxiety and isolation for some families. A few reflected badly or appeared resentful that their treatment had been overlooked and that they had not been informed about surgery earlier. For others there was a sentiment that they had let their children down resulting in an overwhelming sense of guilt and a feeling that they were somehow to blame.

xxv. What happened madam my family didn’t know anything about it they didn’t know anything about surgery... Now my family feel upset that they didn’t know to come for surgery (CH04)

Thankfulness

However, with respect to their current situation, there was a greater focus on looking forward with appreciation that the correct treatment was finally underway and that it was worth the inconvenience to be sure of getting the best treatment.

xxvi. I feel very happy for that I never thought it would happen (CH04)

xxvii. [They] are happy because the surgeon kept whatever promises he had made and kept his word and ensured that the treatment doesn’t cost anything (TB15)

xxviii. Although the travel is extremely difficult he would still not trade it for going elsewhere because of the result. He [father] has seen many kids once his son was under way correction but all of them for what he has seen there are really bad surgeries and he’s glad to be coming here because his son looks the best among the lot (TB07)
9.8.3. **Family impact**

Through talking with the participants and their families, it became clear that the impacts of CLP were not restricted to the participants alone: Indeed, the impact on their families was often quite marked; emotionally, financially and in terms of the time commitments required to obtain treatment.

9.8.3.1. **Parental concern**

Emotionally, apart from the aforementioned guilt, there was an overriding sense of worry and concern regarding their child's well-being: For younger children worries revolved mainly around the difficulty they had in making friends:

xxix. *They won't be able to understand and make with other friends so she will face that problem* [father] (CH01)

xxx. *[mother] …wants to join with friends but they wouldn’t understand* (CH02)

xxxi. *They cannot understand his speech so they won’t make him as a friend* (CH15).

For older children, parental anxiety tended to focus on marriage prospects, concerns which held true equally for both female and male children:

xxxii. *It’s her parents wanting which make her come for appointments [marriageable age]* (GSR01)

xxxiii. *Their parents think is will improve [for marriage] that is the only reason they are coming here* (TB01)

9.8.3.2. **Family finances**

**Out of pocket payments**

Although frequently referred to in the literature, lack of finance was not commonly reported as a primary reason for not seeking treatment although a number of families had suffered OOPs accessing intervention. Only one family, not realising that treatment was available free of charge, had paid for lip surgery and it was
their inability to raise the funds for the palate surgery which prevented them seeking further treatment:

xxxiv. financial problems in the childhood so that's why they pay some amount of money to get it done.... So they wait for after they are getting information about this free surgery so that's why they came.... (CH02)

Treatments offered at the three research sites in this study were all free of charge, however some families still incurred varying degrees of personal costs in relation to transport, subsistence and accommodation during surgery:

xx xv. financially it has been very difficult for them to manage but still they are trying to make it because there has been a difference in how she looks and interacts with everyone (TB15).

Loss of earnings

There were also compounding costs associated with loss of earnings, and although some employers were sympathetic, the expenditure and time involved in travelling to the cleft centres had serious impacts on some of the families, particularly those travelling great distances.

xxxvi. He's [father] a construction worker and whenever he accompanies his daughter he has to miss the days labour and he gets it in the end from his boss can't work for the entire weekend because maximum time where they get money so big cost involved in getting her for treatment (TB06)

xxxvii. a lot of travelling and a lot of time off work and time off school and time off for someone to bring them (TB03)

Although these financial and time constraints were difficult they were seen as necessary for the benefit of the child and parents did not attribute any resentment to their remarks on this topic.
Inability to contribute to household income

A third financial impact for families related to complications surrounding their child's employment. For one family their daughter struggled to help out in the family business until she had her surgery done:

xxxviii. Before her speech correction was done they have a shop and she sits and helps out in the shop. She did have a lot of problem in communication because the customers couldn't really understand what she used to speak, [surgery] has helped her quite a lot and her interactions with the public have definitely improved (TB04)

Although in rural areas several participants were able to find agricultural work from an early age, since communication skills was less of an issue:

xxxix. He's never been to school or college for his studies so from the beginning he was doing work [in agriculture] (GSR03)

xl right from his childhood he was working he seems he was in to farming (GSR04)

For others, poor communication precluded them from finding employment at all which, to their annoyance, dictated that they remained fiscally reliant on their family despite a desire to be autonomous.

xli. I want to make money but no one would employ me 'why are you trying job' [they would say] (CH04)

These perspectives indicate that delayed intervention for CLP can have long term and wide ranging impacts for families as well as the individual requiring surgery.
**QUESTION 2: IN WHAT WAY DOES UNTREATED CLP IMPACT THE ACTIVITY, PARTICIPATION AND WELL-BEING OF THE INDIVIDUAL AND HOW ARE THESE CHANGED BY INTERVENTION?**

### 9.9. ACTIVITY & PARTICIPATION

#### 9.9.1. Communication

**Comprehension by others**

Almost without exception the dominant issue which framed the pre-operative experiences of the participants was difficulties in communication. There was marked variation in the degree of its impact, for some the problem was relatively minor but for a few, verbal communication proved almost impossible:

\[\text{xlii. I understand but no one can understand. No one can understand what I am speaking (CH18)}\]

\[\text{xliii. [earlier] they couldn't understand anything what he spoke (TB10)}\]

For the majority, communication was not such a severe problem at home because the listeners were habituated to their speech, however talking with strangers proved more problematic:

\[\text{xliv. the family members we understand ...... but the people who are outsiders or the unknown people they cannot understand (CH20)}\]

\[\text{xlv. Only my mother and my brother could understand me apart from that that they had to do a very lot of concentration to understanding me (CH09)}\]

\[\text{xlvi. In his home they can understand but outside of his college they can't understand so much (CH13)}\]

Only one participant maintained that their speech had been clear from the beginning however, this individual had received relatively early intervention for their cleft palate and returned for treatment because of regurgitation through a residual fistula and alveolar bone cleft.
9.9.1.2. Improvement after surgery

Following surgery, a great number of the participants remarked that their speech and ability to be understood and communicate improved markedly:

xlvi. After surgery the first little bit the people don't understands what they are speaking (but) now it is everyone understands (CH12).

xlvii. He is much easier to understand and now he will talk with other children and make friends (CH07)

xlviii. In his school all can understand his language and his speech at this time previous they won't (CH14).

l. Before the palatoplasty unclear speech.. after the palatoplasty all can understand his language better (CH22)

It was very evident that participants were pleased with the improvement, even if they did not consider that their speech was perfect:

li. It's [speech] very much improved that I can say but previously it was very bad, many words which I can't pronounce, still now I can't pronounce letter K word it comes out A (CH09)

9.9.1.3. Anxiety

Anxiety about not being able to make themselves understood was the dominant factor which precluded many individuals from socialising. For some the intimidation was so concerning that they barely spoke at all, even in their own homes:

l ii. I can't talk in any language, and fear and hesitation. fear, fear, very much fear.. not many people heard me (CH04)

liii. She has a scary feeling ma'am (about speaking) (GSR02)

liv. Even at home also she used to restrict her speech (GSR01)
This was compounded by the diversity of languages spoken throughout the region and some participants found the additional anxiety of speaking a language in which they were not confident, was quite overwhelming. Many felt that they were setting themselves up for failure and this crushing expectation caused them to stay at home altogether:

_**Iv. He becomes anxious going outside because they may not understand him particularly if they speak a different language or dialect (CH10)**_

### 9.9.1.4. Reaction of others: Social stigma and bullying

Discussing the frustrations with communication prompted a range of feelings from the participants and the issue of social stigma quickly emerged: Participants spoke of being the focus of generalised questioning and teasing but they also endured social injustices such as direct criticism and public ridicule and several participants articulated feelings of stigmatisation and shamed for their CLP.

_**Ivi. The main thing is that if I speak someone will laugh on me they will mess with me and shame me (CH04).**_

_**Ivii. [before surgery] also they criticise him (CH15)**_

_**Iviii. They tease him is because of his speech and since he’s not able to pronounce some words correctly he’s pretty often teased for that (TB07)**_

### 9.9.1.5. Torment and seclusion

The insensitivity with which they were treated often caused anxiety, fear and hesitancy when speaking, causing intimidation, shyness and inhibition:

_**Ivx. before operation that’s what he wasn’t much comfortable going out meeting people or talking to them because they used to upset him (GSR03)**_
lx. Had friends yes ma'am but sometimes used to laugh because they didn't understand so a little afraid to speak (CH23)

lx1. Make her a little bit shy talking to strangers (CH16)

9.9.1.6. Aggression

Some participants even reported episodes of feeling aggressive towards people who bullied them, sometimes, particularly with younger boys, to such an extent that it occasionally even prompted physical violence:

lxii. we have a few other kids who have beat the ones who have asked (about cleft) (general comment by cleft team member)

lxiii. Before he was always hiding or fighting (CH07)

Experiencing such difficulty in communication made it very difficult for many individuals to cope with normal social activity. This is clearly demonstrated in the case of TB10's described in Vignette 1. His levels of distress and anxiety caused such feelings of depression that he became a recluse and almost non functional, refusing to go out or interact and causing great anguish for his mother:
Vignette 1: TB10

When he was born, in a rural part of Telangana, his family thought his cleft was due to the solar eclipse. Although his lip was repaired when he was young the family didn't realise the palate could be repaired so his speech developed badly and his nasal regurgitation was severe:

_Earlier when his palate was un-operated there used to be nasal drainage from the nose food always entering one cavity or the other and they couldn’t understand anything what he spoke._

Because of this he was teased and ridiculed and refused to go out and socialise eventually withdrawing from all community activities:

_He is sort of quite depressed with whatever they ask him so he never really reacts to anything._

Aged 14, he came to the hospital for treatment but the family were very sceptical that it would make a difference. Fortunately their worries were unfounded and his story is an excellent example of the positive impact that intervention can have even at a late stage:

_They can notice a tremendous difference after surgery because everybody used to make fun of him earlier and once his speech started stabilizing 2 months after surgery the questions and comments have stopped almost._

9.9.1.7. Social support mechanisms

Although some participants suffered profound distress others appeared to have developed better coping strategies despite encountering similar confrontation, it quickly became apparent that in these cases participants reported a solid social network of support including family, school and local community which gave them confidence despite their CLP.

_lxiv. in both the schools he didn’t face much difficulty. He used to talk to his friends and all and no-one teased as such so it was ok with his speech (GSR05)_
lxv. Her speech and nasal regurgitation never stopped her going out with friends (CH09)

lxvi. Friends and family been supportive never really teased at all (TB08)

lxvii. She didn’t suffer any teasing in school because everyone was patient (CH01)

The clearest example of social acceptance was expressed by GSR06 (Vignette 2) whose case demonstrates that it is more often people’s perception and reaction to it which causes the distress rather than the CLP directly:

**Vignette 2: CSG06**

This participant attended for treatment aged 33. She was an only child and had never received intervention because her parents thought the cleft was the wish of god and they were scared she might die if she had an operation. She has always been extremely difficult to understand and only a few people recognise what she is saying, however, she has always made herself understood by other means and has always been loved and accepted in her village. She has plenty of friends and is married with 2 children.

*Even though her physical appearance and everything she’s not much bothered about it. She has very good number of friends and she talks to everyone.*

She has developed a technique of eating which means that she can usually avoid nasal regurgitation so her condition does not really bother her at all:

*Even eating wise also she doesn’t have much of problem she’s very happy eating the way she is. She’s managing since a long time so she’s used to it So she’s developed a way of making it work [eating]*

They were told that surgery may improve her speech, so the only reason they decided to come for treatment was because her relatives could never understand her and her husband thought that, if she had a repair, he would no longer have to translate for her all the time.
9.9.1.8. Impact of intervention

The overwhelming conclusion from all the participants was that intervention had brought a positive outcome regarding their ability to communicate.

**lxviii.** Now everybody getting understanding and he is good life, big differences are there (CH10)

The majority conveyed that they were no longer too shy to address strangers and that their enhanced confidence had encouraged them to socialise:

**lxix.** Since the palate surgery and a little speech therapy his speech has improved he has become bolder and will now talk (CH07)

**lxx.** After the operation what she observed was she started speaking with everyone comfortably (and) she was interactive with them (other students) after the palatoplasty was done (GSR01)

**lxxi.** But after surgery he himself felt very comfortable with his speech and he started going out very frequently and meeting new friends and all that (GSR05).

For a few however, their shyness in public persisted post-surgery despite feeling more confident with their speech:

**lxxii.** [field note] Boy’s speech very good but reluctant to speak - looked to mother to do all the talking (CH02)

**lxxiii.** Now is very much clearer and most people understand her if it is quiet ... but still shy of new people in case they won’t understand (CH18)

9.9.2. Activities of daily living

The newly found self-assurance took many far beyond just socialising with friends, it opened up the opportunity to carry out ordinary activities which until then had previously been impossible.
9.9.2.1. Using the phone

Hyderabad is a city which pioneers electronic technology, yet for this cohort speaking on a mobile phone was simply not feasible until after surgery was completed.

lxxiv. Even the family members when he speaks over the phone it's hard to understand some of the words (CH20)

lxxv. Before palate surgery 2016 she could not use a telephone now everyone understands (TB08)

lxxvi. After that time I got my palate repaired so I felt very confident on that then I used to talk and that (CH09).

9.9.2.2. Independent shopping and travelling

Even simple everyday activities involving public encounters were fraught with difficulty for many of the participants. The struggle with communicating whilst shopping or trying to catch a bus filled them with trepidation and frustration. Post surgery all participants reported that their increased confidence in communication allowed them to undertake these daily tasks with relative ease:

lxxvii. Even shopkeepers when she goes to a shop she is very much comfortable talking with them, she is not shy at all it seems (GSR02).

lxxviii. Now everyone understands her, even when she speaks on the phone and she has friends and is able to participate in shopping and travelling (CH01).

lxxix. As of shopping and local transport as of now they don't really have any issues - they are able to manage on their own (TB02)

9.9.3. Regurgitation

The second most common topic of discussion was the issue of nasal regurgitation. Although a few they were able to overcome the problem, and did not allow it to
impede their lifestyle, over two thirds of the participants indicated it to be problematic to varying degrees:

lxxx. Nasal regurgitation was a problem but she learned to avoid it by eating carefully but it was embarrassing (CH01)

lxxxi She had some nasal regurgitation and problems eating and drinking ........People always accepting so no problems socialising.

For a number of participants however, the amount of teasing about regurgitation was so great that they preferred not to socialise or eat in front of strangers at all:

lxxxii. Food was getting stuck [in the cleft] and liquid coming from here always [points to nose], People would laugh with him eat so he didn’t like it (CH23)

lxxxiii. It became very problem for him to eat also he had lots of nasal regurgitation, because it was totally open he had lots of food sticking onto his cavity and onto the palate ... that’s why he wasn’t much comfortable going out meeting people or talking to them because they used to upset him (GSR03)

Fortunately, the regurgitation stopped for most of participants following primary palate surgery, and with it the teasing from those around them.

lxxxiv. When it came to eating she used to get a lot of flak for that, but things have become a little better {post surgery} (TB12)

lxxv. [before surgery].whatever they eaten they get regurgitation [after] now it has improved (CH02)

For a few unfortunately, the oro-nasal fistula remains open and regurgitation continues to be a problem as described for TB15 (vignette 3)
Vignette 3: TB15

This 20 year old girl travelled a distance of 800km to reach the treatment centre for each of her appointments. When she was born the family had thought she had been cursed by god and did not realise the CLP could be treated. She was taken for her lip surgery aged five then the palate was partially closed when she was seven but there was still a large fistula overlying the bony defect in the palate as can be seen in the radiograph and photographs below.

Her speech was really difficult to understand and although her family could make out most of what she said, she could not make herself understood by strangers at all. She was extremely anxious about her appearance and suffered badly with nasal regurgitation causing her to avoid socialising due to the amount of teasing and the frustration she felt at not being able to make herself understood.

Only when she was 17 were they told, at a cleft camp, that further treatment was an option and that the situation could be improved. She was of marriageable age when she first attended so it was agreed that the surgery on her nose would take a priority. Now that her treatment is underway her speech has improved a little although regurgitation is still a problem to her because of the residual fistula:

Before the operation there was a lot of teasing, eating and drinking is still a problem because of the fistula there is fluid exchange between her nose and mouth it makes things difficult.

She knows she has more surgery ahead but now that her nose and her teeth are being corrected and her appearance has improved so much, she finds she can smile with confidence and no longer hides away from the mockery she endured before surgery.
9.9.4. Medical and dental problems

Participants alluded to a number of medical issues: Reference was made to ear infections, throat infections, breathing difficulties and dental anomalies, which would be expected amongst this cohort (Sheahan et al., 2003, Goudy et al., 2006, Smallridge et al., 2015, Hocevar-Boltezar et al., 2006).

lxxxvi. Breathing is the major concern, speech is second (TB10)

lxxvii. He is getting a lot of colds, fever [before surgery], now he doesn't have anything. so a big difference (CH10)

lxxviii. [post surgery] No ear infection or deafness or any of those issues, dental pain was there earlier but since she's coming for regular treatment now those issues have been sorted out

Of these it was the dental problems which were of most concern to them. As described in Vignette 3 dental derangement and ectopic teeth can be a major concern indeed and a number of participants reported bullying. For one girl it was actually her missing anterior teeth which prevented her speaking rather than problems with speech:

lxxxix. She [mother]is happy that his teeth are being set finally because he's had a lot of bullying because of it earlier (TB10)

xc. I used to hesitate talking because I had no teeth here, from here to here so what shall I do (CH09)

xci. Actually before I did not have any teeth this side or this side so they have fixed the teeth. Even the look also it has been changed, 80% it has improved (CH11)

Some observations appeared to be heavily influenced by cultural ideals, with participants and their families reflecting on how important aesthetics were in terms of marriage prospects. This topic emerged as the social norm equally for both genders and was highlighted by one case where both bride and groom had been born with CLP.
xci. With dental and orthodontics to make it look better then she will be happy for marriage (CH10)

cvii. [for marriage] Now he is grown up so the teeth corrections and upper lip corrections are need to do.

cviii. The only thing is he is worried about his physical appearance because right now it the age for marriage, So because of that he wants to get the treatment done (GR05)

For some participants the complex positioning of ectopic teeth was an extreme functional issue: Figure 18 demonstrates the extent to which the alignment of the dentition was affected for one participant: Clearly her facial aesthetics are not a problem but dental function was significantly compromised in terms of eating and speech.

Figure 18: Participant with ectopic teeth associated with CLP

9.10. WELL-BEING

9.10.1. Role in society

Almost all the participants had experienced some social setbacks and been exposed to attitudes of contempt in relation to their CLP and a brief mention was also made in section 9.8.3 relating how untreated CLP affected peoples’ ability to
make a living. This restriction in living a self sufficient, autonomous existence was considered a key factor causing distress for a number of the participants and one of elements which made the biggest transformation when it was overcome. The detrimental effect of such heartless mockery and rejection on participants’ quest for acceptance and independence is succinctly expressed by one man as he described his persistent search for employment (Vignette: 4)

**Vignette: 4: CH04**

This 27 year old man had been born into a rural family who were unaware of treatment options for his isolated CP. As a child and young man he was very difficult to understand and those around him would laugh and mock causing him to shy away and remain silent. He repeatedly tried to find work but nobody would employ him:

*I want to make money but no one would employ me ‘why are you trying job’...and for that reason. ’Til 2005, I’ve not enjoyed any job*

*when I had gone for job before they would react to me they would laugh on me they told me and they feared me that way it has been very bad for me.*

Finally, when he was 26, he got a job as a porter in the hospital in Hyderabad. One of his colleagues approached him and suggested he attend the cleft department to see if they could help him and from that day his life was changed:

*One fine day they called on me ‘we are going to do the surgery in this hospital’. I go for the surgery and I am very happy for that. I want to tell you madam I am really happy for the result. I never expected it would happen. Many friends laughed at us. I never expected, but it has come.*

In describing his new life it is impossible not to be struck by his level of pride and positivity as he looks to the future. In addition to his work as a porter he assists in educating the public about CLP and helping other families with the same condition, a role he would never have dreamed of before his surgery.

He is rightly extremely happy with the result and summarised in his own words - at last he is *living life*. 
9.10.2. Psychological impact

It is clear from the descriptions, quotes and vignettes which precede this section, the extent to which participants were traumatised and marginalised as a direct result of having untreated CLP. Although many participants developed coping mechanisms and had supportive friends and family, the recurrent use of emotional terminology such as; teasing, ridicule, criticism, embarrassment, fear and humiliation throughout all but a few testimonials, is clearly indicative of the distress they had experienced pre-operatively. It is notable that post-surgical comments relate more to levels of confidence and happiness with clear indications that life was easier and more fulfilling.

xcv. [before surgery] friend also the criticise him So his friends are now talking and not criticise him (CH15)

xcvi. So now shopping and things it’s made life easier (CH01)

xcvii. Decent ability to make friends and mingle out in public with no other major issues that they are facing.

xcviii. As of shopping and local transport as of now they don’t really have any issues - they are able to manage on their own

The fact that participants perceived that being subject to such negative attitudes pre-operatively was the source of their disquiet and marginalisation helps to explain their levels of elation post-surgery when the mockery stopped.

Whilst the participants' reflected on the fact that surgery had benefitted their lives significantly in terms of self respect, making friends and socialising, it was also possible to detect a bigger change in their life experiences. Being privy to the life stories of these participants it was apparent that the benefit of their inclusion in society was not merely personal, there was also a related social advantage in terms of autonomy and self reliance. One case demonstrates this better than all others (Vignette 5) revealing how a relatively simple medical procedure can transform an
isolated and intimidated child into a confident, independent young woman with high flying ambition and expectations:

**Vignette 5: GSR01**

This 19 year old girl had travelled from a village 300km away. Her lip had been repaired at 9 months but the first palate repair, which she had had at 6 years old, had failed, leaving her with a large palatal fistula. At this time she would not socialise with anyone, she did not have any friends and she was embarrassed to eat or speak even in front of the family:

*Before the palatoplasty was done she was very uncomfortable with her speech even for eating also. She never used to mingle with the new people even at home also she used to restrict her speech.*

She attended school but was painfully shy, not interacting with the teachers or any of her peers.

*She was very much afraid to her class mates and even the teachers to answer the questions whatever they’re asking*

Until she was ten, the family didn’t realise anything could be done to fix the palate. By chance her father found out about the cleft centre, and immediately sought advice. The family report that the transformation was amazing:

*After the operation she was comfortable talking to her classmates as well as with teachers... she started speaking with everyone comfortably, she did her schooling, she did her pre-university... also she got to meet new people, new friends.*

Meeting this happy, smiling and confident young woman today, flanked by her proud parents, it is hard to imagine the shy and lonely ten year old she once was. Her speech and appearance may never be perfect but without the surgery she would probably have remained disillusioned with her ambitions thwarted by her low self esteem. Now she is living her dream, fulfilling her aspiration to study at university and to live a full and happy life surrounded by family and friends.
9.11. Pre-operative and post-operative comparison

Comparing the results across the framework, there did not appear to be any particular clustering in relation to the different elements within the themes and subthemes: Comments on aesthetics, marriage prospects, socialising and making friends occurred equally amongst all ages and both genders. Individuals from both rural and urban backgrounds appeared to have equal difficulty in accessing information about CLP and communication problems, regurgitation, aesthetics and experience of bullying affected the majority of participants to a similar degree.

Clear differences were only identified in the framework data after separating the pre-operative and post-operative components, at which point an obvious demarcation between negativity and positivity emerged. Although it is acknowledged that the sampling methods of qualitative research are not conducive to quantitative analysis, and that comparing relative frequencies of responses can sometimes be misleading, graphic representation of data can be a useful means of visualising characteristics in the data (Pope et al., 2000, Yvonne Feilzer, 2010, Hancock et al., 1998).

Figure 19: Comparison of comments pre-operatively and post-operatively demonstrates the number of positive and negative comments which participants made in describing their experiences before and after treatment. Irrespective of the numerical values, and without claiming statistical relevance, this graphic tends to indicate a more positive general attitude physically and emotionally post-surgery.

Participants appear to manifest a marked improvement in their speech and suffer less regurgitation issues post-operatively. Additionally, a key element demonstrated in this figure, is the strikingly more positive attitude individuals have when describing aspects of their experiences relating to independence and well-being. Every comment in relation to work is positive post-operatively, very few comments relate to negative emotions and the degree of teasing has reduced
and importantly, it would appear from this comparison that self confidence and independence improve markedly for this cohort following intervention.

Figure 19: Comparison of comments pre-operatively and post-operatively

![Graph showing comparison of comments pre- and post-operatively](image)
As mentioned in section 9.1, the data collected by the cleft centre staff using the app was prospective whereas the TOM-clp scores derived from the interviews compared the current situation with a retrospective opinion of the participants pre-operative state. The two data sets are therefore not comparable and will be discussed separately.

The TOM-clp is designed to be used as an outcome measure for use in audit, and benchmarking as well as to demonstrate change on an individual patient basis. Therefore, in addition to a general analyses of the data, vignettes will be presented as a means of demonstrating how the TOM-clp score might be used as an outcome measure at an individual patient level.

### 9.12. Usability of the TOM-clp

Circumstances prevented the collection of formal feedback from everyone involved in collecting data for this study, so the following relates to two semi-structured interviews, one e-mail response and collated information from informal discussion during the field trips. In order to differentiate between cohorts involved in this study, for the remainder of the thesis the participants implementing the TOM-clp will be referred to as ‘clinicians’ and the interviewees as ‘CLP participants’.

Clinicians found the TOM-clp 'straightforward to understand’ and felt the training was 'helpful and sufficient'. Almost all the clinicians were able to implement the measure reliably after a single training session and only one person required additional training.

Anecdotally the clinicians reported as follows:

- The TOM-clp integrated easily into regular clinical activity as it took no more than 5 minutes to employ.
- Although there was a prompt sheet available, once they had got used to the process they didn’t need it because there was 'nothing [specific] to remember'.
- As it is not paper based it did not require additional resources, which in the views of the clinicians, was valuable on three counts: It could be carried out
anywhere, so they didn’t have to remember to take paperwork with them into every clinic room; It would be great during cleft camps because with the TOM-clp there was ‘nothing to carry’ and it wouldn’t get ‘muddled up’ or ‘lost’ which could easily happen with paperwork; there were no printing costs.

- It could be undertaken in any language: All of the clinicians were multilingual and were used to conversing with patients in a number of different languages. However, there were some local languages and dialects in which they could converse but could not accurately translate a formal questionnaire. They considered the conversational nature of the TOM-clp gave it an advantage over a more prescriptive outcome measure, such as the WHO-QoL-BREF, in this respect.

- It could be used irrespective of age or level of literacy: Clinicians commented that many of their patients had low levels of literacy or would be too young to understand or complete a questionnaire. This was borne out with the WHO-QoL-BREF which then had the additional problem of proving prohibitively time consuming to translate verbatim for each participant.

- It was simple and could be used in the same way for everyone. One clinician described it as 'low tech and culturally appropriate'.

- Clinicians and CLP participants also commented that the process was relaxed and not intimidating: One clinician remarked ‘the patients were really relaxed because it didn’t feel like an interview’. Along similar lines a CLP participant commented that she enjoyed ‘chatting’ and that she had been a bit nervous about the process because she had been expecting some ‘difficult questions’ which she was worried she might not have understood.

- Clinicians reported that the data collection app was straightforward to use, and convenient as it could be completed on any hand-held device.

- Inputting the demographic data was considered a bit time consuming even though it only took an additional five minutes. Whilst participants appreciated that this element would not be required on subsequent visits, it was suggested that, since this part of the process did not require calibration, that the demographics could be completed by a different team member, if clinic time was pressing.
9.13. App data

Owing to the early cessation of data collection in Assam, all the TOM-clp data was pre-operative. It was therefore not possible to make any assessment as to the outcome of intervention or compare it with the Hyderabad outcome data, as had been the intention. Consequently the pre-operative app data from all three sites was amalgamated and used to explore the impact of untreated CLP prior to intervention.

9.13.1. Demographics

Data was available for 43 cases across the two sites: 31 from Assam and 12 from Hyderabad. Ages ranged from 6 to 35, represented according to cleft type and severity as outlined in Figure 20.

*Figure 20: Cleft classification and size of cleft for pre-operative cases (n=43)*

![Cleft classification and size of cleft for pre-operative cases](image_url)
9.13.2. **TOM-clp data**

The average TOM-clp scores, presented in Figure 21 show that overall, the cohort presented with severe impairment associated with a moderate impact on their activity, participation and well-being. The TOM-clp scores were very particular to each individual participant and there did not appear to be any clear correlation between size of cleft or type of cleft, (represented by the impairment domain) with the other three domains; This is clearly demonstrated by the moving trend line in Figure 21, where the data is sorted in relation to the activity domain: Whilst the figure indicates a general corresponding trend with activity, participation and well-being the impairment domain is completely erratic and does not correlate. This represents a potential link between communication, regurgitation, participation and well-being which is independent of the size or type of cleft.

This is also demonstrated by the fact that 37% of the participants scored higher than 4 in all three of the domains activity, participation and well-being, the same cohort had impairment scores ranging from zero to four. This indicates that some individuals with profound impairment had near normal participation and well being.

**Figure 21: Pre-operative TOM-clp scores from app with moving trend lines**
A score of 5, indicating normal function, only occurred four times in the entire data set: Once in both participation and activity for the same participant who had with severe UCLP and the twice for participation in participants who scored zero under impairment for their profound BCLP.

Nobody scored 5 for well-being but 79% of the participants scored 3 or more in both well-being and participation, indicating that CLP had no more than a moderate affect on these participants. Conversely 12% scored 1.5 or less for well-being and participation indicating extreme distress and isolation in this cohort.

9.13.3. Interview data

The average TOM-clp scores for the all the interview cohort are presented in

Table 9:1 however data was only included in the comparative analysis where participants had undergone at least one operative procedure for their CLP at the centre they were currently attending (n=31). In two instances the information from the interview was not considered adequate to provide a TOM-clp score; once for activity and once for well-being. For this reason n=30 for these two domains.
Table 9.2: Average pre-operative and post-operative TOM-clp scores

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<tr>
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<th>P</th>
<th>W</th>
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9.13.4. Analysis of average TOM-clp scores

The average TOM-clp scores are summarised graphically in Figure 22 and indicated marked improvement in all four domains.

**Figure 22: Pre-operative and post-operative TOM-clp scores comparing**

<table>
<thead>
<tr>
<th>Domain</th>
<th>Pre-operative average</th>
<th>Post-operative average</th>
</tr>
</thead>
<tbody>
<tr>
<td>Impairment</td>
<td>0.7</td>
<td>3.6</td>
</tr>
<tr>
<td>Activity</td>
<td>1.8</td>
<td>4.3</td>
</tr>
<tr>
<td>Participation</td>
<td>2.1</td>
<td>4.7</td>
</tr>
<tr>
<td>Well-being</td>
<td>1.9</td>
<td>4.5</td>
</tr>
</tbody>
</table>

The significance of these changes were assessed by applying a repeat-measure paired t-test (2-tailed) using Microsoft® (MS)Excel 2007. Descriptive statistics and T-test results are outlined in Table 9:3. The p value of 0.0001 indicates that there has been a significant change following intervention in all four domains. The negative t value indicates that these changes are positive.

The standard deviation (SD) is large in the activity, participation and well-being domains, indicating that impact of CLP varies widely between individuals; this is less noticeable in the post-operative data than the pre-operative data. SD is smaller in the impairment domain both pre-operatively and post-operatively indicating a lesser diversity in these groups.
Table 9.3: Repeat measure T-test results comparing per-operative and post-operative TOM-clp scores

<table>
<thead>
<tr>
<th></th>
<th>Pre-operative Impairment</th>
<th>Post-operative Impairment</th>
<th>Pre-operative Activity</th>
<th>Post-operative Activity</th>
<th>Pre-operative Participation</th>
<th>Post-operative Participation</th>
<th>Pre-operative Well-being</th>
<th>Post-operative Well-being</th>
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<tr>
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<td>30</td>
<td>30</td>
<td>30</td>
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<td>-8.4</td>
<td>-8.3</td>
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<td>29</td>
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<td>Significant</td>
<td>Significant</td>
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All individuals included in the case mix had undergone surgery for a large palatal defect however, a number of individuals were having this procedure done as secondary surgery following gross surgical breakdown. In addition, 12 of the cohort had received an alveolar bone graft and/or a palatoplasty in conjunction with the secondary surgery. Statistical analysis comparing these variables showed no significant difference in outcome between the groups in any domain.

Mid range scores, where a participant had already received two different interventions, were available for activity, participation and well-being for five individuals (Figure 23). The average scores demonstrate a progressive improvement at each stage. The impairment score was only available pre-operatively and post-operatively for this cohort so no mid point is demonstrated in the figure.
9.13.5. Descriptive analysis

As indicated in section (9.13.2) the TOM-clp scores are very personal to each individual as demonstrated by two extremes: GSR03 had the highest TOM-clp score for activity yet scored zero for participation and well-being. In contrast GSR06 scored zero for impairment and Activity yet showed normal participation and well-being.

90% of the cohort had presented with a cleft greater that 1/2 of the hard palate. Post-operatively 80% had complete palatal closure but all had some residual defect.

More than two thirds suffered severe or profound problems with activity pre-operatively (TOM-clp<2). For post-operative activity only one participant reported severe difficulty, two thirds were functioning above TOM-clp 4, indicating only a mild difficulty and 16% were considered to be functioning normally.

Just over half the participants suffered severe or profound difficulty in participation pre-operatively (TOM-clp<2) and the same individuals scored severe or profound in the impairment, activity and well-being domain. Post-operatively 90% of the participants reported only mild restrictions in participation (Tom-
clp≥4), however the same 90% scored over 3 in the well-being domain, indicating their CLP still had at least a moderate impact following intervention.

In total 50% of the participants scored 5 for well-being post-operatively, of which five had scored less than 0.5 pre-operatively in both participation and well-being.


Four vignettes will be presented in the following section. The first three cases were chosen as, whilst they are not atypical of the general cohort, the changes in TOM-clp scores clearly reflect their case histories. In doing so they provide an insight into how the TOM-clp might be used in clinical practice to assess outcome following surgery.

The forth Vignette is presented as anecdotal indicator of how the TOM-clp might reflect changes relating to intervention other than surgery.
**1. GSR1: Average TOM-clp scores reflecting the testimony described in each domain**

<table>
<thead>
<tr>
<th>Domain</th>
<th>Pre-operatively</th>
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</tr>
</thead>
<tbody>
<tr>
<td><strong>Impairment</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Activity</td>
<td>Her family understand her but not strangers and everything she eats or drinks comes down her nose</td>
<td>Palatal closure was complete but short</td>
</tr>
<tr>
<td>Participation</td>
<td>She doesn’t like to leave the house and will not actively participate in school because people will laugh at her. She does not want to continue education and won’t eat in front of anyone because everything comes down her nose</td>
<td>She will talk with everyone, including strangers, even if they don’t understand her, and happy to go shopping and is confident mixing in school and plans to go to university and get married</td>
</tr>
<tr>
<td>Well-being</td>
<td>She is shy and not happy with life at all</td>
<td>She is happy and confident</td>
</tr>
</tbody>
</table>

**Impairment:** She presented with a complete cleft palate following surgical breakdown of the palate undertaken previously, as a single procedure with lip repair in infancy which scores a zero in the impairment domain pre-operatively and 4.5 following successful repair.

**Activity:** She had severe difficulty in communication which is reflected by the TOM-clp score of 2 in the Activity domain. This scores 4.5 post-operatively indicating that communication and/or regurgitation are only a mild problem.

**Participation and well-being:** Before surgery she would not socialise or eat in front of anyone and was profoundly miserable hence scoring 0.2 for both Participation and well-being. Post-surgery she scores 5 in both these domains indicating normal social functioning and well being following intervention.
Vignette 2: CH04

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</thead>
<tbody>
<tr>
<td>Impairment</td>
<td>He had a complete isolated untreated isolated cleft palate</td>
<td>Following primary surgery he has a small residual fistula and a shortened palate</td>
</tr>
<tr>
<td>Activity</td>
<td>Nobody can understand him and people laugh at him when he tries to communicate but never has any problem with nasal regurgitation.</td>
<td>People can understand him 50% of the time and many people can understand his speech if he speaks slowly</td>
</tr>
<tr>
<td>Participation</td>
<td>He has no confidence and is scared to go out and rarely speaks at all. He cannot get a job so has to rely on his family to look after him</td>
<td>He will try and speak even if they may not understand, he has a job, he enjoys going out with friends and prepared to mix and speak with strangers in quiet environments</td>
</tr>
<tr>
<td>Well-being</td>
<td>He was always afraid to speak, and extremely miserable</td>
<td>His confidence has improved and he is happier with life although he gets frustrated with his communication and hopes it will improve once treatment is complete</td>
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</table>

**Average TOM-clp score**

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<tr>
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<th>3</th>
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<tbody>
<tr>
<td><strong>CH04</strong></td>
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<tr>
<td>Impairment</td>
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<td>Activity</td>
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<td>Participation</td>
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<tr>
<td>Well-being</td>
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</tbody>
</table>

**EXPLANATION**

The Toms scores of zero in all domains pre-operatively reflect the profound effect of CH04’s CLP. Following his repair he has less of a defect and scores 3 for impairment and still only scores 3.2 for activity post-operatively. Despite this is participation and well-being are normal for someone of his age indicating a good quality of life. Since regurgitation was never an issue the activity domain is based on his speech so these scores indicate how a small improvement in communication can have a marked affect on participation and well-being. Had speech been the only measure of outcome for this participant the intervention may not have represented the full benefit on the psychosocial well-fare of CH04.
Vignette 3: GSR06

<table>
<thead>
<tr>
<th>Pre-operatively</th>
<th>Impairment</th>
<th>Activity</th>
<th>Participation</th>
<th>Well-being</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>She has a severe untreated bilateral CLP</td>
<td>Her husband is the only person who can understand her and if it wasn’t for an unusual trick she has learned everything would come down her nose when she eats.</td>
<td>She has plenty of friends and is the life she is married with children and is the life and soul of all village events</td>
<td>She loves life and is completely happy with everything</td>
</tr>
</tbody>
</table>

Average TOM-clp scores

**EXPLANATION**

GSR6 had untreated BCLP which scores a zero in the impairment domain. She has profound difficulty in communication which is reflected by the TOM-clp score of 0.6 in the Activity domain. However, her social functioning and well-being are excellent and normal for a person of her age. This is clearly demonstrated using the TOM-clp but would have meant missed if GSR6 was assessed using perceptual speech analysis alone. Unfortunately there is no post-operative data for this participant as she was awaiting surgery at the time of data collection.

Vignette 4 represents an 18 year old male whose lip was treated at 4 years. Aged 17 he had several procedures to attempt closure of his cleft palate, all of which broke down, so he was given a plate to cover the fistula, which helped his speech and reduced regurgitation. He has now been advised to stop wearing the plate, while he undergoes orthodontic treatment to expand his arch. Later he will have a bone graft and palate repair, but the plate is damaging the soft tissue and will reduce his chances of a successful repair.
### 4: TB18: Average TOM-clp scores reflecting the testimony described in each domain

<table>
<thead>
<tr>
<th>Time Period</th>
<th>Impairment</th>
<th>Activity</th>
<th>Participation</th>
<th>Well-being</th>
</tr>
</thead>
<tbody>
<tr>
<td>Before any surgery</td>
<td>He never had too much problem being understood and most people could understand him if he spoke slowly but he had a great deal of problem with nasal regurgitation everything he ate came down his nose</td>
<td>He never stopped his CLP from stopping him going out and meeting people and has a good job</td>
<td>He never stopped his CLP from stopping him going out and meeting people and has a good job</td>
<td>He was generally happy but had periods of distress because of his nasal regurgitation</td>
</tr>
<tr>
<td>Whilst wearing plate</td>
<td>The fistula decreased by about half after his initial surgeries</td>
<td>His speech and regurgitation were better because he was wearing a plate to cover the fistula</td>
<td>He never stopped his CLP from stopping him going out and meeting people and has a good job</td>
<td>He was much happier and confident wearing the plate only rarely getting upset</td>
</tr>
<tr>
<td>Since orthodontics started</td>
<td>His fistula has increased in size</td>
<td>He thinks his speech has also changed and he 'sounds like he is talking through his nose' which he did before his palate surgery.</td>
<td>He gets embarrassed going out at the moment because his speech and regurgitation is worse</td>
<td>He is a bit embarrassed and less confident since the orthodontics started but he is hoping things will improve once he has his surgery and his teeth are fixed.</td>
</tr>
</tbody>
</table>

Comparing the three points in time, TB18 shows an initial improvement in the impairment domain which reflects the surgery but this drops once the fistula opens up with the start of orthodontics. A similar trend is shown in the activity domain in relation to regurgitation and speech. His participation was normal until he started the orthodontic treatment when he became slightly more reclusive owing to embarrassment with regurgitation whereas his well being became normal after his initial surgery and cover plate but has reduced since the current treatment reflecting his embarrassment about his current speech and regurgitation.
CHAPTER 10: DISCUSSION

10.1. Aim 1: Identifying the reasons why CLP remains untreated

Question 1: What facilitates or limits accessibility of cleft care, what services are currently available and how are they promoted?

The study by Naram, Makhijani et al. (2013), undertaken at one of the Hyderabad cleft centres, indicated that poor education, superstition and a lack of awareness remained the key reasons for patients failing to attend for treatment at an optimal time. This is borne out by the testimonies of 75% of the participants in this study, who revealed their principle reason for treatment delay was a lack of knowledge. This also mirrors the findings of older studies and indicates that a similar trend has continued unchanged for over a decade (Weatherley-White et al., 2005, Loh and Ascoli, 2011, Oginni et al., 2010). However, in contrast to these earlier publications, only a few of the participants in this study felt there was a monetary component preventing them seeking help. This discrepancy may reflect the financial support offered to patients in relation to travel, subsistence and accommodation which may not have been available previously.

The participants in this study generally reported that they were seeking to improve their appearance and speech in the hope of bettering their educational, employment and marriage potential, which mirrors the testimonies given in the literature (Singh, 2009, Raju, 2000, Mednick et al., 2013, Weatherley-White et al., 2005). Another reason, elucidated in this research yet less well reported in the literature, was the request for corrective surgery following failed interventions carried out elsewhere. This held true for the 30% of the participants, who believed that further intervention was not an option, thus delayed seeking help despite residual problems. This supports the voiced concerns of the clinical directors regarding some of the poor quality of care being delivered at certain other centres in their region and, they presumed, across India.

Improving global standards of care for CLP has been a common goal for several decades and has led to guidelines being produced to regulate treatments and improve quality of services: In 1998 the CSAG report recognised that outcomes for
intervention tended to be compromised where small centres were working in isolation. They suggested the gold standard of care should comprise centralised centres of excellence, offering comprehensive care by multidisciplinary teams including a primary and secondary cleft surgeon, an orthodontist, a SLT, a paediatrician, an ENT/audiologist and a specialist cleft nurse, with associated support from a psychologist, a restorative and paediatric dentist and a geneticist (Epstein, 2014, Scott et al., 2014). Comments from the clinical directors in this study suggested that many cleft centres were not heeding this advice and consequently compromising quality of care which they considered problematic on several counts:

- Firstly - unsuitable surgery, being undertaken by inappropriately trained clinicians, was causing patients harm and compromising achievable outcome. This was confirmed in this study where two individuals attended the clinic having had their pre-maxilla completely removed as an infant, by a surgeon who knew no better, which highlights the need for greater regulation of service provision.

- Secondly - if outcomes are poor, patients may be discouraged from seeking surgical correction in the future and potentially deter others from seeking treatment (Shrime et al., 2015).

- Thirdly - clinicians spoke with disquiet regarding the disparity in experience of operators and modes of surgical delivery at different locations around the country: All the centres included in this study offered comprehensive and cohesive care and were run by resident multidisciplinary teams with high levels of experience and expertise. Concerns were twofold: With local and visiting surgeons operating with inappropriate levels of expertise and inadequate supervision, and with short-term surgical missions which deliver high volumes of work yet taking no accountability for poor surgical outcomes. As alluded to in chapter 5, these concerns are echoed in the literature, highlighting that even where overseas surgeons are experienced, their complication rate is increased when operating in unfamiliar surroundings, a problem further accentuated by the fatigue many suffer, when operating on busy satellite missions (McQueen, 2015, Rossell-Perry et al., 2015, Maine et al., 2012). The
directors were also quick to point out the financial issue with parachute missions as being unsustainable and less cost-effective than permanent facilities (Uetani et al., 2006, Butler): An estimated $250 million dollars are spent by NGO’s annually on short term surgical missions (Wall, 2014, Pezzella, 2006) and, in agreement with the literature, the clinicians in this study commonly felt that this money could be better spent on sustainable services: put into perspective, one NGO spent a staggering $30,000 transporting their surgical services to Ghana, yet $60,000 would have built a new wing on the local hospital (Abdullah, 2008).

The clinical directors agreed that there are many confounding factors which contribute to surgical need not being met, such as low availability of medical facilities and shortage of surgeons anaesthetists and ancillary staff (Chirdan et al., 2010). A common desire for qualified staff to work for higher salaries in wealthy city practices or overseas further compounds the problem (Kupfer et al., 2004, Dodani and LaPorte, 2005). In order to overcome this problem the clinicians emphasised how a good comprehensive care centre can provide an excellent training environment for upcoming surgeons which may encourage them to stay (Shrime et al., 2015, Butler, Butler, 2016). The centres in this study provided long term surgical internships for local and visiting surgeons, which they felt led to high levels of competence in line with global standards of care and encourages a shared learning collaboration with international cleft teams (Butler, 2016). By offering high levels of training they hope to encourage local surgeons to remain in the vicinity thus avoiding the 'brain drain' effect where individuals attain high levels of expertise then move away (Dodani and LaPorte, 2005, Kupfer et al., 2004).

Finally, an important point touched on by a couple of clinicians was the need to offer comprehensive surgical care for all patients with CLP irrespective of age or socio-economic status: In contrast to much of the literature, they felt that in their experience, late repairs could prove extremely beneficial and considered their surgery to be largely successful when employing a comprehensive care strategy. As mentioned, the gold standard for intervention for CLP asserts the need for multidisciplinary services and often multiple procedures yet the majority of the studies discussed in chapter 5 reach the conclusion that late intervention for CLP is
not cost effective based on a single surgical procedure, minimal SLT input, no orthodontics and rarely dental or prosthetic intervention.

As discussed in chapter 5, even under optimal conditions, multiple procedures are often required to manage CLP successfully. The literature reports that less than 50% of single procedures result in normal speech (Kuehn and Moller, 2000, Kummer et al., 2012); fistula formation occurs in up to 27% of cases (Murthy, 2011, Hardwicke et al., 2014, Bykowski et al., 2015); VPI and compensatory cleft speech characteristics often persist into adulthood, irrespective of fistula formation (Peterson-Falzone et al., 2001, Britton et al., 2014); and psychological angst is not uncommon even post surgery (Bradbury and Habel, 2008, Endriga and Kapp-Simon, 1999, Millar et al., 2013). Compounded by the fact that the complication rate is increased for surgery in older patients (Schönmeyr et al., 2016), it is surely unreasonable to expect exceptional results following a single procedure repair for a cohort attending for surgery late in life, many of whom have scarring from previous badly performed procedures and few of whom have been offered adjunctive therapy. In addition, it is unlikely that patients who are happy with their CLP will attend for treatment late in life so, by definition, all the older patients attending these clinics are likely to present with amplified complexity. This raises three points: Is the standard bar being set too high? Are appropriate parameters being measured? should we not be offering more comprehensive care to this cohort before we make a judgement as to success of failure of intervention. These points will be discussed further in the following section.

The clinics in Hyderabad all offer comprehensive care, including secondary surgery, alveolar bone grafting, orthodontic treatment and dental care for all patients who wish to proceed irrespective of age or social status. They have a high compliance rate and relate this to offering realistic projected outcomes. This was borne out in this study where several of the participants commented that, although attending multiple appointments was a difficult commitment, they considered it worthwhile for the long term result.

Although some NGO's, including Smile Train, have funded orthodontic and dental care since their inception, the service has not been widely available. This has largely been due to the lack of appropriate facilities and clinical expertise. As
discussed in section 8.5, a personal correspondence with the vice president for Strategy and Evaluation for Smile Train (October, 2017) revealed that, whilst the organisation appreciates the importance of expanding these interventions and offering comprehensive care to a wider population, there is a requirement to find an appropriate outcome measure to audit outcomes and benchmark services in order to guarantee quality of care. In particular they are looking for a measure which accounts for psychosocial and physiological outcomes and which is less unwieldy to implement than a formal questionnaire but, as yet, have not found one suitable to meet their needs. As will be discussed the TOM-clp shows promise in providing such an instrument.

All the clinicians reiterated the importance of education to improve health literacy in relation to CLP and the programmes they have implemented are clearly outlined in section 9.5. These have been developed to be empathetic and culturally sensitive and are in keeping with those described in the literature (Reddy et al., 2017, Mandrano et al., 2017, Mednick et al., 2013, Nutbeam, 2008, Sørensen et al., 2012). Although providing a scientific explanation for CLP can correct misconceptions, care must be taken not to transfer the causal attributions and make the parents feel guilty about other contributory factors which they might have foreseen and avoided such as dietary deficiencies (Wehby and Murray, 2010, Yazdy et al., 2007) and tobacco use (Honein et al., 2007, Little et al., 2004). The teams in this study were aware of the risk of parents interpreting advice regarding how to manage their child’s cleft as a criticism that they have been neglectful which is similarly outlined by Nelson, O’Leary et al. (2009) in their UK study. Although participants in this study did not express personal shame as the result of the CLP one participant alluded to the fact that his parents felt guilty only once he had started his treatment at the age of 22 because they hadn’t realised treatment was an option.

An interesting observation amongst the cleft care providers was the benefit families obtained from mixing with other families with experience of CLP. This was also conveyed by the participants in this study, many of whom had never come across anyone else with CLP before attending the centre. As discussed in section 9.9.1 the literature clearly describes the social stigma associated with CLP (Strauss
et al., 2007) which can be associated with marginalisation and bullying (Wehby et al., 2006, Blas et al., 2011). Similar confrontation was described by many of the participants in this study, often leading them to become shy and reclusive for fear of ridicule. Meeting others who have fallen victim to the same experience, has been shown to reduce personal feelings of isolation reduce social stigma and improve general well-being (Kerr and McIntosh, 2000, El-Shazly et al., 2010). Clinicians recognised that mingling encouraged families to exchange stories which helped dispel their misconceptions and develop a better understanding of what to expect from treatment. This type of activity can prove particularly useful in addressing the "self blame" parents often attribute to their situation such as being punished by god for misdemeanours in a previous life (Weatherley-White et al., 2005, Mednick et al., 2013) or not having realised they could have helped their child earlier (Nelson et al., 2009).

Several of the participants interviewed for this study described the benefit of meeting other individuals with CLP, as it allowed them to reflect on their own difficulties in relation to the experience of others. They expressed that it made them feel less isolated and reinforced their enthusiasm to continue treatment. The whole process of attending the clinic appeared to give them a more positive attitude towards CLP which they projected as optimism for the future. The importance of bolstering individuals personal resilience, teaching coping strategies and focusing on the positive aspects of their life, has been described in the literature (Nelson et al., 2009, Beaune et al., 2004, Eiserman, 2001). Certainly the participant in this study reported a greater confidence and had a better psychosocial attitude to life following treatment which was supported by the post-operative TOM-clp scores amongst the interviewed cohort. In this vein, it must be considered that finding an empathetic support network and encountering a positive attitude to CLP amongst the staff, may be playing as significant a role in improving well-being and participation as the actual surgery. It could be hypothesised that this support structure would be much stronger in coming to regular visits at a comprehensive treatment centre than a single visit to a surgical mission. Unfortunately it is not possible to separate these components in this research but it would be an interesting concept to follow up. Certainly it supports
the need to change the attitude of the general public towards CLP in addition to directing education towards those requiring treatment (Venkatesan, 2015).

There are often cultural barriers which prevent less educated families in India seeking treatment and many perceive that accessing health care is not appropriate for them (Gulliford et al., 2002). Consequently, although many public health services are free of charge uptake is still very low, as reported by the clinicians in this study as one of the reasons CLP remains untreated. A similar problem has been reported in relation to free immunization programmes (Joe, 2014) and maternal health care instruction (Pallikadavath et al., 2004, Pathak et al., 2010). Despite sustained efforts by cleft care providers it is surprising how many families remain unaware of the availability of treatment opportunities for CLP and although cleft camps are considered the most comprehensive mode of education for CLP, less than a quarter of families in this study attended through this route. For the majority of participants their enlightenment regarding treatment opportunities was largely serendipitous, either through a chance sighting of an advertisement or a fortuitous encounter with a knowledgeable stranger. This is in contrast to Upadhyaya et al., (2017) who report 60% of their cohort had been recruited through cleft camps, however it is not possible to interpret much from these data since there is no record of the number of cleft camps nor the amount of alternative advertising in either study. After starting treatment participants spoke passionately about their dedication to broadcast information about timely cleft care and intimated regret at not having had the opportunity themselves, a number have even been recruited by the cleft centres to this end. These figures indicate that there is a need to reflect on the different modes of delivery for CLP education to enhance the dissemination of information to a wider population.

Whilst cleft camps are an invaluable means of identifying individuals with CLP, a lack of trained personnel will limit their capacity for disseminating information over a wide geographical area. In 2005, in acknowledging the need for community health education, the Indian Government initiated the National Rural Health Mission (NRHM) targeting 18 states which were identified as having weaknesses in public health delivery (Agarwal and Sangar, 2005). Part of this incentive included the appointment of volunteer ASHAs and Auxiliary Nurse Midwives (ANM) to
establish an educational link with each community and provide a first point of contact for individuals who would otherwise have limited access to health advice (Gupta et al., 2010). Within this framework much emphasis is made towards delivery of maternal health care and identifying congenital anomalies such as CLP however the clinicians in this study are sceptical that this is undertaken effectively. These thoughts are supported by the findings of Rao and Mant (2012) and by Marjra and Silan (2016) who found that whilst Indian rural nursing care providers, could identify CLP, little was offered to manage the situation. This would indicated that, whilst ASHA and ANM provide excellent potential for education, greater training and support is obviously required for this route to function efficiently.

**In summary,** the cleft centres involved in this study were fully comprehensive and offered a wide range of multidisciplinary services including secondary surgery, orthognathic surgery, dentistry and orthodontic treatment. The clinical teams felt it was important to meet the global gold standard of care for all their clients irrespective of age or social status. They expressed concern about the wide disparity of services being provided in India and felt that more emphasis should be placed on developing centres of excellence rather than perpetuating parachute missions. For this change to be implemented they suggested that auditing outcomes and benchmarking services would be required in order to provide quality data and change the opinion of fund holders at a local, national and international level.

They also recognised that the restrictions around accessing care for CLP have remained unchanged for many years and hinge on poor levels of understanding and cultural misconceptions about CPL and the potential for intervention. It is agreed that there is a need to provide culturally sensitive education to overcome these obstacles, there should be as great an emphasis on changing public attitudes towards CLP and better collaboration with regional health care providers and educators across the nation.

**10.2. Aim 2: Exploring the psychosocial impact of untreated CLP**

In what way does untreated CLP impact the activity, participation and well-being of the individual and how are these changed by intervention?
10.2.1. Demographics

The diversity in distribution of participants between Assam and Telangana reflect the different landscapes of the two states, as outlined in the 2011 Census (Indian Government, 2011): Assam is largely rural in nature with 88% of the population residing in rural communities; in contrast, Telangana is more urbanized and less than 70% of the population is rurally based. Although the overall populations of the two states are not dissimilar, at just over 31,000,000, Guwahati, the largest city in Assam has a population of only 1 million in comparison with Hyderabad which is seven times larger. Bearing these figures in mind it would be expected that the majority of participants in the Assam centre would have been recruited from a rural population. The fact that 14% of the Hyderabad cohort had urban backgrounds may reflect the opportunistic nature of Indian cities, where in a process known as 'urbanisation', where individuals are drawn to the city to find employment, creating large communities of urban poor with inequitable access to health care (Nambiar et al., 2016). So, whilst it is essential that specialist medical facilities continue to be built in rural communities, these figures would indicate that dissemination of educational programmes are equally important for rural and urban communities alike and has been acknowledged by the NRHM programme which was expanded to include an urban component in 2013 (Gupta et al., 2010).

Interestingly, the distances travelled to the cleft centres are greater in Hyderabad than Assam, for which three credible reasons are proposed: Firstly, travelling the length of the Brahmaputra valley, in Assam, is considerably more challenging than using the relatively comprehensive infrastructure of the Telangana travel network. Secondly, the centre in Guwahati had only opened in 2009, so at the time of data collection knowledge of treatment availability may not have disseminated far afield possibly making recruitment more likely from close to the cleft centre. Thirdly, a number of the participants in Hyderabad intimated that they had experienced disappointing treatment elsewhere, consequently they were choosing to travel extensively to visit these particular centres of excellence. This is less likely to happen in Assam since there is only one centre for treatment of CLP and the region is less served by district hospitals where inappropriate surgery could occur.
The gender ratio was equal for this cohort which is in contrast to the epidemiology report from smile train (Kalra, 2014) from which it is suggested that the higher male to female ratio in India reflects not only the natural preponderance for CLP in males but also a cultural preference towards seeking treatment for male children. However the data set in this study is too small to make judgement as to whether the figures represent a general reduction in gender bias.

The diversity in age distribution of the participants in Assam and Hyderabad may well reflect the difference in treatment options offered between the different centres. As mentioned, the centre in Guwahati is still in its relative infancy and, whilst treatment is not age restricted, they currently concentrate on providing primary surgery rather than secondary interventions. In addition, other disciplinary services are restricted in Assam with no availability of orthodontic treatment and only limited speech and language therapy and dental care. which may account for the preponderance towards younger patients in Assam. In contrast, the centres in Hyderabad offer full complement of specialties including secondary cleft surgery, orthognathic surgery and comprehensive orthodontic and dental care, and is more likely to attract greater numbers of patients in their late teens and early twenties, who have become aware of how their speech limitations and different appearance may negatively impact their prospects in relation to marriage and employment opportunities.

10.2.2. Interviews and framework analysis

Chapter 2 described the sheer scale of the problem of CLP in India and alluded to how the disparity in health care provision across different regions was leaving many individuals unable to access appropriate surgical intervention. This is supported by the fact that all of the 85 individuals participating in this study presented with a certain level of deficiency in their CLP treatment and the interview process revealed that, without exception, this was a group of individuals whose lives had been restricted in some way as a result.

As reported in an earlier section, the main reasons given for not receiving intervention mirrored those in the literature as lack of knowledge and superstition. However, an additional problem highlighted in this study, but not
alluded to in other studies, was the amount of misinformation provided to patients: 95% of the participants in this study had CLP and all of them, bar one, had received intervention for their cleft lip before the age of six, indicating they had all had access to medical advice. Even the 33 year old with untreated CLP had been offered surgery, although had declined, which indicates that all the participants had seen someone who should have identified their cleft palate and explained that a procedure was advisable. However, 12 families did not remember being told that the palate needed treating, two families misheard the timing for the second procedure and waited ten years rather than ten months before returning, one family was told to wait, but not for how long, 3 families were told that their child was underweight or too unwell for surgery and took it to mean they could never have it fixed. An additional 35% of patients had received surgery which had failed but were not aware that they had been told of the potential for further treatment.

As discussed, there are many problems associated with the delivery of education about CLP and its success will depend on who is delivering the information, when it is delivered, the level of detail provided, who is receiving the information and the manner in which it is delivered (Naram et al. 2017, Naram et al., 2013, El-Shazly et al., 2010). This is also true of information sharing and it has been shown that unless information is delivered empathetically and appropriately understanding and compliance will be poor (Davis et al., 1998, Williams et al., 2002, Williams et al., 1995). Information transfer has clearly not been successful for these individuals and caused a barrier to treatment. This indicates the need for more optimal modes of communication within a clinic situation, particularly with patients of low literacy, to avoid these potential misinterpretations in the future.

10.3. Family impact

The psychological impact of having a child with CLP is widely reported in the literature (Turner et al., 1997, Berger and Dalton, 2009, Hunt et al., 2005) and similar findings were made evident during this research. The issue of marriage prospects quickly emerged during the interviews and was a cause for pre-occupation, with outward appearance being highlighted as an influential factor.
There was a similar conviction that intervention would improve marriage prospects as seen by Weatherley-White, Eiserman et al. (2005) however, in contrast to their study, the level of concern was equally reported by the parents of boys and girls and interestingly the concerns tended to be raised by parents rather than their child. Although it is reported that modern Indian children exert more influence in choosing a marriage partner than in the past (Kamble et al., 2014, Ghimire et al., 2006), this is less pronounced in lower socio-economic classes and rural communities (Banerjee et al., 2013), so these findings may have reflected a parental feeling of responsibility for finding a prosperous match.

Parents of younger children tended to voice concerns about the influence of CLP on their child's ability to socialise and make friends. As mentioned earlier, parents reported feelings of guilt on this front but there was also a sense that they were aggrieved and disappointed about being given poor treatment and misinformation elsewhere.

The financial impacts to the family were complex. As mentioned in the previous section, in agreement with the literature, participants reported the struggle to take time off work and associated costs with attending appointments. However, an additional financial burden which manifested itself in this study was the restrictions that CLP placed on earning money to support the family: Two families reported that their child could not work in the family shop before treatment because the customers could not understand them, and several individuals were traumatised by the fact they could not find work and had to rely on their family for support. One participant commented that it made him feel like a ‘burden’ which exposes the importance of employment not only in fiscal terms to the family but also in relation to autonomy and self-worth. These factors highlight the importance of accounting for potential social impacts of CLP repair such as personal and financial autonomy which are currently under reported in the literature.

A problem which was touched on was the financial impact of some of the services. Owing to lack of parity in external funding for cleft services, some participants had to pay a nominal fee for their orthodontic treatment at one of the centres. Although they were only charged for the orthodontic brackets this is an expense which is not incurred elsewhere and highlights the need to achieve equity in funding provision.
10.3.1. Activity

Speech was by far the most talked about impact of CLP and affected all bar four of the participants to some degree. In general participants perceived that their speech improved following intervention; post-operatively all of participants could at least converse with friends and family, half could be understood by most people and 35% reported that they had no problem communicating with anyone. This was a stark difference from pre-operative reports where 35% of the participants could not be understood by anyone outside their family and 15% could not really make themselves understood at all, even by those most habituated to their speech. These findings mirrored the perception of the participants in the study by Schönmeyr, Wendby et al. (2015b) despite their conclusion that intervention was of limited value based on speech outcome. This identifies the importance of embracing multiple parameters when assessing outcome, including patient perspectives. The accuracy of the patients perceptions was supported by their observations during the interviews where 80% of the comments about speech had negative connotations pre-operatively and the trend was reversed to 80% positive comments post-operatively.

A similar trend was seen for participants perception of regurgitation, with only 28% reporting a little regurgitation post-operatively. Pre-operatively more than three quarters of the cohort had suffered regurgitation to some degree and for a third of the participants this was every time they ate or drank. Although regurgitation was mentioned less often than speech, 85% of the pre-operative comments had negative connotations in comparison with only 12% post-operatively which supports the reports of the participants perceptions as accurate. The results of this study were the reverse of those reported by Schönmeyr, Wendby et al. (2015a) who only reported 4-6% of patients with nasal regurgitation pre-operatively and 26-28% post-operatively. Schönmeyr, Wendby et al. suggest that the high levels of post-operative regurgitation relates to altered musculature however they do also refer to 11% of their recall patients having developed fistulae which is likely to have contributed. Since this study comprises patients who have had secondary repair and fistula closure the results are not directly comparable, however they potentially indicate the importance of
secondary surgery in the elimination of regurgitation in an older cohort which could be the focus of further research.

Two of the participants in this study reported increased nasality and regurgitation after starting orthodontic treatment, but the expectation was that it would improve with reconstructive surgery. Both had previously had their palate repaired using a tongue flap which had broken down to develop a fistula which is a commonly reported cause of nasality (Henningsson and Isberg, 1987, Henningsson et al., 2008, Sell et al., 2001, Harding and Grunwell, 1996) and regurgitation (Jeffery et al., 2000). The problem had possibly worsened with the initiation of orthodontic treatment potentially owing to the arch expansion accentuating the fistula.

Oral health and function manifested itself as a specific problem for 25% of the participants, yet, as far as can be established by the PI, it is not mentioned in the literature in relation to late intervention for cleft palate. Comments related to an inability to eat properly, particularly where the dental arches had collapsed as well as the more obvious aesthetic component and impact on speech. Poor dental health and malocclusion are acknowledged as important considerations in CLP (Antoun et al., 2015, Gherunpong et al., 2004, Williams et al., 2001, Masood et al., 2013), yet the correlation has not been made in relation to the older patient who remains untreated, indicating the need for future studies.

10.3.2. Participation

The impact of CLP on participation was profound and almost all the participants reported that they felt their involvement and development was being restricted as a result of negative influence of people around them. These worries focused primarily around speech and their ability, or inability, to be understood causing many to stay silent before surgery to avoid embarrassment and frustration. Many expressed an underlying sense of frustration when they had been excluded and ambitions had not been attainable. This was particularly demonstrable in relation to the rejection many felt when trying to make school friends which made them
reluctant to participate in education for risk of ridicule. The impression of loneliness and isolation some of the participants described pre-operatively was palpable and even where positivity was expressed there was a sense that the CLP had lowered their self esteem. For others it was possible to identify a sense of resignation to partake in normal activity despite their reservations, it was noticeable that they still expressed feelings of anxiety even where they had a supportive network of family and friends. In contrast, where friends and family were patient and supportive, participants were actively encouraged to participate in everyday activities and there was clearly a sense that these individuals had followed a well integrated childhood and had been able to realise their potential in education unimpeded.

There was a stark contrast post-operatively where educational success or finding employment accounted for an overwhelming expression of accomplishment when they were finally embraced into something they enjoyed. Where pre-operatively they had felt that socialising was setting themselves up for failure, post-operatively they were more confident in meeting people as they had a conviction that they would be better understood, irrespective of any imperfection in their speech. There were also more specific examples of how a level of improvement in speech had impacted their everyday lives and being able to speak on the telephone was important to many. Although they had been able to text their friends in the past, the ability to speak on the phone made them feel less of an outsider when making plans; Being able to go shopping or travel by bus knowing that they could convey their meaning without needing assistance or fearing mockery was considered a liberating move towards independence.

These everyday impacts of CLP and the potential improvement post surgery are largely overlooked in previous studies on late intervention, yet clearly played an important role in improving life quality for this cohort. Once again this indicates the importance of taking a more holistic view of measuring outcome as none of these factors would have become illuminated in a study monitoring changes in speech and intelligibility performance alone.
10.3.3. well-being

Facial difference and poor dentofacial aesthetics are known to elicit an unfavourable response in many societies (Cunningham, 1999, Macgregor, 1970, Shaw, 1981) which in turn leads to peer victimisation and bullying (Berger and Dalton, 2009, Hunt et al., 2006, Seehra et al., 2011). This has been reported at all ages with implications such as; anxiety and depression (Van der Wal et al., 2003, Whitney and Smith, 1993, Fekkes et al., 2004, Due et al., 2005, Bond et al., 2001); reduced academic prowess (Glew et al., 2005) and less chance of making friends (Helm et al., 1985).

Social bullying is widely reported with CLP (Hunt et al., 2006) and was demonstrated by Reddy et al., (2017) using the ICF framework. They demonstrated that, even where adults with CLP treated optimally, they reported being restricted in maintaining interpersonal relationships, employment and major life areas and felt like an observer rather than a participant in conversations. These traits and concerns were mirrored by many of the participants in this study and were associated with the largest proportion of negative emotions. Participants reported feeling upset, shy, fearful, humiliated, embarrassed and even aggressive in response to unkindness for those around them. Equally dominant in the course of the interviews was their experience of mockery which shattered their confidence and crushed their self esteem and caused them to shy away from society rather than risk suffering the demoralising and derisive behaviour of their peers. Such behaviour is commonly cited in the literature and can lead to a vicious cycle of negative emotion; victims of bullying become insecure, they lack assertiveness which is seen as a sign of weakness and leads to more bullying (Schwartz et al., 1993, Bernstein and Watson, 1997). Even if the source of the bullying is removed, the personality traits often persist; this was seen in a number of individuals in this study who, despite improvement in communication and reduction in regurgitation, remained shy and unwilling to socialise. It was anxiety from this type of scathing treatment which appeared to be the most salient cause of individuals' feeling marginalised and failing to reach their true potential, particularly in relation to education and employment; Participants mentioned how difficult it was to participate to full capacity during their early years in education, how teasing from
peers and difficulty in communicating with the teaching staff gave them an inferiority complex and put them at a disadvantage in fulfilling their potential. This mirrors the findings of other researchers who report individuals with poor speech and intelligibility having difficulty in forming friendships, being more likely to be rejected by their peers (Boyd et al., 2006), feeling inferior (Broder et al., 1994) and perceived by others as less intelligent (Allard and Williams, 2008). These findings indicate the importance of acknowledging the psychological impact of CLP and might account for the benefit participants gleaned from mixing with others who could relate to their experiences.

Robinson Ramsey et al., (1996) describes a similar negative interaction cycle, whereby individuals with facial disfigurement avoid interaction, in anticipation of a painful encounter, based on previous experience (Robinson et al., 1996). If this cycle is broken, a more positive attitude to socialising may give the impression of improved communication, whether or not the speech is actually better. This might well have been true for some of the participants in this study who report a more positive attitude towards socialising post-surgery. This appeared particularly common for participants whose secondary surgery had given them enhanced confidence in their appearance and communication skills. This process might be partly responsible for patients perceived benefits from intervention reported in the literature, despite speech analysis indicating otherwise.

Personal dissatisfaction with facial appearance is also known to have a negative impact on self esteem (Shaw et al., 1980). This was the case for many of the participants in this study and there was a commonly held belief that surgery would improve this situation. Post surgery, there was a marked improvement in self opinion with 80% of the remarks having positive connotations in comparison to just 20% pre-operatively. This level of expectation helps to explain the importance they imposed not only on the functional but also the aesthetic aspects of surgery.

**In summary**, the participants’ comments in the interviews indicate how activity, participation and well-being are inextricably linked in a complex web of cause and effect: Difficulty in communication and regurgitation can cause embarrassment and anxiety about socialising. Socialising can lead to bullying which in turn affects
self esteem, causes shyness and a reluctance to socialise. Marginalisation and bullying can interfere with social progress and prevent people from reaching their true potential in society. However, the outcomes are not predictable; given the right social support individuals thrive despite problems in communication, this gives them confidence and enthusiasm to socialise which in turn allows them autonomy and a prominent social role in society. Although there was an overwhelming conclusion, from all the participants, that the corrective surgery they had received had been beneficial in a number of different dimensions, their comments also echo the findings in the previous section which suggest that psychological support and education are equally important as surgery in achieving an optimal outcome.

**10.4. Aim 3: Examining the value and viability of the TOM-clp**

To explore the impact of untreated CLP and evaluate how well the TOM-clp can be implemented as a culturally sensitive tool to demonstrate change in relation to intervention.

**10.4.1. Teaching and implementing the TOM-clp in clinical practice**

The pilot study suggested that the TOM-clp could be taught reliably after a short training session even where the learners were using English as a second language. This hypothesis was borne out in the clinical situation where training was achievable with equal reliability using face-to-face and remote teaching methods. This indicates that the TOM-clp could be disseminated as a standardised outcome measure without the need for expensive and time consuming field visits. Only one assessor was trained by another trainee, but they were able to achieve reliability after just two sessions, even though the trainer had little experience of using the TOM-clp themselves and was not conversant in the local language. Although further research would be required to confirm reliability of disseminated training, these results indicate the ease of training across language and cultural barriers.

The clinical directors confirmed that demand for cleft surgery is high which meant that their clinics could be extremely busy. This emphasised the need for an outcome measure which was quick and simple to integrate into normal clinical
routines. Anecdotally users reported that the TOM-clp was easy to understand and simple to deploy and they felt it could easily be integrated into a clinical routine. Collecting demographic data extended the process slightly but it was suggested that this element could be undertaken by a member of the administrative staff using the patient ID to link the demographic data with the TOM-clp scores so the clinician would only need to spend a few minutes per patient documenting the scores in the patient record and/or a central database. Overall the consensus was that the TOM-clp would prove a useful measure for monitoring change associated with intervention and as a benchmarking tool to compare different services.

The fact that the TOM-clp was based on normal conversation meant that it could be applied equally well for all participants without alteration or compromise: Since there was no specific wording or complex terminology, it was understood by all participants irrespective of age or level of education and literacy. This was supported by clinicians and clp participants who remarked that they felt relaxed because the process did not feel like an interview. Since the CLP participants did not feel that they were 'answering questions' this may have reduced the response bias which is known to be a particular problem with direct questioning in self-reported questionnaires and structured interviews (Furnham, 1986, Bowling, 2005, Broder et al., 2007).

Similarly, since there are no set questions in the TOM-clp, it proved equally simple to administer with participants of all ages, genders and cultures without alteration or adaptation. This is in contrast to the WHOQoL-BREF, where some of the questions needed to be missed out or altered to make it appropriate for particular participants, despite having specifically selected an adapted version previously used in research with similar cohorts (Chen et al., 2006, Saxena et al., 1998, Yao et al., 2002). Implementation of the WHO-QoL-BREF was also limited by language as it was not available in appropriate translations and it demanded a degree of literacy above that of the majority of participants in this study. The requirement of a translator, to overcome these limitations, rendered the WHO-QoL-BREF prohibitively costly and time consuming to complete. These shortcomings highlighted a number of general issues using self-reported questionnaires in mixed
socio-economic communities which may also prove problematic for the Cleft-Q questionnaire but which has been overcome in the design of the TOM.

Another two issues which may have become problematic had the use of the WHOQoL-BREF continued, and potentially with the Cleft-Q, were the cost of printing the three page questionnaire for each participant (Shirima et al., 2007) and also the time taken to upload the information into a data base (Tomlinson et al., 2009), neither of which restrict the TOM-clp. Clinicians alluded to both these points when describing the benefit of using the TOM-clp in that it is not paper based. Although the Cleft Q is also being piloted as an on-line PROM to minimise these problems, as mentioned in section 3.12, this brings with it the requirement for literacy and availability of appropriate electronic devices. The data collection app for the TOM-clp has been designed to work on-line and off-line to reduce the problem of intermittent IT links and since it is completed by the clinician not the patient there is not an issue of computer literacy. In addition, even had the TOM-clp data been notated on paper rather than downloaded, the nature of the data comprises four digits which would not be time consuming to upload into a data base at a later date thus reducing the time and accuracy limitation of extended double entry in paper based questionnaires.

10.4.2. Interpreting impacts of CLP using the TOM-clp

10.5. Analysis of the TOM-clp scores

When assessing the pre-operative condition, there did not appear to be any correlation between size of cleft or type of cleft with TOM-clp rating in the activity domain. This is contrary to the literature which reports that the presence of cleft speech characteristics are closely linked to cleft type and size both in the treated (Hardin-Jones and Jones, 2005, Persson et al., 2002, Mahoney et al., 2013) and untreated cleft (Sell, 1991). Since communication played a large role in the assessment of activity in this study, it might have been supposed that the two domains would correlate more closely. However, unlike the reported literature, which drew their conclusions based primarily on assessment of speech production
and recorded intelligibility, the TOM-clp takes a broader outlook on activity and includes regurgitation, overall communication and the ability to be understood. Communication not only comprises spoken language, it also includes other non-verbal cues which are essential for interacting with others (Wharton, 2009, Payrató, 2009) thus communication is influenced by body language, self confidence and capacity to handle social situations (Dumbravă and Koronka, 2009, Goman, 2009). Putting this into context; If an individuals with CLP is shy and uncomfortable in social circumstances, they are likely to mumble and keep their face hidden which will make them less clearly understood than someone projecting their voice with confidence, whilst holding their head high. Also, a number of individuals alluded to the fact that their speech was better when they spoke quietly which might make them more difficult to understand in a louder social situation than in a quiet home environment. This complex interaction between speech, language and communication means that an individual's perception and their confidence in their environment may have had just as strong an influence on the participants’ communication skills as their ability to produce words. This is likely to have influence on the TOM-clp activity domain in that it reflects generalised communication and may have accounted for the apparent positive impact of intervention in this study when compared with those evaluating speech and intelligibility alone.

The correlation between activity, participation and well-being would tend to indicate that, overall, patients perceive that their communication and regurgitation improve in a manner which parallels a change in participation and well-being. However, the individual vignettes for CH04 and GSR06 indicate that these three domains are not necessarily directly linked, and a low activity score does not always correspond to poor participation and well-being. This is important on two counts: Firstly; it indicates the uniqueness of an individual response to CLP insinuating that every case should be assessed on its own merit. Secondly; research that focuses on speech characteristics and regurgitation as outcome measures will not reliably reflect psychosocial change, so the choice of outcome parameter is critical in making a judgement as to whether an intervention is effective.
Although only five cases were available with mid range TOM-clp scores, the graphic (Figure 23) on page 204, demonstrates a progressive improvement during treatment indicating that the TOM-clp is sensitive as an outcome measure at each stage. This is also demonstrated by the vignette for TB18 which demonstrates the negative impact of the fistula which developed after the onset of orthodontic treatment. It is acknowledged that this is presented only as a single case, however its relevance to this study lies in the potential use of the TOM-clp as an outcome measure for different aspects of cleft care such as orthodontic intervention which requires further investigation.

**In summary**, the TOM-clp appeared to be well accepted as it was considered easy to implement within a busy clinical setting. All the participants felt that surgery had produced positive results, which was reflected in all four domains. A Comparison of pre-operative, post-operative and mid scores indicated that, as an outcome measure, the TOM-clp was sensitive to positive and negative changes as treatment progresses. Close analysis of these changes demonstrated the personal nature of CLP and how different the impacts were for each individual. What was made clear by this variation was, that the size of the cleft, the ability to communicate and the degree of regurgitation did not correlate closely with regard well-being and social functioning. This would indicate that speech and intelligibility, which are commonly used as predictors of outcome, may not reliably reveal all the benefits individuals experience from intervention. This demonstrates the potential gain in widening the parameters of research in this field to include psychosocial measures so as make a more holistic judgement of the potential benefit of late intervention for this cohort.

**10.6. Strengths and Weaknesses**

Several limitations must be considered in the interpretation of this study in relation to their potential impact on the findings. Firstly, the choice of locations and the use of purposeful sampling restricted the participants to individuals who had access to comprehensive multidisciplinary care by experienced clinicians. Further investigation is required to explore how this cohort compares with individuals managed via alternative care pathways such as; optimally timed
intervention, surgery undertaken in transient cleft camps, use of a single intervention approach or an age matched comparison in individuals without CLP. Although 85 participants were recruited for this study, there was a wide variation in demographics, cleft characteristics and management regime, so further investigation on a larger scale is required to explore whether patient age, cleft type, severity of cleft or choice of intervention have an impact on the outcome and explore whether these results translate across different cultures and different countries.

Attempts have been made to increase the validity of this study by employing triangulation, multiple assessors, blinding and randomisation when dealing with the analysis of the TOM-clp data and maintaining transparency by using framework analysis. However, there are potential biases which must be acknowledged:

*Recall bias and patient reported response shift:* Circumstances stipulated that the pre-operative data collected in this study was retrospective and it is possible that participants recall of events may not have been wholly accurate. People's perception of events often alters over time and self-reported data may be subject to exaggeration and embellishment, however this effect could be minimised by repeating the process as a longitudinal prospective study.

*Responders bias:* Attempts were made to minimise responder bias by ensuring a relaxed environment and involving clinicians with whom the participants were already comfortable. However, the presence of an international PI and the fact that the interview process was perceived as relating to the clinic providing treatment may have influenced the participants responses. This impact could be reduced by using independent, non clinical researchers to collect data in the participants home environment.

*Multiple languages and the use of translators:* The TOM-clp was undertaken using simple conversation in the patient's own language which meant it was equally applicable for all ages, cultures and levels of literacy and did not need translation. However, the informal interview process was undertaken via an interpreter which meant the meaning and nuances of the interviewee may have been lost and there
may have been miscommunications or misunderstandings in the dialogue. Attempts were made to minimise this effect through re-iteration and clarification of meaning by the PI during the interview process. Although consideration was given to the use of a professional interpreter and transcribing the participants responses verbatim to minimise this problem, this would have proven prohibitively time consuming and expensive for the purposes of this study.

Training in the TOM-clp was generally straightforward and reliable after only a short training session however, all the clinicians in this study were either native English speakers or frequently use English as a second language. If the study is to be repeated on a more global scale, further investigation is required to confirm that the training of TOM-clp is not only compatible with other languages but also that it can be taught reliably to and by non-English speakers.

From the outset, this study was designed to collect information longitudinally, from different locations and clinical settings and compare the results across cultures. However, factors beyond the control of the PI prevented the study reaching its planned potential: It was felt that the political conflict which impeded data collection in Assam may have related to cultural issues that the PI was unaware of from the onset. Additionally, some difficulty was encountered in maintaining motivation for data collection by some of the clinicians involved. It is possible that if the research was backed by a larger organisation, rather than being run by an individual researcher, the cleft teams might see more personal benefit in data collection and be more enthused in the process.

Finally, at the initiation of this research process there was not another outcome measure available with which to compare the validity of this research. An attempt was made to implement the WHOQoL-BREF to this end but the questionnaire did not prove fit for purpose. Since this time the Cleft-Q has been developed as a PROM directed towards assessing quality of life in a similar cohort to this study. It will be interesting to compare the results when the instrument becomes available for general use.
10.7. Conclusion

It is clear from the discussions with clinical directors that great inroads are being made to improve parity and quality of care for CLP at a local, national and international level. The realisation that lack of understanding about CLP creates a barrier to treatment has lead to the development of various educational strategies. However, even as these developments take place, there remain a significant number of policy makers and even responsible medical professionals, who remain sceptical about clefts; Kalra (2014) quotes statements from commissioners such as "You must be out of your mind if you think this is the most serious medical problem facing our country" and "No child ever died of a cleft" indicating that there is a great deal still to be done in educating the entire population from grass-roots to government.

With regards late intervention for CLP, there is the additional hurdle of the suggestion in a large proportion of the literature that intervention is not beneficial and ill-advised. A few studies in this field have reported anecdotally that patients perceive an improvement in quality of life following late intervention for CLP then have dismissed it as irrelevant as it is not supported by outcomes of speech and intelligibility. Yet there is growing evidence that the psychosocial benefits of late intervention are significant and allow patients to fulfil their potential and independence. These findings are supported by the results of this study, with the additional suggestion that, by providing comprehensive intervention, improvement can be seen in multiple domains. A greater depth of evidence will be required to convince the wider population that a comprehensive strategy is cost effective for late intervention and the TOM-clp shows potential for collecting such data.

The clinical directors interviewed in this study emphasised the need for cleft services to be developed in line with international gold standards to include wider provision of secondary surgery and allied services. The importance of training and centralising services should not be underestimated with a move away from parachute missions and treatments being based on clinical need rather than 'funding by numbers'. A key element in ensuring quality is tighter regulation, auditing and monitoring which requires an holistic outcome measure which is
quick and simple to integrate into normal daily routine. The results of this study show the potential of the TOM-clp for fulfilling this role as it can be adapted for monitoring physiological and psychosocial change to assess the progress of individuals through treatment or benchmarking services on a wider scale. The results of this study also indicate that the TOM-clp can be taught equally reliably through face-to-face and internet training. This indicates that it could be disseminated widely as a standardised outcome measure without the need for expensive and time consuming field visits.

It is hoped that this research can pave the way to improve quality of care for patients with untreated CLP not only at an individual level but also informing policy and changing attitude of the health culture surrounding CLP. In addition, the results indicated that the TOM-clp might be adaptable to detect psychosocial change relating to orthodontic treatment and oral health and a follow on study is planned to investigate this element further in the future.


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**CHAPTER 12: APPENDICES**

**Appendix 1: TOM-clp**

<table>
<thead>
<tr>
<th>IMPAIRMENT</th>
<th>Description</th>
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<tbody>
<tr>
<td>0</td>
<td>Profound. Bilateral cleft lip and/or complete cleft palate.</td>
</tr>
<tr>
<td>1</td>
<td>Severe. Complete unilateral or bilateral cleft lip and/or cleft palate including more than 1/2 half of the hard palate.</td>
</tr>
<tr>
<td>2</td>
<td>Severe/Moderate. Extensive cleft lip and/or cleft palate including more than 1/2 of hard palate.</td>
</tr>
<tr>
<td>3</td>
<td>Moderate. Moderate lip clefting and/or cleft palate including less than 1/2 of hard palate.</td>
</tr>
<tr>
<td>4</td>
<td>Mild. Minimal lip clefting and/or cleft palate only of the soft palate.</td>
</tr>
<tr>
<td>5</td>
<td>Normal lip and palate.</td>
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<tr>
<th>ACTIVITY</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>0</td>
<td>Profound difficulty with speech. Unable to make recognisable words or sounds. Not understood by familiar or non-familiar listeners. Profound difficulty eating and drinking.</td>
</tr>
<tr>
<td>1</td>
<td>Severe problems with articulation. Limited to a few recognisable words. Understood by family and friends with only with help of hand signals but not by strangers. Severe problems eating or drinking, much food and all liquids coming down the nose when eating.</td>
</tr>
<tr>
<td>2</td>
<td>Severe/moderate problems with articulation. Can produce single words but difficulty using whole sentences and sounds very nasal. Can usually be understood by friends and family in calm quiet surroundings but strangers only able to understand occasional words. Severe/moderate difficulty eating and drinking, some food and most liquids coming down the nose when eating.</td>
</tr>
<tr>
<td>3</td>
<td>Moderate difficulty with articulation and speech. Can be understood by family and friend in most circumstances but understood by non-familiar listeners only when speaking in context. Generally difficult to understand when excited or speaking quickly. Moderate difficulty eating and drinking; no problem with solid food but some liquids coming down the nose.</td>
</tr>
<tr>
<td>4</td>
<td>Mild difficulty with articulation and being understood but sound a little bit nasal. Understood almost all the time only occasionally unintelligible when very excited, speaking fast or loudly. Mild problems with nasal regurgitation when drinking.</td>
</tr>
<tr>
<td>5</td>
<td>No difficulty in being understood. Normal speech and feeding for someone of their age.</td>
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<tr>
<th>PARTICIPATION</th>
<th>Description</th>
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<tbody>
<tr>
<td>0</td>
<td>Profound difficulty in participating. Unable to participate in any social/educational/family role. No social integration.</td>
</tr>
<tr>
<td>1</td>
<td>Severe difficulty in participating. Very low opinion of themselves. Very little confidence. Doesn't mix socially. Never eats or speaks in public. Not taking advantage of any opportunities around them. Will not participate in daily activities like shopping or using the phone.</td>
</tr>
<tr>
<td>2</td>
<td>Severe difficulty in participating. Some self confidence will mix socially in familiar situations. Avoids mixing and eating with strangers. Needs a lot of support in order to join in. Will accompany friends family but will not participate in daily activities such as shopping.</td>
</tr>
<tr>
<td>3</td>
<td>Moderate difficulty in participating. Reasonably confident. Will mix socially in certain environments. Participates in educational activities but needs a lot of help and encouragement. Will occasionally make the most of opportunities but seeks assistance with daily activities such as shopping &amp; using the phone.</td>
</tr>
<tr>
<td>4</td>
<td>Mild difficulty in participating. General self confidence and willing to mix socially in most environments. Participates in family/social/educational decision making. Will make the most of opportunities in certain situations but occasionally still seek assistance with daily activities such as shopping and using the phone.</td>
</tr>
<tr>
<td>5</td>
<td>No difficulty in participating. Fully confident and participates in normal activities appropriate to their age. Well integrated into family/educational/social environment. Will undertake daily activities such as shopping and using the phone without assistance.</td>
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<tr>
<th>WELL-BEING/DISTRESS</th>
<th>Description</th>
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<tbody>
<tr>
<td>0</td>
<td>Profound distress. Constantly severely distressed and embarrassed. Always severely withdrawn, frustrated, apathetic and depressed. Unable to express or control emotions appropriately.</td>
</tr>
<tr>
<td>1</td>
<td>Very severe distress. Very frequent episodes of severe distress and embarrassment. Often extremely withdrawn, frustrated, apathetic and depressed in most situations. Needs constant reassurance, encouragement and support. Poor control of emotions.</td>
</tr>
<tr>
<td>2</td>
<td>Severe distress. Frequent episodes of distress and embarrassment. Often withdrawn, frustrated, apathetic and depressed in unfamiliar situations. Needs frequent reassurance in unfamiliar situations. Frequent emotional outbursts but generally able to control emotions.</td>
</tr>
<tr>
<td>4</td>
<td>Mild distress. Generally well adjusted with only a few episodes of distress/embarrassment/withdrawal/frustration/apathy depression. Needs occasional reassurance in unfamiliar environments.</td>
</tr>
<tr>
<td>5</td>
<td>No distress. Able to cope in most environments and accepts limitations.</td>
</tr>
</tbody>
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Appendix 2: Ethics approval for pilot project for TOM-clp

Joanna Rees
Registration number: 120252576
Human Communication Sciences
Programme: PhD pilot study department HCS

Dear Joanna

PROJECT TITLE: Piloting the training manual relating to a research project relating to Intervention For Cleft Lip And Palate In Adulthood
APPLICATION: Reference Number 002397

On behalf of the University ethics reviewers who reviewed your project, I am pleased to inform you that on 30/01/2015 the above-named project was approved on ethics grounds, on the basis that you will adhere to the following documentation that you submitted for ethics review:

- University research ethics application form 002397 (dated 10/12/2014).
- Participant information sheet 003927 version 1 (08/12/2014).
- Participant information sheet 003928 version 1 (08/12/2014).
- Participant consent form 003929 version 1 (08/12/2014).

The following optional amendments were suggested:

Establish an alternative to the use of home address as contact, as suggested by reviewer.

If during the course of the project you need to deviate significantly from the above-approved documentation please inform me since written approval will be required.

Yours sincerely

Rachel Ralph
Ethics Administrator
Human Communication Sciences
Appendix 3: Ethics approval for data collection

The University Of Sheffield.

Downloaded: 15/04/2017
Approved: 11/02/2016

Joanna Rees
Registration number: 120252576

Human Communication Sciences
Programme: post graduate research

Dear Joanna

PROJECT TITLE: Measuring the psycho-social impact of late intervention for cleft lip/palate across different countries: Adaptation and application of the Therapy Outcome Measure (TOM)

APPLICATION: Reference Number 006653

On behalf of the University ethics reviewers who reviewed your project, I am pleased to inform you that on 11/02/2016 the above-named project was approved on ethics grounds, on the basis that you will adhere to the following documentation that you submitted for ethics review:

- University research ethics application form 006653 (dated 09/02/2016).
- Participant information sheet 1014778 version 4 (09/02/2016).
- Participant information sheet 1014777 version 4 (09/02/2016).
- Participant consent form 1014780 version 1 (15/01/2016).
- Participant consent form 1014779 version 1 (15/01/2016).

If during the course of the project you need to deviate significantly from the above-approved documentation please inform me since written approval will be required.

Yours sincerely

John Mason
Ethics Administrator
Human Communication Sciences
Appendix 4: Consent form A: Adults

Participant ID number:

TITLE: Using An Adapted Version Of The Therapy Outcome Measure To Collect Data On The Impact Of Intervention For Adults Undergoing Treatment For Palate

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<td>I have had the opportunity to ask questions and understand that my participation is voluntary and that I am free to withdraw at any time, without giving reason</td>
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<td>I am happy for sessions to be recorded and/or filmed and agree for the resulting material to be used for the purpose of this study and publication purposes where it is appropriately anonymised.</td>
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Name of Participant | Date | Signature

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Name of Researcher | Date | Signature

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Appendix 5: Consent form B: minors

Participant ID number:

TITLE: Using An Adapted Version Of The Therapy Outcome Measure To Collect Data On The Impact Of Intervention For Adults Undergoing Treatment For Palate

Name, position of Researcher: Joanna Rees. PhD Student. University of Sheffield, Department of Human Communication Sciences

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Name and relationship of guardian | Date | Signature |
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Name of Participant | Date | Signature |
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Name of Researcher | Date | Signature |
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Appendix 6: Participant information sheet for cleft data collection

TITLE: Using An Adapted Version Of The Therapy Outcome Measure To Collect Data On The Impact Of Intervention For Adults Undergoing Treatment For Cleft Palate


Invitation to participate

You are being invited to take part in the above research study. Before you decide whether to participate it is essential that you fully understand why the research is being done and what will be involved. Please take time to read/listen to this explanation leaflet before you make your decision.

What is the purpose of this study?

I am collecting information on peoples experience of living with a cleft lip or palate and to see in what way things are changed as the result of treatment. This information will help surgeons and the supporting team to understand your needs, and the needs of similar people, so they can help provide treatment in a way which will suit people better in the future. To do this I want to talk with people with a cleft palate as well as their family and friends in order to find out what challenges you have faced and may be facing now. This will allow me to identify factors associated with having a cleft palate and incorporate them into the questionnaire.

Who are the researchers and how is the research funded?

I am a student in the Department of Human Communication Sciences at the University of Sheffield in the UK. I am also a dentist and surgeon involved in the provision of care for cleft lip and palate patients. I am undertaking this study because I believe that the continuing progress of cleft care in the developing world is something which is of great importance.

Why am I asking you to be involved?

I am looking for people who are seeking or are receiving treatment for a cleft lip or palate in later life because they missed the opportunity to have treatment in infancy. Agreeing to participate in this research will not influence the cleft care you personally receive through the centre but may help others in the future.

What will you have to do?

I would like you and your friends and family to take part in an informal chat to discuss your own experiences of your cleft lip and palate and how it affects your daily life. Each session will last about 20
minutes and I will keep a record of your answers on a special form so that I can assess how things are changing as your treatment progresses. Our name will not be used on the form so no-one will be able to identify specifically what you have said from these notes.

I would like to meet up with you on at least three occasions during your treatment to chat about how you are getting on and if your life has changed in any way as time goes on. During this time I will also be teaching other people how to fill in the assessment form so the chat sessions will sometimes be undertaken by trainees.

**What will happen to the information you provide?**

With your permission the chat sessions will be recorded or videoed so that I have a reminder of what was said and can assess how well the trainees are managing the assessments. If you do not wish your discussion to be recorded but wish to participate that is not a problem so please let me know.

**Will the information be confidential?**

All the information will be kept confidential and if you prefer you will be able to use a made up which will protect you from being recognised on the recording. This means that you can speak freely about your thoughts and opinions without anyone being able to identify you individually. The information will be in a safe way so that it can only be accessed for use in this study and for teaching purposes.

**What if I change my mind about participating?**

Your participation is entirely voluntary and you can withdraw from the study at any time without needing to give a reason.

**Will I get any expenses covered?**

Unfortunately, owing to limited resources I am unable to offer any expenses for time and travel however all the sessions will be undertaken on days that you are already attending the clinic for regular appointments so there should not be any additional costs involved.

**What do I need to do now if I want to be involved?**

If you would like to be involved you will be asked to sign a consent form to say that you understand what is involved in the project and that you are happy to go ahead.

You will then be offered the chance to attend your first chat session either today or at a time in the near future.

**Please contact me if you have any questions about this project using the following e-mail:**

jsrees1@sheffield.ac.uk

**If you have any concerns about the project at any point, please contact me directly or the project supervisor using the e-mail below:**
Appendix 7: Training manual

PROJECT TO HELP US UNDERSTAND MORE ABOUT TREATING CLEFT PALATE IN ADULTS

Is operating on cleft palate later in life a good thing to do?
INTRODUCTION - what is a cleft palate?

At the moment many people in this country have a hole in the roof of their mouth called a cleft palate. This is a condition that people are born with and is often associated with a split lip known as a cleft lip. A cleft happens because the skin and bone in the face don't join together properly as the baby develops leaving gaps or 'clefts' in and around the mouth when the baby is born.

Every cleft palate is different, sometimes it is just a tiny hole, sometimes it can be a huge hole covering almost the whole palate or it can be any size in between. Some examples of what they look like can be seen below:

Because it is hidden inside the mouth a cleft palate may not be noticed, particularly if it occurs without a cleft lip. Sometimes a cleft palate is left untreated even when the lip was fixed when still a baby or a young child.

A cleft palate can cause many problems if it is not fixed because it affects speaking, eating and drinking.

WHY DO CLEFTS OCCUR?

Clefts are very common and there are many myths and stories about why they happen. The exact cause of them is not known. There are some things that may make them more common though.

- Sometimes more than one person in different generations of a family may be born with a cleft. When this happens it is known as a 'genetic problem'.
- Sometimes clefts occur because of things happening during pregnancy such as mother's illness, smoking and alcohol, taking of drugs and medication or a deficiency in vitamin B12 or folic acid.
WHY IS IT A PROBLEM?

Most clefts can be treated with one or two operations when the child is still a baby but if it is not treated it can cause many problems.

Because of cultural beliefs and a lack of understanding people can be embarrassed and ashamed if their child is born with a cleft. Because of this children may be abandoned or the families shunned and no longer accepted into normal society.

If a child has a cleft lip they look very different and are often teased because of it. A cleft palate causes difficulty eating and speaking which makes children difficult to understand and causes food and liquid to dribble from their nose which is another reason that children are teased.

Children with clefts are often not allowed to attend school they may be hidden away by the family and cannot attend social occasions, find a job, get married or live a normal life.

People who have a cleft palate often find it difficult to make themselves understood, particularly when speaking with strangers who are not used to hearing them speak. For this reason people with clefts are often reluctant or find it impossible to do everyday things like shopping at the market, speaking with friends or answering the phone since they know they won’t be able to communicate properly. This is very frustrating for them and makes them feel isolated and lonely. It also means that they cannot become independent because they rely heavily on friends and family to help them on a daily basis.

Because people with untreated clefts are often destined to live as outcasts and as objects of ridicule they are often depressed and live a lonely existence in isolation.
WHAT CAN BE DONE FOR CLEFT PALATE THAT HASN'T BEEN TREATED

It is possible to treat a cleft palate in adulthood but the operation is generally more difficult than it would be if it were performed when the patient was still a baby. It is also more difficult to predict how well the operation will turn out and whether or not it will make things better or not when the operation is done for older patients.

SPEECH

Because we learn to speak when we are very young, it is very difficult to change the way we produce words when we are older. Learning to speak with a cleft palate is often very difficult because the roof of the mouth is the wrong shape and it allows air to escape through the nose which changes the way sounds are made. This is why it is often difficult to understand people with a cleft palate.

Although you would expect speech to improve if the hole was closed by an operation, it does not always work like that, sometimes it does not improve speech as much as you might hope.

At the moment it is difficult to know which operations will work and which ones won't and we need to collect information about this in order to decide.

EATING AND DRINKING

We think that the problem of food and liquid coming down the noses of people with clefts is likely to improve following surgery but because we haven't done enough research we don't know for sure.

TREATING CLEFT PALATE IN BABIES AND YOUNG CHILDREN

Repairing Cleft lip and palate in young children is a very common and successful operation. Many thousands of operations are carried out around the world every day and lots of information has been collected to find out how successful they have been. Because of this we know that an operation which costs only a few hundred dollars per patient, will very likely allow a child to lead a full and normal life. Knowing this fact means that many organisations will provide funding so that the operations can be done for free because they know it is worthwhile and will help a lot of people.

This type of decision making is known as a cost/benefit analysis. If an organisation has a certain amount of money to spend, they want to know exactly how much benefit people will get as a result. So they need as much information as they can in order that they can decide which operations they will fund and which ones they won't. It's a bit like being in the market and trying to decide whether to spend your money on a big bag of rice which will help everyone in the family or spending it all on a new pair of shoes which will only benefit one person.
TREATING CLEFT PALATE IN OLDER CHILDREN AND ADULTS

Although some older patients are treated for cleft palate it is not very common. This is partly because we don’t know if it will be successful and we don’t want to offer people an operation if we don’t know whether it will make a difference or not. Also, because we don’t have enough information about these operations, it is difficult to find funding because fund holders would rather spend money on things which they know will definitely make a difference. This is why there is plenty of opportunity to treat cleft palate in young children but much less as people get older.

WHAT DO WE WANT TO DO?

We want to find out whether operating on cleft palate later in life is a good thing to do or not. We have lots of questions about operating on older patients which we want to find the answer to such as:

- Will it help people to be understood better?
- Will it stop food and drink coming from their noses?
- Will it make them happier?
- Did they think it was worthwhile and if so in what way?
- Was it more successful if cleft was very big or very small or didn’t it make a difference?

In order to answer these questions and many more, we need to collect information about all the operations we do for cleft palate in older patients. We will then use it to decide what to do for the best: if we find that an operation can make things better we hope we can get funding to do this for more patients. If we find out that it doesn’t help we can explain that to patients and try and help them in other ways but avoid an unnecessary operation.

HOW WILL WE COLLECT THE INFORMATION?

Most of the information we need can be simply collected by talking to the patient and filling in a questionnaire which we have already prepared. You will have a chance to use this and ask questions during the workshop session.

For the final part of the questionnaire you will be asked to assess how much you think the cleft is affecting the person in their everyday life. This will be done using a system called the 'Therapy Outcome Measure'. The rest of this manual gives a brief introduction to the system for you to look through.

Don’t worry too much if it seems complicated, it really is very simple to use and it will be explained fully during the workshop session. Just have a read through and look at the case scenarios for now, you will have a chance to have a go and ask questions during the workshop.
WHAT IS THE 'THERAPY OUTCOME MEASURE'?

An outcome measure is simply a system which records the affect or 'outcome' of an event. In this case the 'event' is the cleft palate and the 'outcome' is the way in which the person is affected by it.

In order that we can work out if things have changed following the operation, we first need to make a record of how the patient feels beforehand and then compare this with how they feel afterwards. This means that we will repeat the same questionnaire before surgery, just after the surgery and then again about 6 months later. By comparing the results we should be able to decide which things got better, which things got worse and which things stayed the same.

The system we are going to use looks at four different things which may change by operating on the cleft:

**IMPAIRMENT - HOW BIG IS THE CLEFT?**

At the time of the operation the surgeons will measure the size of the cleft by taking a photograph inside the mouth with a tiny ruler. This will allow us to see how big the cleft was from front to back and from side to side. In order that we can see the difference 6 months later we will need a similar photograph to compare it with and we will explain how this can be arranged nearer the time.

**ACTIVITY - HOW WELL THE PATIENT CAN PERFORM ACTIVITIES?**

To assess this you will need to talk to the patient and ask them questions to decide how difficult they find it to perform various activities. There is a list of these at the end of the manual but they include things like: making themselves understood by friends and family or complete, how much food and liquid comes down their nose when they are eating and drinking, how much help they need doing everyday things like shopping or answering the telephone.

**PARTICIPATION - HOW MUCH ARE THEY PREPARED TO PARTICIPATE IN THINGS AND SOCIALISE?**

For this section you will need to find out whether they have been willing to attend school or college? do they go to social events or eat in public? will they go shopping in the market and mix with strangers?

**WELL BEING - HOW DO THEY FEEL ABOUT DOING THINGS?**

This section is all about how the person feels emotionally; how anxious do they get socializing? Do they get embarrassed, angry or frustrated when people don't understand them? Do they feel generally happy or unhappy? Are they out-going or withdrawn?

By answering these questions we hope to find out how the patient feels about their cleft, how it affects them on a daily basis and if these things get better or worse or stay the same after the operation.

**DOES THIS SOUND ALL VERY COMPLICATED?**

As I mentioned before, don't worry if it doesn't seem very clear at the moment, we will practice doing this at the end of the session and there will be plenty of time to ask questions then.
CASE SCENARIOS

CASE SCENARIO 1
JUANA

BEFORE SURGERY
Juana was born in Mexico and had a cleft lip and palate. She was teased a great deal because of how she looked and how different she sounded but she was determined to finish school so she ignored the bullies. Unfortunately Juana’s family were killed in an accident when she was only 10 so she became an orphan. With no family support and very few friends Juana became very lonely and depressed. Although her speech was quite good and most people understood her, she was shy and hid away from everyone. She didn’t like the way she looked or sounded and because she was embarrassed, she rarely went out or socialised. Even after she got married and had two beautiful children she remained depressed and often thought of taking her own life. She felt responsible for the teasing that her children suffered because of her condition.

AFTER SURGERY
Juana’s husband and children were worried about her and persuaded her to get some help. Although her speech was quite good she had some speech therapy which helped her become more confident communicating and she had a small operation to improve the appearance of her lip. She also had some help to manage with her depression and is gradually starting to feel more confident. After 25 years of humiliation she is starting to feel better about herself and learned to be proud of herself and her family. She is still shy and prefers to be with people she knows but she will go to the market on her own to do the family shopping and go to celebrations but she is unwilling to speak on the phone or mingle with strangers unless her family are by her side.
CASE SCENARIO 2

AVEROOT

BEFORE SURGERY

Averooot is a teenage boy who was born in India with a cleft lip and palate. Although he had surgery to repair the cleft when he was age 6 – 12 months, unfortunately a small hole was left in his palate which sometimes let liquid to come down his nose when he drank. He sounded a bit funny when he spoke but, unless he tried to talk too fast or too loudly, people could usually understand him. He had many friends at school who looked after him when he got frustrated and protected him when he got bullied. He enjoyed going to the cinema and spending time with his friends but he would get anxious about going out shopping alone or meeting and socialising with strangers in case they would laugh or not understand him. Although he liked to go to social events like weddings and celebrations he would get anxious if he needed to eat in front of people because he was embarrassed when liquid leaked from his nose.

AFTER SURGERY

When Averooot was 16 he had an operation to fix the hole in his palate. He can now eat and drink without the risk of liquid leaking from his nose. His speech has got so much better since the surgery and it no longer worries him. He is happy to mix with strangers, he can be understood on the phone and he has started a new course at college. Sometimes he gets upset because he misses a lot of college because he has to attend many hospital appointments to straighten his teeth and will have more surgery to make his lip and nose look better but he is a generally contented and outgoing young man now that his speech is no longer a concern.
CASE SCENARIO 3
NOSANTUI

BEFORE SURGERY
Nosantui is from Kenya. She is 23 and lives in a traditional Masi community. She knew her lip and palate were unusual but did not know there was anything that could be done to fix them. Most people in the village could understand Nosantui with the help of pointing and gestures, but her speech was not good which made her too shy to leave her village. Her friends realised that people would not understand her in the town so they took all her merchandise to market and did her shopping for her. Although she went to all the village festivals and celebrations she usually hid at the back of the crowd wishing she was brave enough to join in. The villagers were used to the fact that Nosantui could not eat without food and liquid coming down her nose but she found it very embarrassing and preferred to eat alone. When Nosantui went to Nairobi for her surgery it was the first time she had ever left her village. She was very scared and it took a great deal of persuading to make her go.

AFTER SURGERY
When Nosantui first saw her repaired face she forgot to be scared of Nairobi, she was really excited about returning to her village to show her friends her new smile. Now her palate is repaired she is happy to eat in public because the food doesn't come down her nose so often. Although her speech is not much clearer, Nosantui feels more confident and is happy to use gestures with strangers to make herself understood. Nosantui is much more confident and she now has to persuade her friends to take her to the market because she enjoys seeing all the new things. She joins in all the dancing and proudly participates at every festival and is even starting to sing now that she is so much more confident about her appearance and willingness to speak.
CASE SCENARIO 4
LOKHI

BEFORE SURGERY
Lokhi is a 15 year old boy Assamese boy whose is known in his remote hillside village as “happy boy”. Even if his family had realised that his cleft could be repaired they could never have afforded the journey to town. Although his family and the villagers were taken aback by his appearance when he was born, because he was such a happy outgoing child, they soon didn’t notice his cleft anymore. Lokhi loved school and played with all the other children. He was always cheeky and liked to be centre of attention during village festivals. Nobody could understand what he was trying to say but he could make himself understood by pointing and waving his arms or he would simply write things down and everyone was patient and took time to understand him. The fact that eating was difficult and messy did not seem to matter to anyone, he just laughed and other people laughed with him. Everyone in the village liked 'happy boy'.

AFTER SURGERY
Lokhi loves his new smile and certainly prefers the way he looks now. He is getting better at talking more clearly and his eating has improved enormously and is a much less messy affair. People can usually understand him now, as long as he doesn’t talk too fast or get too excited. He is still a cheeky teenager, energetic and full of fun and still the centre of attention at all the village festivities. He is hoping to go to college and university and would like travel the world and make lots of new friends in the future.
CASE SCENARIO 5
KIRAN

BEFORE SURGERY
Kiran is a 14 year old Assamese girl who was born with severe breathing problems. She spent a lot of time in hospital because she couldn’t feed and her family were told she would be unlikely to live. Because she was so sick she didn’t have an operation for her cleft lip and palate. The village thought that she was cursed which is why Kiran looked the way she did and the family were excluded from the community. The family were made to leave the village and could not find work so they couldn’t afford to return to the hospital for treatment. Kiran went to school sometimes but was teased because nobody could understand her so she was unhappy and stopped going. It made her shy and lonely and she spent her time helping at home and looking after the other children. She could make a few words which the family could understand but usually she relied on pointing and gesturing. Nobody had no time to teach her to read or write. It made her sad that she couldn’t join in with the other children. She was worried about what would happen to her when she got older and there was nobody to look after her which made her withdrawn and depressed.

AFTER SURGERY
Kiran is much happier about her appearance since her surgery and will venture outside but never on her own. She is still shy and hasn’t got any friends, even though her speech is better nobody outside the family can more than a few words and Kiran does not like talking with strangers in case they can’t understand her. She no longer finds that food or liquid come down her nose when she eats but she is still too shy to eat or drink in public. Kiran gets very upset when she watches other children playing and gets frustrated and angry because people don’t understand her but with the help of her family she is starting to get more confident.
CASE SCENARIO 6
KEO NEANG

BEFORE SURGERY
Keo Neang is 81. She’s from Singapore and has a small cleft in her lip and a tiny cleft in her palate. She has always looked that way and the people who know her do not really notice anymore. She was teased a little bit at school but the other children soon accepted her and didn’t bother any more. Everybody could understand her and food and fluid didn’t come down her nose when she ate so the cleft palate did not bother her. Keo Neang left school when she was 13 so she could help work on the land and help her mother in the home but that was usual in her village and not because she had a cleft. She was never particularly bothered about her cleft and she was able to get married and have four beautiful children and now has 20 grandchildren. She is very well respected and plays a major part in the decision making in her village.

AFTER SURGERY
Keo Neang went to her screening appointment by travelling 30km on a motorbike taxi. She was operated on the following day and loves her new straight lip, although it made her wish she hadn’t waited so long to have the treatment. It hasn’t changed her life very much, her speech is no different and the way she eats is just the same but she enjoys seeing her new face in the mirror everyday and is happy the cleft been repaired. She enjoys teaching the others people in her village not to be scared of surgery and persuades them to be brave and not wait so long to have their treatment as she did.
Before Surgery

Jacqueline was born with a cleft lip and palate in Rwanda 16 years ago. She was a very timid little girl but her mother did not want her to miss out on her education and made her go to school. Jacqueline would come running home from school everyday crying but her mother insisted that she went back because she knew it was important. Sometimes she hid away but one day two girls came to make friends with her and tried to stop her getting bullied. They encouraged Jacqueline to speak out loud and gradually people got more used to her unusual speech and eventually most people could understand her. She was still shy and didn’t like talking with people she had not met before but Jacqueline was very bright and worked hard. She passed all her exams and even helped her friends to study for theirs. Because she had good friends she became more confident and eventually would go to the market on her own and take part in festivities even though she was still anxious. She worried that she would find a husband or have children of her own because of her cleft so although outwardly she appeared to be happy, inside she was quite upset and depressed about her cleft.

After Surgery

When Jacqueline had her surgery it transformed her life. She practiced everything her speech therapist taught her and steadily got easier to understand. This made her more confident when chatting with strangers and now almost everybody can understand what she is saying. She even uses a telephone which she would never have been brave enough to do before. Because Jacqueline worked so hard she won a scholarship to go to a city college and her ambition is to train as a doctor so she can help other children with clefts. She is now pleased that her mother forced her to go to school and doesn’t regret it although she hated it at the time. She doesn’t have a boyfriend but she is sure that she will one day marry and have a family which she could not have done before her cleft was repaired. She now feels like a normal happy student just like everyone else around her.
CASE SCENARIO 8
DEMITRIA

BEFORE SURGERY

Demetria is an 18 year old Peruvian girl. Her cleft lip and palate was so severe that she was teased badly and didn’t go to school at all. Her brothers and sisters taught her to read and write and she could speak clearly enough for her family to understand her. Unfortunately she found speaking very difficult and because she was shy she spoke very quietly which made it difficult to hear so strangers did not understand her properly. When she ate she liquids would sometimes pour out from her cleft palate and come down her nose. This embarrassed her and she refused to eat in public so she hid away at home when there were festivals in the village. She usually avoided leaving the house when she could but kept her face covered and would look at the ground if she did go outside so she couldn’t see people staring. She wished she could join in when she saw other children playing but she never made any friends of her own to play with.

AFTER SURGERY

After Demetria’s surgery she felt much better and is now proud of her appearance. She loves to go outside and mix with other people and happily goes to the market on her own because people can almost always understand her now, even though her speech is not perfect. She realises that it was not her fault that she had a cleft lip and palate and has explained this to the people in the village. Now that they understand the villagers are no longer scared of her and don’t stare. They have embraced Demetria which makes her much happier. She is now included in all the festivals and celebrations and she has her first boyfriend who is very supportive of her. She is also happy to eat in public and is proud to leave her face uncovered which makes it even easier for people to understand her. She works as an assistant in the primary school where she is also allowed to study so is gradually catching up with her education.
CASE SCENARIO 9
KAZA

BEFORE SURGERY

When Kaza spoke the words would not form properly. However much he tried to say the letters 'p' 'b' and 'm' they came out all wrong. Even if people were patient and listened really hard they could not understand even a quarter of what he was trying to say. It made simple things like shopping or speaking on the telephone impossible. This used to make him angry and frustrated and sometimes he used to lash out and want to fight even though he was normally a quiet gentle man. People then became scared of him and kept away so he became lonely and isolated, rarely venturing out into town. He felt downtrodden, miserable and thought nobody cared.

AFTER SURGERY

Kaza saved every penny he could to get to the hospital. The surgery transformed his life. When he went back to his wife in his remote Congolese village she hardly recognised him. Not only because of how he looked but also because of how he was. Kaza was no longer frustrated and angry, he was calm and proud. His speech was not perfect but because he was more relaxed and spoke calmly people could understand him better. He still finds it difficult to make himself understood on the telephone but the surgery has made him a much happier man because he feels he can now play an active part in village and family life and enjoys socialising particularly now he is so well liked in his village.
CASE SCENARIO 10
ISAAC

BEFORE SURGERY

Isaac was a clever boy. He was happy to go to school as long as he was not asked to read out loud and could sit at the back where nobody could see him. His teachers respected this and his classmates hardly noticed his cleft. Isaac always helped them with their work when they got stuck so they were happy he was there and Isaac was glad to help. He could be understood most of the time but he preferred to write things down rather than to speak out loud because that way everyone understood him. Isaac was very good at football which made him a popular boy, everyone wanted him on their team. He really loved to play because it was one time that his cleft did not make any difference, he was just like all the other boys.

At home things were more difficult because his father was embarrassed about Isaac's cleft, he thought it was his fault and didn't want to accept that Isaac was his son. Isaac loved his father but it made him sad that he would not look at him. If Isaac was careful he could eat well but every now and then a little bit of liquid would escape from his nose and upset his father so he was not allowed to eat at the same table. His brothers also looked down on him which made Isaac feel abandoned even though he had lots of friends at school.

AFTER SURGERY

One day Isaac's father took him to have treatment for his cleft. It hasn't really changed Isaac's appearance or ability to speak clearly and he still gets an occasional escape of liquid from his nose if he drinks too quickly. His siblings have moved away and Isaac now lives with his mother so he can eat at the family table for meals and his father acknowledges him as his son which makes Isaac a little happier. He still has lots of friends and is studying science at college and continues to be a football star at his college. He is confident that he will get a good job, a wife and family which is all he ever wanted.
Appendix 8: Information sheet for pilot phase 1

Participants information sheet - Phase 1 question 1

TITLE: Piloting the training manual relating to a research project relating to Intervention For Cleft Lip And Palate In Adulthood

Name, position of Researcher: Joanna Rees. PhD Student. University of Sheffield, Department of Human Communication Sciences

Invitation to participate
You are being invited to take part in the above research study. Before you decide whether to participate it is essential that you fully understand why the research is being done and what will be involved. Please take time to read/listen to this explanation leaflet before you make your decision.

What is the purpose of this study?
I am developing a training manual to train health workers in the developing world to collect information about people’s experiences of living with a cleft palate and the changes they experience as a result of surgical repair. By collecting this information I hope to gain a better understanding of the implications associated with untreated cleft palate and in turn to facilitate better treatment to suit the needs of such individuals in the future.

Before I introduce the training manual into a clinical environment I want to ensure that it is easy to understand and contains an appropriate level of information for its purpose. In order to do this I would like to arrange a small informal discussion group to look at the wording and content of the manual and make changes which would benefit it’s clinical application.

Who are the researchers and how is the research funded?
I am a PhD student in the Department of Human Communication Sciences at the University of Sheffield in the UK. I am also a dentist and surgeon working in the UK and I am undertaking this study because I believe that the continuing progress of cleft care in the developing world is something which is of great importance.

The information from this research will initially be used for purposes of my PhD, to collect information about the impact of intervention for cleft palate in later life in order and to clarify whether the procedure is beneficial. In addition it hoped that the resulting training manual and data collection system will continue to be used in cleft services worldwide to collect information for the purposes of quality control and benchmarking.

This study has been ethically reviewed by the Department of Human Communication Sciences Research Ethics Committee at the University of Sheffield.

Why am I asking you to be involved?
You are being asked to be involved in this project because you are a fluent English speaker with an interest in teaching and research and may be interested in participating in this project.

What will you have to do?
If you agree to participate you will be asked to read through the teaching manual that I will send you by email in early January. A few weeks after you have received the manual I will arrange a discussion group in a meeting room in Penrith, the details of which will be sent to you with as much notice as possible. The group will consist in total of a small group of local physiotherapists and myself and will be of an informal nature.

During the meeting I would like to work through the training manual and ask you for your verbal feedback regarding the wording and content. Although the manual is designed to be used together with a training workshop, it is important that the language and content are suitable for health-workers whose first language is not English. Your verbal contributions may be noted down by me, but your name or any identifying details about you will not be to ensure the information is kept confidential.
Once we have been through the manual together you will be asked to assess ten case scenarios using a data collection sheet similar to the one attached. The purpose of the assessment is to see if you consider each scenario to be easily understood and contain enough information to be effective teaching aids.

The session is expected to last approximately three hours in its entirety. Breaks and refreshments will be available throughout.

**What will happen to the information I provide?**

The notes I make during our discussion and the completed data collection sheet will be used by me to adapt the teaching manual before undertaking a full working pilot study. If the pilot study is successful then the manual will be introduced into the field to train health workers in developing countries to collect data relating to the psycho-social impact of living with untreated cleft palate and the affect of its subsequent treatment.

**Will the information be confidential?**

In order to keep your answers confidential I will use a unique identification number to label all information that I collect from you.

The information you provide will be used only for the purposes of developing the training manual and will be stored securely to ensure that it is only accessible by me personally. Direct quotes from my notes may be used for purposes of teaching and publication but, because your name will not be included in the stored data, it will not be possible to realise your identity from this material.

All material will be deleted at once the study is complete. The date of this is estimated to be October 2018.

**What if I change my mind about participating?**

Your participation is entirely voluntary and you can withdraw from the study at any time without needing to give a reason.

**Will I get any expenses covered?**

Unfortunately, owing to limited resources I am unable to offer any expenses for time and travel. There will however be refreshments provided during the sessions.

**What do I need to do now if I want to be involved?**

If you would like to be involved please complete the consent form attached to the end of this form to say that you understand what is involved in the project and that you are happy to go ahead. Please return a scanned copy of the completed consent form to jsrees1@sheffield.ac.uk

If you do not wish to participate you need do nothing but I would like to thank you for your time and consideration.

If you are intending to participate in this study please keep a copy of this information sheet for your records.

If you have any questions about this project please don't hesitate to contact me using the e-mail jsrees1@sheffield.ac.uk

If you have any concerns about the project at any point, please contact me directly or the project supervisor using the e-mail: t.muskett@sheffield.ac.uk

If you still feel that you complaint has not been handled to your satisfaction you will be able to contact the head of department Professor Patricia Cowell using the e-mail: p.e.cowell@sheffield.ac.uk. If you are still dissatisfied, please contact the University Registrar at The Registrar and Secretary's Office, University of Sheffield, Firth Court, Western Bank, Sheffield S10 2TN, UK.
ASSESSMENT OF CASE SCENARIOS - example of data collection sheet

Please read the case scenarios in the research manual provided. Using a scoring from 1 to 5 please mark the appropriate box in the score sheet to indicate how easily are you able to assess the following from the information provided:

<table>
<thead>
<tr>
<th>ACTIVITY</th>
<th>The person’s ability to be understood</th>
<th>Problems the person has eating and/or drinking</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>PARTICIPATION</td>
<td>The person’s willingness to socialise and join in activities</td>
<td>The person’s willingness to speak to strangers</td>
</tr>
<tr>
<td></td>
<td></td>
<td>The person’s willingness to eat in public</td>
</tr>
<tr>
<td>WELL-BEING</td>
<td>The person’s level of anxiety/happiness/embarrassment</td>
<td>your general feeling of knowing the subject</td>
</tr>
</tbody>
</table>

**SCORING SYSTEM GRADED FROM 1 TO 5**

1 = All the information is available and assessment is made easily

5 = None of the information is available and assessment is not possible

<table>
<thead>
<tr>
<th>Before surgery</th>
<th>ACTIVITY</th>
<th>Ability to be understood</th>
<th>Problems eating and/or drinking</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>easily</td>
<td>1 2 3 4 5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>PARTICIPATION</td>
<td>Willingness to socialise and join in activities</td>
<td>Willingness to speak to strangers</td>
</tr>
<tr>
<td></td>
<td>easily</td>
<td>1 2 3 4 5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>WELL-BEING</td>
<td>level of anxiety/happiness/embarrassment</td>
<td>General feeling of knowing the subject</td>
</tr>
<tr>
<td></td>
<td>easily</td>
<td>1 2 3 4 5</td>
<td>Not at all</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>After surgery</th>
<th>ACTIVITY</th>
<th>Ability to be understood</th>
<th>Problems eating and/or drinking</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>easily</td>
<td>1 2 3 4 5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>PARTICIPATION</td>
<td>Willingness to socialise and join in activities</td>
<td>Willingness to speak to strangers</td>
</tr>
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<td></td>
<td>easily</td>
<td>1 2 3 4 5</td>
<td>Not at all</td>
</tr>
<tr>
<td></td>
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</tr>
<tr>
<td></td>
<td>WELL-BEING</td>
<td>level of anxiety/happiness/embarrassment</td>
<td>General feeling of knowing the subject</td>
</tr>
<tr>
<td></td>
<td>easily</td>
<td>1 2 3 4 5</td>
<td>Not at all</td>
</tr>
</tbody>
</table>

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Appendix 9: consent form for pilot

CONSENT FORM

Participant ID number

TITLE: Piloting the training manual relating to a research project relating to Intervention For Cleft Lip And Palate In Adulthood

Researcher: Joanna Rees. PhD Student. University of Sheffield, Department of Human Communication Sciences

Please read the following and tick the boxes where appropriate. Then return a scanned copy of the completed consent form to jsrees1@sheffield.ac.uk or by post to Joanna Rees, 3 Inglewood Terrace, Penrith, CA11 7SZ.

<table>
<thead>
<tr>
<th>Date</th>
<th>Please initial box</th>
</tr>
</thead>
<tbody>
<tr>
<td>I confirm that I have read / understand the information sheet for the above study and am happy to participate</td>
<td></td>
</tr>
<tr>
<td>I have had the opportunity to ask questions and understand that my participation is voluntary and that I am free to withdraw at any time, without giving reason</td>
<td></td>
</tr>
<tr>
<td>I am happy for resulting material to be used for the purpose of this study, teaching and publication purposes where it is appropriately anonymised.</td>
<td></td>
</tr>
<tr>
<td>I understand that my responses will be kept confidential and that my name will not be linked to any of the research material</td>
<td></td>
</tr>
<tr>
<td>I agree to the use of anonymised quotes in publications</td>
<td></td>
</tr>
</tbody>
</table>

Please complete and sign the section below and thank you for considering to participate in this project.

<table>
<thead>
<tr>
<th>Name of Participant</th>
<th>Date</th>
<th>Signature</th>
</tr>
</thead>
</table>

<table>
<thead>
<tr>
<th>Name of Researcher</th>
<th>Date</th>
<th>Signature</th>
</tr>
</thead>
</table>

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Appendix 10: WHOQoL-BREF user agreement

User Agreement for “WHOQOL-100” and/or WHOQOL-BREF and related materials

This agreement is between the World Health Organization (“WHO”) and Joanna Rees. WHO hereby grants the User a nonexclusive, royalty-free license to use the World Health Organization Quality of Life Questionnaire and/or related materials (hereafter referred to as “WHOQOL-100” or “WHOQOL-BREF”) in User’s study outlined below. The term of this User Agreement shall be for a period of 1 year, commencing on (date) 6th March 2017.

The approved study for this User Agreement is:

| Study Title | Measuring the psycho-social impact of late intervention for cleft lip/palate - adaptation and application of the Therapy Outcome Measure (TOM) |
| Principal Investigator | Joanna Rees |
| Sample characteristics | Individuals who have not received optimal timing for cleft lip and palate but are now undergoing intervention at a number of cleft centres in India |
| Sample size | 20 |
| Treatment Intervention | Routine surgical cleft lip/palate repair |
| Total number of assessments | 40 |
| Assessment time points | Pre-surgery and 6 months post surgery |
| “WHOQOL-100” or WHOQOL-BREF version – Please specify language version(s) you would like to receive. | WHOQOL-DNCF - English and Punjabi and Hindi if available |
| Other measures | the therapy outcome measure TOM MIP |

This User Agreement is based upon the following conditions:

1. User shall not modify, abridge, condense, translate, adapt, recast or transform the WHOQOL-100 or BREF in any manner or form, including but not limited to any minor or significant change in wording or organization, or administration procedures, of the WHOQOL-100 or BREF. If User thinks that changes are necessary for its work, or if translation is necessary, User must obtain written approval from WHO in advance of making such changes.

2. User shall not reproduce WHOQOL-100 or DNCF, except for the limited purpose of generating sufficient copies for its own uses and shall in no event distribute copies of the WHOQOL-100 or BREF to third parties by sale, rental, lease, lending, or any other means. In addition, User agrees that it will not use the WHOQOL-100 or BREF for any purpose other than conducting studies as specified above, unless agreed in writing by WHO. In any event, the WHOQOL-100 or BREF should not be used for research or clinical purposes without prior written authorization from WHO.

10/17/13
1 of 3
3. User agrees to provide WHO with an annual update regarding activities related to the WHOQOL-100 or BREF.

4. User agrees to provide WHO with a complete copy of User’s raw data and data code books, including the WHOQOL-100 or BREF and any other instruments used in the study. This data set must be forwarded to WHO upon the conclusion of User’s work. While User remains the owner of the data collected in User’s studies, these data may be used in WHO analyses for further examining the psychometric properties of the WHOQOL 100 or BREF. WHO asserts the right to present and publish these results, with due credit to the User as the primary investigator, as part of the overall WHOQOL-100 or BREF development strategy.

5. WHO shall be responsible for preparing and publishing the overall WHOQOL-100 or BREF results under WHO copyright, including:
   a. the overall strategy, administrative set-up and design of the study including the instruments employed;
   b. common methods used by two or more Users;
   c. the data reported from two or more Users;
   d. the comparisons made between the data reported from the Users;
   e. the overall findings and conclusions.

6. User shall be responsible for publications concerning information developed exclusively by User and methods employed only by User. Publications describing results obtained by User will be published in User’s name and shall include an acknowledgement of WHO. User agrees to send to WHO a copy of each such paper prior to its submission for publication.

7. WHO may terminate this User Agreement at any time, in any event. Should WHO terminate this User Agreement, User shall immediately cease all use of the WHOQOL100 or BREF and destroy or return all copies of the WHOQOL 100 or BREF. In the event of such termination, all other collateral materials shall be destroyed and no copy thereof shall be retained by User. Notwithstanding the return or destruction of the WHOQOL-100 or BREF and its collateral materials, User will continue to be bound by the terms of this User Agreement.

8. It is understood that this User Agreement does not create any employer/employee relationship. User and its affiliates are not entitled to describe themselves as staff members of WHO. User shall be solely responsible for the manner in which work on the project is carried out and accordingly shall assume full liability for any damage arising therefrom. No liability shall attach to WHO, its advisers, agents or employees.
Please confirm your agreement with the foregoing by signing and returning one copy of this letter to WHO, whereupon this letter agreement shall become a binding agreement between User and WHO.

WHO:

[Signature]

Dr. Somnath Chatterji
Health Statistics and Health Information Systems (HSI)
World Health Organization
Avenue Appia
Geneva 27
CH 1211 Switzerland

Date: 6th March 2017

USER:

By: Joanna Rees
Title: Dr
Institution: University of Sheffield
Address: 3 Inglewood Tce
Lowther Street
Cumberland
CA11 7SZ

Date: 6th March 2017
## Appendix 11: Code comparisons for the first 50 phrases appendix

<table>
<thead>
<tr>
<th>ID</th>
<th>Quote</th>
<th>Coding from 2nd reviewer</th>
<th>Coding from PI</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Ch01 [father] then can’t understand [but] now it is better</td>
<td>Being understood Parental concern</td>
<td>Change in communication</td>
</tr>
<tr>
<td>2</td>
<td>Ch01 [whether it could be fixed] that they didn’t know, the parents are uneducated we don’t know</td>
<td>Understanding of CLP Family social circumstances</td>
<td>Low education of family No knowledge of CLP Getting repair</td>
</tr>
<tr>
<td>3</td>
<td>Ch01 [finding out about repair] i was in Hyderabad and i heard about it with a advertisement</td>
<td>Understanding of CLP Learning about CLP</td>
<td>Knowledge about CLP Getting repair</td>
</tr>
<tr>
<td>4</td>
<td>Ch01 She had some small of holes there in palate so now it was clear [speech]</td>
<td>Understanding of CLP Being understood</td>
<td>Communication Surgical</td>
</tr>
<tr>
<td>5</td>
<td>Ch01 [father] she has faced a lot of problems in the communication</td>
<td>Being understood Socialising Parental concern</td>
<td>Communication Participation Ability to make friends</td>
</tr>
<tr>
<td>6</td>
<td>Ch01 They won’t be able to understand and make with other friends so she will face that problem</td>
<td>Being understood Marginalisation /socialising Parental concern</td>
<td>Communication Participation Ability to make friends</td>
</tr>
<tr>
<td>7</td>
<td>Ch01 [father] now is ok, speech is much better</td>
<td>Speech Being understood</td>
<td>Communication</td>
</tr>
<tr>
<td>8</td>
<td>Ch01 Now all are able to understand what she is speaking</td>
<td>Being understood Socialising</td>
<td>Communication Participation</td>
</tr>
<tr>
<td>9</td>
<td>Ch01 So now shopping and things it’s made life easier</td>
<td>ADL [activity of daily living] Life getting better</td>
<td>Daily life improved Participation Well-being</td>
</tr>
<tr>
<td>10</td>
<td>Ch01 Did you know anyone with CLP before? No ma’am</td>
<td>Understanding of CLP</td>
<td>Knowledge about CLP</td>
</tr>
<tr>
<td>11</td>
<td>Ch01 So now shopping and things it’s made life easier</td>
<td>ADL Life getting better [same as above]</td>
<td>Repeat phrase</td>
</tr>
<tr>
<td>12</td>
<td>Ch01 They won’t allow her go on her own, not yet [cultural not because of cleft]</td>
<td>Socialising but Relevant as not cleft but cultural</td>
<td>Cultural Participation</td>
</tr>
<tr>
<td>13</td>
<td>Ch01 She didn’t suffer any teasing in school because everyone was patient</td>
<td>Reaction of outsiders Education</td>
<td>Attitude of society/acceptance</td>
</tr>
<tr>
<td>14</td>
<td>Ch01 She didn’t really make any friends</td>
<td>Making friends Socialising</td>
<td>Not making friends</td>
</tr>
<tr>
<td>15</td>
<td>Ch01 Nasal regurgitation was a problem but she learned to avoid it by eating carefully but it was embarrassing</td>
<td>Regurgitation Emotion - embarrassment</td>
<td>Regurgitation Emotion</td>
</tr>
<tr>
<td>16</td>
<td>Ch01 Now everyone understands her, even when she speaks on the phone</td>
<td>Being Making friends ADL</td>
<td>Communication Participation Daily life</td>
</tr>
</tbody>
</table>
and she has friends and is able to participate in shopping and travelling

<table>
<thead>
<tr>
<th>Page</th>
<th>Ch</th>
<th>Text</th>
<th>Understanding</th>
<th>Emotion</th>
<th>Communication</th>
<th>Well-being</th>
</tr>
</thead>
<tbody>
<tr>
<td>17</td>
<td>Ch01</td>
<td>Nasal regurgitation has reduced but happens occasionally which is embarrassing.</td>
<td>Regurgitation</td>
<td>Emotion - embarrassment</td>
<td>Communication</td>
<td>Emotion</td>
</tr>
<tr>
<td>18</td>
<td>Ch02</td>
<td>Speech also improved... And they understand what he is telling</td>
<td>Speech</td>
<td>Being understood</td>
<td>Communication</td>
<td></td>
</tr>
<tr>
<td>19</td>
<td>Ch02</td>
<td>[before].whatever they eaten they get regurgitation [after] now it has improved ma’am</td>
<td>Regurgitation</td>
<td>Regurgitation</td>
<td></td>
<td></td>
</tr>
<tr>
<td>20</td>
<td>Ch02</td>
<td>Had financial problems in the childhood so that’s why they pay some amount of money to get it done</td>
<td>Family finance</td>
<td>Understanding CLP</td>
<td>Social status</td>
<td>Financial</td>
</tr>
<tr>
<td>21</td>
<td>Ch02</td>
<td>So they wait for after they are getting information about this free surgery so that’s why they came</td>
<td>Financial</td>
<td>Education about CLP</td>
<td>Financial</td>
<td></td>
</tr>
<tr>
<td>22</td>
<td>Ch02</td>
<td>They wants to join with friends but they wouldn’t understand or something small, but now he is also facing some problems</td>
<td>Making friends/socialising</td>
<td>Being understood</td>
<td>Emotion - frustration</td>
<td>Making friends negative</td>
</tr>
<tr>
<td>23</td>
<td>Ch02</td>
<td>Whenever he mingles with friends some sort of difficulties there ma’am speaking</td>
<td>Socialising</td>
<td>Being understood</td>
<td>Having friends</td>
<td>Participation</td>
</tr>
<tr>
<td>24</td>
<td>Ch02</td>
<td>They won’t understand some words, so that’s why they are asking frequently ‘what are you saying’</td>
<td>Being understood</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>25</td>
<td>Ch02</td>
<td>If mother is there with her she can explain to the friends</td>
<td>Being understood</td>
<td>Socialising</td>
<td>Lack of autonomy</td>
<td>Autonomy</td>
</tr>
<tr>
<td>26</td>
<td>Ch02</td>
<td>After surgery there is some sort of hole there now the doctor says that if this surgery Is also done he will be able to speak better</td>
<td>Understanding CLP</td>
<td>Knowledge about CLP</td>
<td></td>
<td></td>
</tr>
<tr>
<td>27</td>
<td>Ch02</td>
<td>Boys speech very good but reluctant to speak - looked to mother to do all the talking</td>
<td>Speech</td>
<td>Emotion - shy</td>
<td>Lack of autonomy</td>
<td>Emotion</td>
</tr>
<tr>
<td>28</td>
<td>Ch02</td>
<td>Would not do interview without mother as too shy</td>
<td>Emotion - shy</td>
<td>Lack of autonomy</td>
<td>Emotion</td>
<td>Well-being</td>
</tr>
<tr>
<td>29</td>
<td>Ch03</td>
<td>N/a</td>
<td>N/a</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>30</td>
<td>Ch04</td>
<td>What happened madam my family didn’t know anything about it they didn’t know anything about surgery</td>
<td>Understanding of CLP</td>
<td>Family knowledge</td>
<td>Knowledge about CLP</td>
<td></td>
</tr>
<tr>
<td>31</td>
<td>Ch04</td>
<td>Now my family feel upset that that they didn’t know to come for surgery</td>
<td>Understanding of CLP</td>
<td>Parental concerns</td>
<td>Emotion</td>
<td>Effect on family</td>
</tr>
<tr>
<td>32</td>
<td>Ch04</td>
<td>When i got my job here the doctor came and said to me to go for the surgery</td>
<td>Getting employment</td>
<td>Understanding of CLP</td>
<td>Employment</td>
<td>Knowledge about CLP</td>
</tr>
<tr>
<td>33</td>
<td>Ch04</td>
<td>It would give me confidence and my doctor is suggesting one further surgery</td>
<td>Emotion confidence</td>
<td>Understanding of CLP</td>
<td>Emotion</td>
<td>Well-being</td>
</tr>
<tr>
<td>34</td>
<td>Ch04</td>
<td>around 50% then I recovered and now more than 50%. I feel very happy for that</td>
<td>Emotion - happy</td>
<td>Emotion</td>
<td>Well-being</td>
<td></td>
</tr>
<tr>
<td>35</td>
<td>Ch04</td>
<td>I feel very happy for that i never thought it would happen</td>
<td>Emotion happy</td>
<td>Emotion - expectation</td>
<td>Emotion</td>
<td>Well-being</td>
</tr>
<tr>
<td>Ch04</td>
<td>[regurgitation] never had any problem with that</td>
<td>No regurgitation</td>
<td>Regurgitation</td>
<td></td>
<td></td>
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<tr>
<td>Ch04</td>
<td>[before] i can’t talk in any language, and fear and hesitation. Fear, fear, very much fear</td>
<td>Speech</td>
<td>Emotion fear</td>
<td>Communication Emotion</td>
<td></td>
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<tr>
<td>Ch04</td>
<td>The main thing is that if I speak someone will laugh on me they will mess with me and shame ..</td>
<td>Speech</td>
<td>Attitude of others</td>
<td>Teasing Emotion</td>
<td></td>
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<tr>
<td>Ch04</td>
<td>Not many people heard me</td>
<td>Speech</td>
<td>Emotion shy</td>
<td>Communication Emotion</td>
<td></td>
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<tr>
<td>Ch04</td>
<td>I want to make money but no one would employ me ‘why are you trying job’</td>
<td>Attitude of others</td>
<td>Emotion - fear</td>
<td>Employment Emotion</td>
<td></td>
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<tr>
<td>Ch04</td>
<td>I have not done any job and for that reason. Til 2005, I’ve not enjoyed any job .</td>
<td>Emotion frustration</td>
<td>Not finding work</td>
<td>Employment Finance</td>
<td></td>
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<tr>
<td>Ch04</td>
<td>I had my own business but in 2008</td>
<td>Finding work</td>
<td>Employment</td>
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<tr>
<td>Ch04</td>
<td>I applied for [and got] this job [working at the hospital]</td>
<td>Finding work</td>
<td>Employment Participation</td>
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<tr>
<td>Ch04</td>
<td>When i had gone for job before they would react to me they would laugh on me</td>
<td>Not finding work</td>
<td>Emotion</td>
<td>Employment</td>
<td></td>
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<tr>
<td>Ch04</td>
<td>[about getting a job] they told me and they feared me that way it has been very bad for me.</td>
<td>Finding work</td>
<td>Emotion - fear</td>
<td>Employment</td>
<td></td>
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<tr>
<td>Ch04</td>
<td>One line day they called on me we are going to do the surgery in this hospital – i go for the surgery and i am very happy for that.</td>
<td>Emotion happy</td>
<td>Employment</td>
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<tr>
<td>Ch04</td>
<td>Works with the public helping families coming for cleft treatment which he would not have dared to do before the operation.</td>
<td>Self respect</td>
<td>Socialising</td>
<td></td>
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<tr>
<td>Ch04</td>
<td>And now i have confidence for 8 year,</td>
<td>Emotion confidence</td>
<td>Employment</td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Ch04</td>
<td>Everybody [in the hospital] know me very well</td>
<td>Attitude of others</td>
<td>Friends Participation Employment</td>
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<tr>
<td>Ch04</td>
<td>And i am living my life</td>
<td>Emotion happy</td>
<td>Socialising</td>
<td>Employment</td>
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</tr>
</tbody>
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