Quality of life, psychological wellbeing and distress in those with an intellectual disability

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Declaration

I confirm that this thesis has not been submitted for any other degree or to any other institution.

Word count

Literature review	6598 excluding tables, references and appendices
	7665 including tables
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	14230 including tables, references and appendices
Research report	7973 excluding tables, references and appendices
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	9664 including tables and references
	13059 including tables, references and appendices
Total	14571 excluding tables, references and appendices
	27289 including tables, references and appendices
	27520 including tables, references, appendices and overall abstract

Overall abstract

Literature review: The present study is a systematic literature review of findings from 16 studies investigating the quality of life of adults with an intellectual disability. It was concluded that those with intellectual disability experience a lower quality of life than other groups. The interrater reliability between self-reports and proxy reports were found to be fair. However, proxies were found to consistently underestimate quality of life for individuals with intellectual disability, and physical wellbeing was identified as a problematic domain for agreement between self-report and proxy reports. Employment and good social or family supports emerged as factors most important to good quality of life. Those with severe or profound intellectual disability were under represented in the literature. Clinical implications and recommendations for future research are discussed.

Empirical report: There is limited research into how psychological wellbeing and distress are identified in those with a severe or profound intellectual disability. Therefore, the present study aimed to explore carer's view on how these inner emotional states are identified in those they care for. A qualitative exploratory design was used and 18 paid carers and family members participated in semi-structured interviews. Analysis of transcripts yielded eight superordinate themes relating to: relationship; physical contact; body; vocalisation, things that affect their emotions; facial expression; unknown and telling their story. Agreement between the dyads was fair. Clinical implications and recommendations for future research are outlined.

Acknowledgements

Firstly, I would like to thank the family and carers who took part in this research. I have appreciated their candour, and without their time this project would not have been possible. I would also like to thank Professor Nigel Beail for his guidance and support. Thank you to Lee and Frankie, for the support and levity they brought to this journey. I would also like to thank Sandy, my mother, who has supported and encouraged me throughout the years in my efforts to pursue a career in psychology. Last but not least, I would like to thank my children, Ethan and Eve, who have had to share me with the course. I could not have done this without your love and patience. We only have one life, so you may as well follow your dreams.

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Section one: Literature Review

Quality of life for those with an intellectual disability: A systematic review

Abstract

Objectives: There is limited literature examining the quality of life and areas of impact in those with an intellectual disability. The literature is also not clear how well individuals and their proxies agree on quality of life measures. To date, there has been no systematic review of the literature that includes an emotion component in the assessment of quality of life. Therefore, this systematic review summarises and synthesises the literature: investigating the quality of life in those with an intellectual disability; the areas of impact; and the inter-rater reliability between individuals and their proxies.

Method: A search of five databases comprising: Scopus, MEDLINE, PsycINFO, The Cochrane Library and CINAHL was conducted to identify articles published between January 2007 and February 2018. These studies were then assessed against inclusion criteria and a quality rating checklist.

Results: Sixteen quantitative studies met the inclusion criteria. Overall, agreement between individuals and their proxies was fair. There was good agreement overall on the emotion domain. Proxies were found to consistently underestimate quality of life for individuals with intellectual disability, and physical wellbeing was identified as a problematic domain for agreement between self-report and proxy reports. Employment and good social or family supports emerged as factors most important to good quality of life. Those with severe or profound intellectual disability were underrepresented in the literature.

Conclusions: It was concluded that those with intellectual disability experience a lower quality of life than other groups, but that employment and good social and family support can have a positive impact on quality of life. It was also concluded that using proxy measures, as well as self-report, is a valid method to get different

perspectives and that further research is needed for those with severe or profound intellectual disability.

Practitioner Points

Clinical Implications

- Obtaining self-report and proxy measures is a useful way to get different perspectives of quality of life. Clinicians should be aware that proxies may underestimate an individual’s quality of life.
- Consideration of family and social support networks, as well as potential employment opportunities, could inform care planning and service provision and improve an individual’s quality of life.

Limitations

- The results are less representative of women and those with a severe or profound intellectual disability and were not ethnically diverse.
- Some research has not been accounted for in this review. Prominent journals were not hand searched, the grey literature was not consulted, and only papers written in English were considered.

Understanding, measuring, and improving the human experience is a major goal of researchers, individuals and governments (Costanza et al., 2007). There has been a change in focus away from a belief that medical or technological advances alone will result in an improved life experience, towards an understanding that family, community, personal and societal well-being, values, perceptions and environmental conditions are the key to understanding and improving individuals' lives (Schalock & Felce, 2004). In research, the overall assessment of this human experience has been more commonly termed *quality of life* (QOL).

There is some disagreement about the similarities and differences between the QOL of those with an intellectual disability (ID) and those without, with some studies showing people with ID having poorer QOL (Sands & Kozleski, 1994; Watson & Keith, 2002). However, others show no differences between the two groups (Pretty, Rapley, & Bramston, 2002). Given the increased risk of exposure to poverty, social exclusion, poor health, disempowerment, victimisation and abuse for people with ID (Emerson & Hatton, 2008), research that aims to understand and improve QOL is vital. Assessment of QOL for those with ID is particularly important in health care systems to enable planning and implementing personalised care packages and interventions. Bertelli and Brown (2006) stated that QOL should be considered as the ultimate keyword for those who work with people with an ID.

QOL models take the essential dimensions of a person's life situation and operationalise them for use in enhancing and evaluating personal outcomes (Buntinx & Schalock, 2010). There is an general agreed consensus around Schalock and Verdugo's (2002) model of QOL for those with ID, which is a multidimensional framework with subjective and objective factors, and takes into account the personal and environmental characteristics of the person. They state that core QOL domains are the same for all people, although they can vary individually in relative value and importance (Schalock, Keith, Verdugo & Gomez, 2010).

In this framework there are eight core QOL domains, which have been cross-culturally validated (Brown et al., 1998; Schalock et al., 2002). They are: self-determination; Social inclusion; material wellbeing; physical wellbeing; emotional wellbeing; personal development; interpersonal relations; and rights (Cummins 2004; Schalock et al., 2002; Wang et al., 2010). Some ID QOL models do not include an emotional/psychological wellbeing component, and focus rather on other more objective or environmental characteristics. In recent years emotional wellbeing, however, has begun to be considered a key component of the QOL for those with ID (Cummins, 2005; Schalock et al., 2002; Schalock & Felce, 2004). However, it is not clear how well individuals and proxies agree on this domain. As such, the following review will include studies that have an emotional wellbeing component in their assessment of QOL.

In measuring QOL, Schalock et al., (2002) suggested that a valid assessment should comprise of both self-report and proxy measures. However self-report ratings can be difficult to obtain from people with ID, when compared to those without an ID, because of their lower cognitive abilities (McGillivray, Lau, Cummins & Davey, 2009). Because of this, proxy ratings are often utilised, usually from paid carers or family members. However, the research has shown a mixed picture of the inter-rater reliability between self-report and proxy reports, with some studies showing agreement between the raters and others showing significant differences (McVilly, Burton Smith, & Davidson, 2000). It is particularly difficult to measure QOL for those with severe or profound ID, as it is not possible for the individual to self-report on their QOL, therefore any proxy measures cannot be compared to assess validity. Perhaps, as a result of this, the expanding body of research on QOL and people with ID, has, it has been suggested, given little attention to people with severe or profound ID (Lyons, 2005). This review will, therefore, examine reports of interrater reliability between self-report and proxy report measures of QOL, as well as identify how much of the literature includes those with a severe or profound ID.

To summarise, the aims of this systematic literature review are to synthesise the literature investigating the QOL of people with ID. Existing literature does not include a systematic review on QOL for those with ID, with emotional wellbeing as a component. Additionally, it is not clear how much agreement exists between self-report and proxy reports of QOL. It has been suggested there is a lack of QOL data on those with severe or profound ID. Therefore, the available literature will be reviewed to:

1) Identify any difference in QOL for those with ID.
2) Examine any areas of impact on QOL for those with ID.
4) Examine the inter-rater reliability of self-report and proxy reports of QOL, paying particular attention to the emotion domain.
5) Identify whether the studies included those with a severe and profound ID.

Method

Search strategy

Studies were identified by systematic searches of Scopus, Medline, Psycinfo, Cochrane Library and Cinahl, time limited from January 2007 to February 2018. Titles and abstracts were screened for eligibility. When an indication of a cross sectional study on the QOL of those with ID was found, the entire article was reviewed for possible inclusion. References and citations of the selected papers were checked for additional papers. The following criteria were applied in the search terms: quality of life OR QOL AND intellectual disabil* OR learning disabil* or mental retardation anywhere in the document, AND adult as a keyword.

A filtering process, adapted from Moher, Liberati, Tetzlaff, and Altman (2009), of the reviewed papers was used and is shown in figure 1. In total, 17 articles were included. Methodological quality was assessed using the AXIS checklist (Downes, Brennan, Williams & Dean, 2016).

Inclusion and exclusion criteria

Articles published in English and with a study sample concerning adult populations (16+ years), where an emotion domain was used as part of the QOL measure, were considered for inclusion. During the first stage of the search process, any article that focused on the QOL of those with ID was included. Papers were excluded based on the following criteria: a main focus on epilepsy; a main focus on autism; interventional studies; studies validating an outcome measure and studies focused on learning difficulty. After titles were screened for the first stage inclusion and exclusion criteria, 187 articles were retained. During the second stage of the search strategy, the abstracts were screened and 39 articles were retained. In the third stage of the search, the full articles were reviewed and 22 articles were removed for the following reasons: using a child/adolescent sample; having a learning difficulty focus; validating of an outcome measure; being an intervention study or the level of ID not being expressly mentioned. Reference lists were then searched from the remaining articles and a further 2 papers were identified for inclusion. In total, 17 papers were then put forward for quality appraisal.

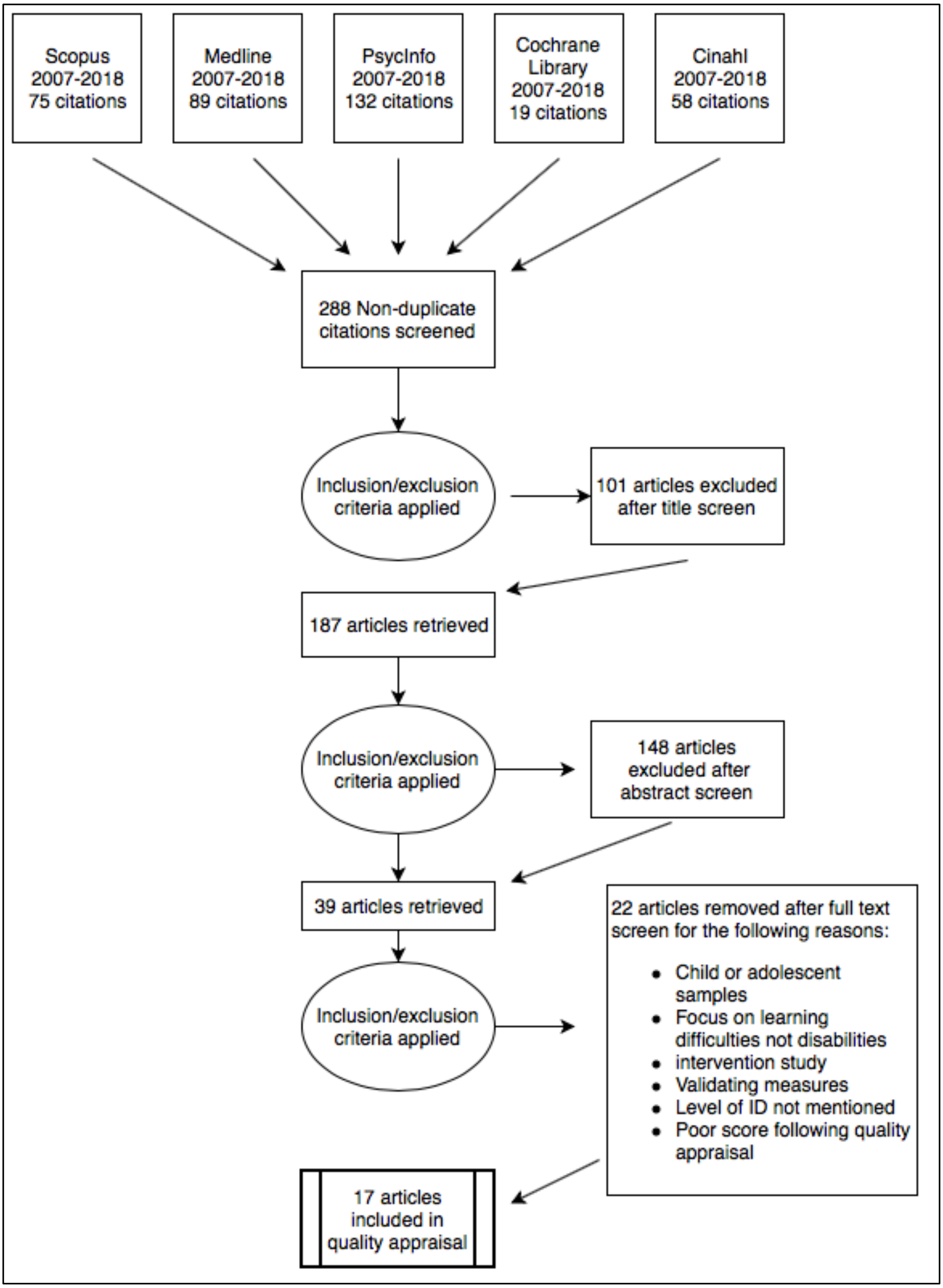

Figure 1. PRISMA flow diagram of study selection process

Quality appraisal

Quality appraisal is used to systematically assess and judge the reliability of a given article. The AXIS appraisal tool was specifically developed for appraising observational cross-sectional studies (Downes et al. 2016). It was designed to address issues most often apparent in cross-sectional research and to generally aid the reader to assess the quality of a given study. The AXIS has 20 items with a yes or no response format, and was adapted, in this case, for use in the present summary table as presented with an overall score out of 20 (yes=1, no=0). All papers were appraised by the author and 4 were chosen at random and scored by an independent rater. An interclass correlation analysis was performed assessing inter-rater reliability, with excellent reliability between raters being achieved. In total, 17 papers were retained and examined for the present systematic review.

Results

Overview

Table 1. summarises the key findings from the reviewed studies. The specific focus of the papers in this review have lent themselves to being grouped. Consequently, results are presented with papers in one of two groups (i) Self report QOL vs proxy views of QOL or (ii) variables or predictors of QOL.

Critique of papers

As show in Table 1, 17 of the papers in this review were assessed as having poor to excellent quality. Ratings were varied with scores from 7 to 19. Papers with higher quality ratings were characterised by robust study design, clear justification of the target sample, well defined outcome measures and justified discussions and conclusions.

Table 1.

Study characteristics

Author (Country)	Study aims	Outcome measure	Sample	Severe/ profound included	Residential status of persons with ID	Main findings	Quality rating /20	Included in review
Balboni, Coscarelli, Giunti & Schalock, 2013 (Italy)	To investigate self-report QOL & compare with carer's estimation of client point of view as well as their own view.	POS	176 adults with Mild, moderate & severe ID	Y	family/ Care home	Fair to poor correlation between self-report & proxy, but good to excellent correlation between self-report & carers estimation of clients point of view.	15	Y
Bertelli, Bianco, Rossi, Scuticchio & Brown, 2011 (Italy)	To investigate the relationship between individual QOL and family QOL	QOL-IP	27 adults with ID Aetiology of disability and how well person speaks, but no level of ID	Y (but categorised as level of support and language ability)	Residential home/ Family home	QOL perceived differently by those with ID and their family.	15	Y
Beyer, Brown, Akandi & Rapley, 2010 (UK)	To compare QOL for People with ID in employment and those not in employment	ComQol-I.	17 adults with ID	N	?	Higher QOL for non-ID workers. Higher QOL for those with ID in supported employment.	16	Y
Chou & Schalock, 2009 (Taiwan)	To compare QOL for People with ID in Taiwan with other countries	CCQoLI	233 adults with Mild ID.	N	Family home	Those in Taiwan have lower QOL when compared to other countries.	7	Y

Author (Country)	Study aims	Outcome measure	Sample	Severe/ profound included	Residential status of person with ID	Main findings	Quality rating /20	Included in review
Claes, Hove, Vandevelde, loon & Schalock, 2012 (Netherlands)	To investigate how supports strategies, environmental factors, & client Characteristics impact on QOL for those with ID	POS	186 adults with borderline, mild, moderate severe & profound ID	Y	Group residences/ Independent living	QOL sig. impacted by support strategies, living arrangements, status of employment and level of ID.	16	Y
Claes, Vandevelde, Hove, Loon, Verschelden & Schalock, 2012 (Netherlands)	To evaluate self-report vs proxy ratings of QOL	POS	161 adults with mild and moderate ID	N	?	Some differences between self-report and staff ratings but no significant differences between self-report and family ratings.	16	Y
Emerson & Hatton, 2008 (UK)	Explored factors associated with variation in self-reported wellbeing and sociability	Semi-structured interviews	1273 adults with ID	N	Private household/ Residential care	Association between socioeconomic disadvantage, contact with friends with ID, and well-being.	16	Y

Author (Country)	Study aims	Outcome measure	Sample	Severe/ profound included	Residential status of persons with ID	Main findings	Quality rating /20	Included in review
Koch, Vogel, Becker, Salize, Voss, Werner, Arnold, & Schutzwohl, 2015 (Germany)	To investigate self-report & proxy measures of QOL & the Impact of psychiatric symptoms, problem behaviour, medication & unmet needs.	WHOQOL-BREF & WHOQOL-DIS	102 adults with mild or moderate ID	N	Lives alone/ residential accommodation	Agreement between self-report and proxy was poor. Unmet needs and medication most important predictors of reduced self-rated QOL.	19	Y
Lombardi, Croce, Claes, Vandevelde & Schalock, 2016 (Italy)	Investigates the factors predicting QOL for people with ID	POS.	1,285 adults mild, mod & severe ID	Y	Family home/ residential home/ Lives alone	Outcomes can be predicted by providing activities aligned with personal needs/goals.	18	Y
Memisevic, Hadzic, Zecic, Mujkanovic, 2016. (Bosnia & Herzegovina)	Investigated demographic predictors of QOL in people with ID	POS.	152 adults with mild or mod ID	N	?	Level of ID and employment status were sig. predictors of QOL.	16	Y
Rey, Extremera, Duran, Ortiz-Tallo, 2012 (Spain)	Investigating the role of role of emotional competence on QOL in those with ID	SWLS	139 adults with ID	N	?	Emotional competence positive association with life satisfaction and happiness.	15	Y

Author (Country)	Study aims	Outcome measure	Sample	Severe/ profound included	Residential status of persons with ID	Main findings	Quality rating /20	Included in review
Schmidt, Green, Lucas-Carrasco, Eser, Dragomirecka & Fleck, 2010 (UK, Spain, Turkey, Czech.R & Brazil)	Investigating inter-rater reliability between self and family & paid carer rating of QOL in adults with ID.	WHOQOL-BREF.	614 adults with ID. Level not recorded	?	Lives alone/residential accommodation/ hospital	Moderate to strong association between self-report and both proxies.	14	Y
Sharfi & Rosenblum, 2016 (Israel)	Executive Functions, Time Organization and Quality of Life among Adults with ID	WHOQOL.	55 adults – level not recorded	N	?	Lower QOL for those with ID. Executive function of emotional control, initiation & emotional reaction sig. predictors of psychological QOL.	15	Y
Simoes & Santos, 2016a (Portugal)	To examine consistency between QOL self-report and two proxies	POS	69 adults with mild or moderate ID	N	Family home/ Care home	Moderate to excellent agreement between self and proxies, except for physical wellbeing domain, which was poor. Agreement was better with support staff than their family members.	16	Y
Simoes & Santos, 2016b (Portugal)	To investigate the differences between self-report & proxy measures of QOL in those with ID and factors that predict QOL	POS	1264 adults with mild or moderate ID	N	Own home/ Family home/ Care home	Self-report and proxy correlations were good to excellent. Factors of diagnosis, age, living circumstances, & daily activity strongly affected QOL.	16	Y

Author (Country)	Study aims	Outcome measure	Sample	Severe/ profound included	Residential status of persons with ID	Main findings	Quality rating /20	Included in review
Simoes & Santos, 2016c (Portugal)	Aims to compare the QOL of adults with and without ID and any predictors	POS.	1264 adults with mild or moderate ID	N	Own home/ family home/ Care facility	Sig. higher QOL for those without ID. Health status highest mutual predictor of QOL.	16	Y
Van Asselt-Goverts, Embregts, Hendriks, 2015 (Netherlands)	To investigate which social network characteristics relate to self-reported QOL for those with ID	IDQOL-16	33 adults with mild to borderline ID	N	Independent residence	Affection related to higher QOL.	18	Y

Note. POS= Personal Outcomes Scale (van Loon, Van Hove, Schalock, & Claes, 2008). QOL-IP= Quality of Life-Instrument Package (Brown, Raphael, & Renwick, 1998). ComQol-I= Comprehensive Quality of Life Scale-Intellectual Disability (Cummins, 1997). CCQoLI= Cross-cultural QoL Indicators Survey (Verdugo & Schalock, 2003). WHOQOL-BREF= World Health Organization Quality of Life Instrument, Brief (Angermeyer, Kilian, & Matschinger, 2000). WHOQOL-DIS= World Health Organization Quality of Life instrument for people with disabilities (Power & Green, 2010). SWLS= Satisfaction with Life Scale (Atienza, Pons, Balaguer, & Garcia-Merita, 2000). IDQOL-16=Intellectual Disability Quality of Life-16 (Hoekman, Douma, Kersten, Schuurman, & Koopman, 2001).

A limitation of the AXIS tool is that the checklist does not take in to account the psychometric properties of the measures used in the articles. The articles used in this review contain a range of QOL measures and the quality and validity of these measures has not been considered when assessing the study research quality.

One paper (Chou & Schalock, 2009) scored only 7 in the quality check as they did not report on some important participant demographics and had a less robust study design. This study was less reliable and therefore excluded from the synthesis.

Self-report and proxy views of QOL

Six papers had a focus on comparing the self-reported views of those with ID with those of a proxy. An overall total of 2,386 participants with ID, 58% men and 42% women, were included in the studies. Participants' ages ranged from 18 to 85 years. All the studies used a range of psychometric measures, observational methods or semi structured interviews. Only one of these studies (Balboni, 2013) reported on participants with a severe or profound disability. Four of these studies reported at least a moderate agreement between self-report and proxies, with two of the studies showing a fair or poor agreement.

Schmidt, Power, Green, Lucas-Carrasco, Eser, Dragonmirecka & Fleck (2010) investigated the agreement between self and proxy reports, from family and paid carers, and looked at any factors that may contribute to differences. They found overall, that both family and paid care proxy reports showed a moderate to strong association with self-reports of QOL, having good overall agreement on the emotion domain. In general, they found that people with ID rated their QOL a little higher than proxies with the exception of two items on the physical domain, and on the negative feelings domain, with a negative association reported between the self and proxy report. The authors conclude that, despite the proxy report showing high agreement with self-reports, they still recommend using both methods for future assessments of QOL. However, this study did not report on the level of ID of the participants. This may be due to using various national centres as its sampling strategy, and as different countries have slightly different level of ID parameters, this makes reporting on the ID level very difficult. A further limitation of this study was its use of opportunistic sampling.

Claes, Vandevelde, Hove, Loon, Verschelden, and Schalock, (2012) evaluated the consistency between self-report and direct observation by support staff as well as family members. They found some difference between individual and staff proxies ratings, and no significant difference in ratings from self-reports and family proxy respondents, including the emotion domain. This suggests there was poor agreement between individuals and staff, but good agreement between individuals and family proxies. Although, much like Schmidt et al., (2010), overall proxy observation ratings were lower than self-report ratings. There were also significant differences found on the three domains of interpersonal relations, social inclusion and physical wellbeing. Again, however, this study did not include those with a more severe or profound ID. A further limitation of this study is the authors did not include information on whether any ethical approval was sought.

Simoes and Santos (2016a) examined the consistency between QOL self-reports and staff as well as family members. Overall, they found a moderate to excellent agreement between self-report and proxies, and again, good agreement on the emotion domain. Physical wellbeing domain had a poor agreement, which is in line with the results of the Schmidt et al., (2010) and Claes and Vandevelde et al., (2012) studies. Additionally, It was found that individuals reported their QOL as higher than their proxies, and agreement was better with care staff than it was with family members. They concluded that different perspectives should be taken into account when measuring QOL in those with ID. However, results of the study should be interpreted with caution as the sample size was small as well as having a lack of male family members and support staff in the sample.

Balboni, Coscarelli, Giunti, and Schalock (2013) investigated QOL self-reports and compared them to proxies' views of their QOL, and is the only study that also looks at the proxies' estimations of their clients' point of view. Unlike the above studies, they found a fair to poor correlation between self-report and proxy reports, but good to excellent correlation was found between self-report and the carers' estimation of the client's point of view for all levels of ID, with the exception of the emotional, interpersonal relationship and, again, physical wellbeing domains, for which the correlations were only fair. One of the strengths of this study is its inclusion of those with a more severe disability. This group of participants could not complete a self-report measure, although there was a strong correlation between both proxies' estimation of the client's point of view. Limitations of this study include the sample size not being justified and information on attrition or non-responders was not described.

Koch et al., (2015) investigated the self-report and proxy measure of QOL for adults with mild or moderate ID, as well as the impact of psychiatric symptoms, problem behaviour and medication. Overall, they found that agreement between self-report and the proxy respondents were poor, including the emotion domain, and that proxies tend to consistently underestimate the QOL subjectively experienced by people with ID, which is in line with Schmidt et al., (2010) and Claes and Vandevelde et al., (2012) findings. They also reported that unmet needs and medication were the most important predictors of reduced self-rated QOL. The main limitation of this study is the omission of family carers as proxies and also there were no participants with severe and profound ID.

Simoes and Santos (2016b) examined the differences between self-report and proxy measures of QOL, together with personal and environmental characteristics influencing QOL. They found good to excellent correlations between self and proxy reports, with good agreement on the emotion domain. Factors of diagnosis, age, living circumstances and daily living activity strongly affecting QOL for those with ID. Limitations of this study are that the participants were not randomly selected and only persons with a mild or moderate ID took part. There was also a lack of participants over the age of 50 years.

To summarise, four of these studies showed an overall good to excellent agreement between self-report and proxy reports on QOL, with good agreement on the emotion domain. Two of the studies show poor agreement overall. However, Balboni et al., (2013) also measured proxies' estimation of the client's point of view and found good to excellent correlation between this and the client's self-report overall, although there was still poor agreement on the emotion domain. Common themes across these studies are that proxies tend to consistently underestimate QOL when compared to the person's self-report. Additionally, in four out of the six studies, physical wellbeing seems to be the most problematic domain to find agreement between self-reports and proxies.

Variables or predictors of QOL

Ten studies used quantitative research methods to investigate variables that affect or predict QOL for people with ID. An overall total of 4451 participants with ID, 55% men and 45% women, participated in the studies. Participants' ages ranged from 16 to 83 years. All the studies used a range of psychometric measures, observational methods or semi structured interviews to assess a range of variables. Three of the papers had those with severe or profound ID as participants.

The first study by Simoes and Santos (2016c) essentially compared the QOL of those with and without ID and any fundamental predictors of QOL. They found QOL was significantly higher for those without an ID and that health status was the highest predictor of QOL for both those with and without ID. They also found that people living in their family home were more likely to have a higher QOL than those living in residential homes, and those in paid employment were more likely to have a higher QOL. A limitation of this study is its lack of participants with severe or profound ID, and participants were not randomly selected. It should also be noted that it is connected to another paper in this review (Simoes & Santos, 2016b).

Memisevic, Hadzic, Zecic and Mujkanovic (2016) investigated employment status, as well as other demographic variables such as: age, sex, level of ID, and marital status, to find any potential predictors of QOL for people with mild or moderate ID. They found that the level of ID and employment status were significant predictors of QOL. Specifically, they reported that having a mild ID meant having a better QOL compared to having a moderate ID. They concluded that people with ID should be supported to find suitable employment. A limitation of this study is its non-random sample of participants and its lack of residential status being taken into account.

Beyer, Brown, Akandi and Rapley (2010) looked at the variable of employment in more detail and compared QOL for people with ID in those in either supported employment, employment enterprises and day services. They also compared QOL with those not in employment, as well as people without an ID. They found that QOL was higher for those without an ID in employment. For those with ID, they found that those in supported employment reported higher QOL than those in employment enterprises and day services, particularly in the domains of health, productivity and emotional wellbeing. A limitation of this study is that the authors did not include information on the residence of the participants with ID, or the level of ID. The authors did not describe non-responder or information on attrition. These results agree with Memisevic et al., (2016) that those with ID in employment tend to have a higher QOL, but adds that being in *supported* employment is the optimal form of employment.

Claes, Hove, Vandevelde, Loon and Schalock (2012) investigated how support strategies, environmental factors and client characteristics impact on QOL for those with mild, moderate, severe and profound ID. Similar to the findings of Beyer et al., (2010) and Memisevic et al., (2016), they found that QOL was significantly affected by the status of employment, with employment significantly correlated with higher QOL scores. They reported that QOL outcomes were significantly impacted by level of ID, with those with mild ID experiencing better QOL than those with more severe ID. They also reported that QOL was also affected by support strategies and living arrangements. Having natural support (from family and friends) was reported to have a significant impact on individual QOL, and they concluded that those with a more severe ID often have to rely on more professional support, and this is counterproductive to the establishment and maintenance of close emotional relationships and natural support, which are important for a good QOL The main limitation of this study is that all the data was collected from one organisation which is fully committed to improving QOL and societal inclusion for users of their service. This may mean that the sample is not representative of the wider population and that the results are skewed.

Bertelli, Bianco, Rossi, Scuticchio and Brown (2011) investigated the relationship between individual and family QOL. They found that only some aspects of individual QOL (being, belonging and becoming) had statistical correlations with family QOL, but could not explain the nature of this relationship, suggesting this deserved further research. A strength of this study was its inclusion of people without expressive language, although they did not expressly use the term 'severe or profound' the suggestion is that the authors did include this group. Further delineation of ID level would have been useful. Further limitations are that the sample size was small and that family participants were all over the age of 64 years, which may have biased the results.

Van Asselt-Goverts, Embregts and Hendriks (2015) investigated which social network characteristics related to self-reported QOL for those with ID. They found that the majority of participants were satisfied with their social networks, with affection towards family and professionals being strongly related to QOL. Again, a limitation of this study is the lack of severe or profound participants. It would also have been useful if the authors compared characteristics of individual's social networks.

Similarly, Emerson and Hatton (2008) explored factors associated with the QOL domains of wellbeing and sociability. They found an association between economic disadvantage, contact with friends with ID and the subjective wellbeing component of QOL. Participants in this study who had more contact with friends who also had an ID, were more likely to have a higher QOL. Additionally, they also unexpectedly found wellbeing was strongly influenced by gender, with women having greater wellbeing on all indicators when their relationship status was single. The main limitation of this study was its lack of reporting on participants' level of ID. This study, along with Bertelli et al., (2011) and Van Asselt-Goverts et al., (2015) suggests that, perhaps unsurprisingly, having close personal relationships is an important aspect of having a good QOL.

Focussing on factors of cognitive functioning, Sharfi and Rosenblum (2016) investigated executive function and organisation in time, and their role in QOL in those with and without ID. They found that those with ID reported lower QOL and had significantly poorer executive function, organisation in time abilities, and more negative emotional responses. These were significant predictors of psychological QOL. The authors did not specify the level of ID in this study and participant gender was significantly biased towards women, which does not represent the wider population and is a limitation.

Investigating more psychological factors, Rey, Extremera, Duran and Ortiz-Tallo (2013) investigated the role of emotional competence on QOL for those with ID. They found that emotional competence had a positive association with QOL and happiness. They conclude that supporting and teaching those with ID how to manage negative emotions should be an integral part of their care plan to improve QOL. This study is limited by its small sample size, and it does also not report on the participant levels of ID.

Lombardi, Croce, Claes, Vandevelde and Schalock (2016) investigated the sociodemographic, clinical and functional factors predicting QOL for people with ID. They found QOL outcomes were significantly impacted by support needs, client characteristics, personal goals and personal desires. They reported that those with mild ID experienced better QOL than those with more severe ID. They concluded that QOL in those with ID could be improved by providing support activities aligned to the specific personal needs and goals of the individual. This study had a particularly robust design and benefited from the inclusion of those with severe ID, as well as those with mild and moderate ID. However, case managers selected participants for inclusion which is a limiting factor for external validity.

The variables or predictors investigated in the above studies are varied. However, there are themes around employment and social or family supports that have emerged. Three of the studies directly compared those without ID with those with ID and all found those with an ID had a lower QOL. The theme around the importance of social and family network support having an impact on the QOL of those with ID is interesting to consider for those with more severe or profound ID who perhaps rely on professional services more than those with mild or moderate ID.

Discussion

This review aimed to: synthesise the literature investigating the QOL of people with ID; identify any difference in QOL; examine areas of impact; examine the inter-rater reliability of self-report and proxy reports of QOL, particularly in regard to the emotion domain; and identify whether the available literature includes those with a severe or profound ID. Results from the studies included in this review followed some main themes (i) Those with ID may have a lower QOL than those without an ID; (ii) Employment seems to predict a higher QOL score; (iii) Social and family networks have an impact on QOL; and (iv) There is fair agreement between self-report and proxy reports of QOL for the emotion domain, but poor agreement for the physical domain.

Summary of Findings

Difference in QOL . Of the four studies which directly compared the QOL of those with ID and those without an ID, three found those with an ID had a lower QOL than those without, and one study showed the opposite result. This seems to reflect other research which shows a mix of results, but mostly shows lower QOL for those with ID (McVilly, Burton Smith & Davidson, 2000; Sands & Kozleski, 1994; Watson & Keith, 2002; Pretty et al, 2002). Continued research, it has been said, into the comparison between those with and without ID, is important to establish social equity and to identify lifestyle characteristics that impact upon it (Schalock et al. 2002).

Areas of impact

The variables or predictors investigated are varied. However, there are themes around employment and social or family supports that have emerged.

Employment. The four studies in this review that looked at the relationship of employment with QOL all reported that QOL ratings were higher for those in employment (Simoes, et. al, 2016c; Memisevic, et. al, 2016; Beyer, et. al, 2010; Claes and Hove et al.,2012). This is in line with other research that has reported that employment directly predicts higher QOL in those with ID (Gardner & Carran, 2005; Keith & Bonham, 2005). Individuals with ID and their families have also reported employment as being desirable. In a study conducted by Migliore, Mank, Grossi and Rogan (2007), individuals were asked whether they preferred employment within the community or day centre workshops, with 74% of those with ID preferring paid employment that is community-integrated, and 67% of families preferring the same, asserting that paid employment provides opportunities for skill development and community involvement in an inclusive setting (Dague, 2012; Donelly, Hillman, Stancliffe, Knox, Whitaker & Parmenter, 2010; Timmons, Hall, Bose, Wolfe & Winsor, 2011). It should be noted, that three of the four studies did not include those with severe or profound ID. One did include these individuals (Claes & Hove et al., 2012). However, these participants were not included in the analysis on employment. It would be difficult for those with severe or profound ID to find employment, which, according to these results, might be a barrier to good QOL.

Social and family networks. One of the studies included reported that living in the family home was related to a higher QOL (Simoes et al., 2016c) despite having no effect on QOL in the general population. Another reported that having more natural support (from family and friends) had a positive effect on QOL (Claes & Hove et al., 2012). Another study reported that affection directed towards family and professionals was strongly related to higher QOL (Van Asset-Govert et al., 2015), with another reporting that regular contact with other friends who also have ID was related to higher QOL (Emerson & Hatton, 2008). These findings build upon previous research that have highlighted the importance of social support for good QOL in a general population (Helgeson, 2003), and the QOL benefits of friendships between people with ID (Bayley, 1997; Emerson & McVilly, 2004; Robertson et al., 2001). This theme is interesting to consider for those with more severe or profound ID as Claes and Hove et al., (2012) highlight the fact that this group are likely to rely on professional services more than those with mild or moderate ID. This could impede their important close relationships, thereby impacting on their QOL.

Self-report and proxy views of QOL

The studies in this review largely show fair agreement between self-report and proxy reports of QOL, except on the physical domain. Of the six studies that looked at inter-rater reliability, four showed good agreement between individuals and their proxies on the emotion domain. This is in contrast to other research which has reported less agreement in psychological domains and more agreement in more observable aspects of QOL, such as environmental aspects (Zimmermann & Endermann, 2008).

Previous research has shown a mixed picture of the inter-rater reliability between self-report and proxy reports (McVilly, Burton Smith, & Davidson, 2000). In this review, proxies tend to consistently underestimate QOL when compared to the person’s self-report. This is compatible with other research in this field (Janssen, Schuengel & Stolk, 2005) and varied explanations for this have been presented in the literature. Some have suggested that those with ID are less critical of their environmental circumstances (McVilly et al., 2000), and have a positive outlook (Hartley & Maclean, 2006). Some have suggested that those with ID compare their situation to that of other people with ID (Stancliffe, 1999, p.192), while proxies may underestimate their QOL as they are comparing the individual to those without an ID (Arlt, Hornung, Eichenlaub, Jahn, Bullinger & Peterson, 2008).

In four out of the six studies, physical wellbeing seems to be the most problematic domain with individuals and proxies having poor agreement. Cooper, Smiley, Morrison, Williamson and Allan (2007) reported that those with ID have higher rates of physical illness, being 2.5 times more likely to have health problems than those without ID (Lantman-De Valk, Metsemakers, Haveman & Crebolder, 2000), and they report more barriers in health service access, so physical wellbeing is undoubtedly an important issue in QOL ID research. For other groups, such as for those with physical disabilities, concordance of self-report and proxy reports in the physical domain has been shown to be high (Schmidt, Petersen & Muhlan, 2006). Schmidt et al., (2010) suggests that perhaps for persons with ID, some physical aspects elicit different concepts, which may explain the lack of agreement in this domain between individuals and proxies. Further research is needed to properly explore this issue.

Severe and profound inclusion. One of the aims of this review was to identify whether the current literature included those with a severe and profound ID. Of the sixteen studies in the review, four included those with a severe and profound ID (Balboni et al., 2013; Bertelli et al., 2011; Lombardi et al., 2016; and Claes and Hove et al., 2013), with the rest including only those with a mild or moderate ID. Lombardi et al., (2016) and Claes and Hove et al., (2012) both reported that those with mild ID experience better QOL than those with more severe ID, and Balboni et al., (2013) found that there was good agreement between two proxies' estimations of the individual's point of view on their QOL, suggesting that this is a valid way to measure QOL for those who are unable to self-report. Bertelli et al., (2011) did not utilise level of ID as a variable. As few of the studies reported on severe and profound ID, it is not possible to offer any conclusions about the QOL of those with severe or profound ID. This supports Lyons' (2005) assertion of there being a lack of research into the QOL of these individuals.

Limitations

The studies in this review used a cross-sectional design, which means changes across time were not measured, and so causality cannot be determined. The quality appraisal scores of the papers were varied, and as such, findings from the higher rated studies may have more credibility than the findings from the lower rated studies. However, the studies used in this review were scored as having moderate to high quality, and the difference in these scores may reflect the diversity of the methodologies used. The variety of outcome measures used also hinders direct comparison, and these measures were not assessed in this review for validity or quality.

The studies used had a wide variation in sample sizes, ranging from 17 (Beyer et al., 2010) to 1, 285 (Lombardi et al., 2016), which reflects the diverse research methodology. However, it was difficult to assess the possibility of selection bias as none of the studies adequately described the source population. The demographics were also characterised by a majority male sample (56.5%), and a lack of ethnic diversity, despite recruitment being across many developed countries. The variety of countries used for recruitment between the papers, may have affected outcomes due to heterogeneity of cultural norms. For example, previous research has shown that emotions are a superior predictor of life satisfaction in individualistic nations, whereas social norms are an equally strong predictor of life satisfaction in collectivist cultures (Eunkook, Diener, Oishi, Traiandis, 1998), which suggests QOL may have different meanings in different cultures. Therefore, the generalisability of finding to wider populations is limited.

Finally, it is possible that some research has not been accounted for in this review, as prominent journals were not hand searched, the grey literature was not consulted, and only papers written in English were considered.

Clinical implications

Clinicians should be aware that those with an ID are possibly more likely to have a lower QOL, and so assessment of QOL is important to measure, to enable planning and to implement personalised care packages and interventions. Clinicians should consider routine use of QOL outcome measures when working with those with ID, using self-report measures where possible, and complimenting these with proxy measures. Different perspectives are useful when measuring QOL in those with ID, but clinicians should be aware of the potential for proxies to underestimate QOL when compared to the person's self-report. The studies in this review have also highlighted the positive impact of social and family relationships and employment, on the QOL of those with ID. Therefore, clinicians should consider the importance and potential role of an individual's social and family support network and their employment opportunities into care planning and service provision.

Recommendations for future research

Future research should continue to examine the inter-rater reliability of QOL for those with ID. It would be particularly helpful to explore potential explanations as to why the physical domain has the poorest agreement between self and proxy reports, while emotion domains largely find good agreement. Research of QOL in those with ID might further benefit from building on Balboni et al's., (2013) research by investigating proxy reports of the client's estimation of their QOL, as an alternative to proxy point of view of the person's QOL. Inter-rater reliability may be improved by measuring QOL in this way, as well as this being a potentially valid and useful way of assessing QOL in those who are not able to self-report.

This review has added to the notion that there is a lack of research into the QOL of those with severe or profound ID and future research needs to address this. It would be particularly helpful to explore the idea of social and family relationships being important to a person's QOL, given the findings that those with severe or profound ID have limited networks which often include a majority of professionals (Campo, Sharpton, Thompson, and Sexton, 1997). The impact of this on individual's QOL should be explored.

Conclusion

The findings of this review bring together the limited literature that is available on the QOL of people with ID. The results contribute to the literature base and suggests that those with ID may have a lower QOL than those without an ID. This review also offers some support to the notion that proxy and self-rated reports of QOL show, overall, fair inter-rater reliability, particularly in relation to emotions. However, It should be noted that this could not be confirmed for people with severe or profound ID as they are unable to self-report. However, the results do suggest that proxy reports may give a fairly accurate estimation of QOL for the emotion domain for these individuals, when self-report is not possible. This review has highlighted physical domains in QOL measures as particularly problematic, finding poor agreement between self and proxy reports. Therefore, further research is needed. Clinicians are advised that consideration should be given to an individual's social and family support networks and potential employment opportunities when planning service provision. They are also advised to include both self-report and proxy QOL measures where possible, but consider that it is possible that proxy reporters tend to underestimate QOL for those they care for when it relates to the physical domain. Finally, few of the studies included in this review reported on severe and profound ID. This supports Lyon's (2005) assertion of there being a lack of research for these individuals. As there is so little research for this group it is not clear whether they experience a good or poor level of QOL. Further research in to the QOL of those with severe or profound ID is needed to see what, if any, elements of QOL could be improved upon in health care settings for this group.

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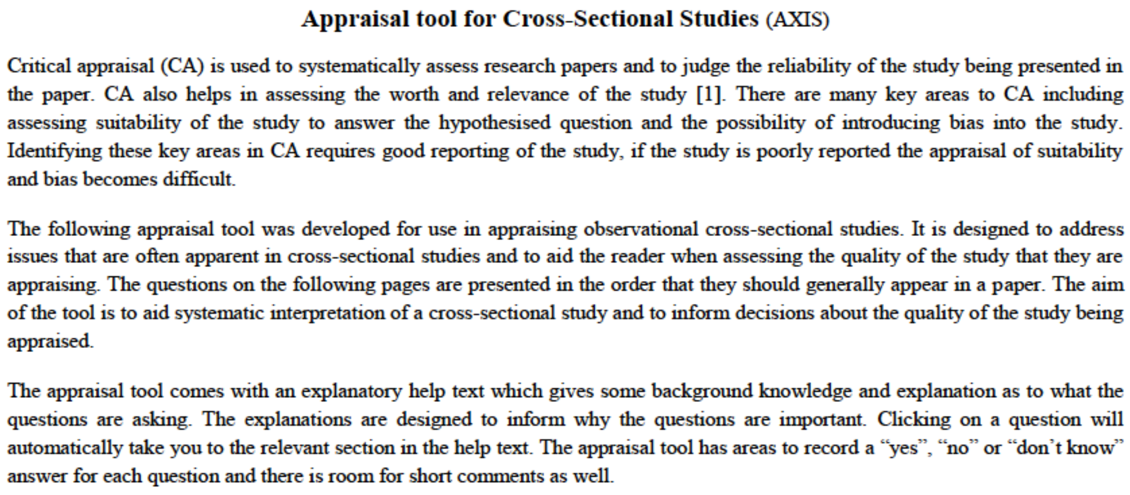
*Denotes key papers in review

Appendix 1. Appraisal tool for Cross-sectional studies (AXIS)



Appraisal tool for Cross-Sectional Studies (AXIS)

Critical appraisal (CA) is used to systematically assess research papers and to judge the reliability of the study being presented in the paper. CA also helps in assessing the worth and relevance of the study [1]. There are many key areas to CA including assessing suitability of the study to answer the hypothesised question and the possibility of introducing bias into the study. Identifying these key areas in CA requires good reporting of the study, if the study is poorly reported the appraisal of suitability and bias becomes difficult.

The following appraisal tool was developed for use in appraising observational cross-sectional studies. It is designed to address issues that are often apparent in cross-sectional studies and to aid the reader when assessing the quality of the study that they are appraising. The questions on the following pages are presented in the order that they should generally appear in a paper. The aim of the tool is to aid systematic interpretation of a cross-sectional study and to inform decisions about the quality of the study being appraised.

The appraisal tool comes with an explanatory help text which gives some background knowledge and explanation as to what the questions are asking. The explanations are designed to inform why the questions are important. Clicking on a question will automatically take you to the relevant section in the help text. The appraisal tool has areas to record a "yes", "no" or "don't know" answer for each question and there is room for short comments as well.

Appraisal of Cross-sectional Studies

	Question	**Yes**	**No**	**Don't know/ Comment**
Introduction				
1	Were the aims/objectives of the study clear?			
Methods				
2	Was the study design appropriate for the stated aim(s)?			
3	Was the sample size justified?			
4	Was the target/reference population clearly defined? (Is it clear who the research was about?)			
5	Was the sample frame taken from an appropriate population base so that it closely represented the target/reference population under investigation?			
6	Was the selection process likely to select subjects/participants that were representative of the target/reference population under investigation?			
7	Were measures undertaken to address and categorise non-responders?			
8	Were the risk factor and outcome variables measured appropriate to the aims of the study?			
9	Were the risk factor and outcome variables measured correctly using instruments/measurements that had been trialled, piloted or published previously?			
10	Is it clear what was used to determined statistical significance and/or precision estimates? (e.g. p-values, confidence intervals)			
11	Were the methods (including statistical methods) sufficiently described to enable them to be repeated?			
Results				
12	Were the basic data adequately described?			
13	Does the response rate raise concerns about non-response bias?			
14	If appropriate, was information about non-responders described?			
15	Were the results internally consistent?			
16	Were the results presented for all the analyses described in the methods?			
Discussion				
17	Were the authors' discussions and conclusions justified by the results?			
18	Were the limitations of the study discussed?			
Other				
19	Were there any funding sources or conflicts of interest that may affect the authors' interpretation of the results?			
20	Was ethical approval or consent of participants attained?			

Introduction

The introduction serves to establish the context of the work that is about to be presented in the text of the paper. Relevant primary literature should be discussed and referenced throughout the introduction. The history and current understanding of the problem being researched should be presented. This should be concluded giving a rational as to why the current study is being presented and what the aims and/or hypothesis under investigated are [2,3].

Aims

The aim(s) of the study tells us if the study addresses an appropriate and clearly focused question. If the aim is not clearly stated or not stated at all, it will be difficult and in some cases impossible to assess the extent to which the study objectives were achieved. Ideally, an aim should be stated both at the beginning of the abstract and at the end of the introduction [3]. If the answer to question 1 is no, then it will make it difficult to assess some of the other questions in the critical appraisal process.

Methods

The methods section is used to present the experimental study design of the paper. The methods should be described clearly in easy to understand language and clearly identify measures, exposures and outcomes being used in the study [4]. More specific issues are addressed below.

Study Design

Question 2 is used to assess the appropriateness of using a cross-sectional study to achieve the aim(s) of the study. Cross-sectional studies are observational studies that provide a description of a population at a given time, and are useful in assessing prevalence and for testing for associations and differences between groups [5]. Examples of cross-sectional designs include point-in-time surveys, analysis of records and audits of practice [6]. The reader should try and decipher if a cross-sectional study design is appropriate for the questions being asked by the researcher.

Sample Size Justification

Sample size justification is crucial as sample size profoundly affects the significance of the outcomes of the study. If the sample size is too small then the conclusions drawn from the study will be under powered and may be inaccurate. This can occur by failing to detect an effect which truly exists (type II error) sometimes referred to as a "false negative". The probability of a type I error is also taken into account when determining sample size. A type I error is drawing significant conclusions when no real difference exists and is a function of the p-value (see Statistics section below) sometimes referred to as a "false positive".

Question 3 asks if sample size justification was reported, but it should also be clear what methods were used to determine the sample size. In some cases clustering of observations within groups can occur (e.g. patients within hospitals or livestock within herds) and this should be taken into account if sample size has been determined. It should be clear whether the inferences drawn actually relate to the attributes for which the sample size was calculated [7]. If sample size justification isn't given or restrictions make it difficult to reach the desired sample size then this should be declared in the text.

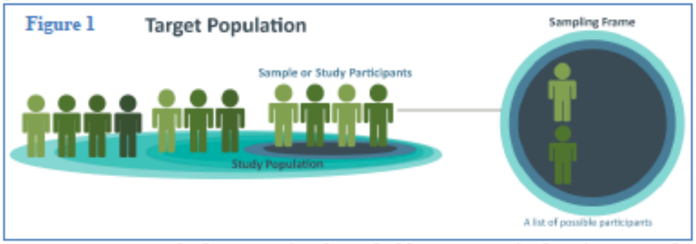
Target (Reference) Population

The target or reference population is the overall population that the research is directed towards. When doing a cross-sectional study, a target population is the overall population you are undertaking the study to make conclusions about or the population at risk of acquiring the condition being investigated [8–10] e.g. the total female population in the UK, or all dogs in the USA with cardiovascular disease. (See Figure 1) Question 4 asks if this is clearly defined in the study. It is important that this is understood both by the researcher and the reader; if it is not clearly defined then inferences made by the researcher may be inappropriate.

Sampling Frame

As a reader you need to determine if the sample frame being used is representative of the target population. The study population should be taken from the target population; units from this study population have information that is accessible and available which allows them to be placed in the study. The sampling frame is the list or source of the study population that the researcher has used when trying to recruit participants into the study (Figure 1). Ideally it should be exactly the same composition or structure as the target population. In practice it is generally much smaller, but should still be representative of the target population. Generally, for convenience, the sampling frame is a list of units that are within the target population e.g. list of telephone owning households, computerised patient records etc. A sample of units is selected from the study population to take part in the study and is generally only a small proportion of the study population (see Sample Selection below) - this proportion ratio is known as the sampling fraction. It is very important that the sampling frame is representative of the target population as results from the study are going to be used to make assumptions about the target population [8–10].

Figure 1



Convenience sampling can be carried out in some situations and are used because the participants are easy to recruit. Convenience samples generally lead to non-representative or biased samples and therefore cannot be used to make assumptions about the characteristics of the target population [11]. Convenience samples are often used for pilot or analytical studies where the need for a representative sample is not required [12], however the authors should make this clear in the text.

Census

A census is where the target population and the study participants are the same at the time the census is taken. In theory questions 5, 6 and 7 don't apply to census studies. However even if a study is described as a census it should be very clearly stated where the study participants have been recruited from, and the reader should make the decision if the study truly is a census. A census may include all the population from the sample frame, but not all the target population; in this scenario questions 5 to 7 need to be addressed.

Sample Selection

Question 6 is used to establish how the researchers got from the sample frame to the participants in the study. It examines the potential for selection bias and how the researcher developed methods to deal with this. The sample selection process is important in determining to what extent the results of the study are generalizable to the target population. For question 6 we are looking in depth at how the sample (study participants) was selected from the sampling frame. It is important to know if there were any inclusion or exclusion criteria used, as inappropriate criteria can dramatically shift how representative the sample is of the target population [8,10,13].

Selection bias can occur if every unit in the sample frame doesn't have an equal chance of been included in the final study [11,14]. Randomisation is used to ensure that each participant in the sampling frame has an equal chance of being included in the sample. If methods of randomisation are not used, not described or are not truly random, this may lead to a non-representative sample being selected and hence affect the results of the study [10,11].

There are many other situational issues to take into account when determining if the population in the sample is likely to represent the target population. Often these issues are outside the control of the researcher, but sometimes are overlooked. One such issue is the healthy worker effect which is a well-known phenomenon in human cross-sectional studies [13]. An example of this is, a researcher trying to do a cross-sectional study to determine health factors in a factory population and decides to sample from workers at work on a particular day. Unfortunately there is a tendency to over select healthy workers as ill workers may tend to be at home on the day of selection. This will in turn lead to inferences been made about the health of the worker population but is only relevant to healthy workers and not ill workers. A veterinary example of this is a researcher trying to do a cross-sectional study to determine health factors in the general dog population and decides to sample from a local park. Unfortunately there is a tendency to over select healthy animals as sick animals will tend to be left at home and not taken for a walk. This will in turn lead to inference been made about the health of the dog population but is only relevant to healthy dogs and not sick dogs.

Self-selection is another example of selection bias that can be introduced and should be assessed [13]. For example, when using a postal questionnaire to examine eating habits and weight control, people who are overweight might read the survey and be less inclined to complete and return the survey than those with normal weight leading to over representation of people with normal weight. Similarly, if using a postal questionnaire to examine mastitis levels on cattle farms, farmers that have a high somatic cell counts (SCC) might be less inclined to complete the survey than those with normal or low SCC leading to over representation of farms with good SCC (see Non-responders below).

Non-responders

Non-response in cross-sectional studies is a difficult area to address. A non-responder is someone who does not respond either because they refuse to, cannot be contacted, or because their details cannot be documented. As a rule, if participants don't respond it is often difficult and sometimes impossible to gain any information about them. However other baseline statistics may exist that can be used as a comparator to assess how representative the sample is [14] e.g. age, sex, socio-economic classification. Methods used, if any, should be well described so that the results from the analyses can be interpreted. This is important as non-responders may be from a specific group, which can lead to a shift in the baseline data away from that group. This shift can lead to results that don't represent the target population. In some situations the sampling frame doesn't have a finite list or a fully defined baseline population. This also makes it difficult, and in some cases impossible, to quantify non-response and it may be inappropriate to do so in these situations. If the researchers are using non-defined populations this should also be declared clearly in the materials and methods section [15,16].

Measurement Validity & Reliability

Measurement validity is a gauge of how accurately the study measurements used assess the concepts that the researcher is attempting to explore. Measurement reliability is a gauge of the accuracy of the measurements taken or the procedures used during the study. Question 8 is used to address the concepts of measurement validity, and is specifically aimed to address the appropriateness of the measurements being used.

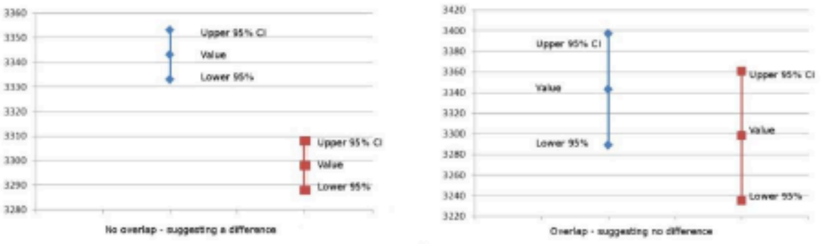
The importance of measurement validity is that it gives weight to applying the statistical inferences from the study to members of the target population. If inappropriate measures are used in the study it could lead to misclassification bias and it will be difficult to determine to what extent the study results are relevant to the target population [12,17].

Question 9 is an attempt to gauge the measurement reliability of the study measures. Measurements must be able to be reproduced and produce identical results if measured repeatedly, so that the measurements would be exactly the same if performed by another researcher. With this in mind, the measurements must be of international or globally accepted standards (e.g. IU standards) where possible and appropriate. If they are being used for the first time they must be trialled, or in the case of questionnaires, they should be piloted before being used.

Statistics

While interpretation of statistics can be quite difficult, a basic understanding of statistics can help you to assess the quality of the paper. Often many different methods can be used correctly to test the same data, but as there is such a wide range available, knowing what tests are most appropriate in particular situations can be hard to decipher. There is an expectation that the researcher has this understanding or has at least sought statistical assistance to ensure that the correct methods are used. Therefore for question 10 the emphasis for the reader is that the statistical methods, software packages used and the statistical significance levels are clearly stated even if the paper is just presenting descriptive statistics. The statistical significance level is usually described as a p-value. In most cases the p-value, at which the null hypothesis is rejected, is set at 0.05. The higher the p-value is set the greater the possibility of introducing a type I error. Confidence intervals should also be declared with p-values or instead of p-values as an indication of the precision of the estimates. It is usual to present a confidence interval of 95% which means that the researchers were 95 per cent confident that the true population value of the outcome lies between these intervals. This can be used to compare groups where an overlap would suggest no difference and a gap between confidence intervals would suggest a difference (Figure 2).

Figure 2



Overall Methods

Question 11 asks if the methods are sufficiently described to enable them to be repeated. If there are sections or even small pieces of information missing it could make a great difference for the reader when interpreting the results and the discussion as they may be unsure if the correct methods are being used.

Results

The results section of a paper is solely for the purpose of declaring the results of the data analysis and no opinion should be stated in this section. This gives the reader the opportunity to examine the results unhindered by the opinion of the researcher. It is important for the reader to form their own ideas or opinions about the results before progressing to the discussion stages.

Basic Data

Question 12 asks for a description of the basic data. Basic descriptive analysis aims to summarise the data, giving detailed information about the sample and the measurements taken in the study. The basic data gives an overview of the process of recruitment and if the sampling methods used to recruit individuals were successful in selecting a representative sample of the target population. If the sampling methods are unsuccessful in selecting a representative sample of the target population, those participants included in the study can often be different to the target population; this leads to inaccurate estimates of prevalence, incidence or risk factors for disease. Descriptive data of the measurements taken in the study give an overview of any differences between the groups, and may give insight into some of the reasons for statistical inferences that are made later in the paper.

Response Rate

As stated previously it can often be difficult to deal with non-responders. Question 13 requires that there is some attempt made to quantify the level of non-response by the researchers and asks the reader to interpret if the response rate is likely to lead to non-response bias. Question 14 is examining if any information on non-responders was available and if so were they comparable to those that did respond as this could help in answering question 13. Non-response bias occurs if the non-responders are substantially different to the rest of the population in the sample [15].

Internally Consistent Results

Question 15 is an exploration of the basic data and asks that the reader spends some time exploring the numbers given in the results; in the text, figures and tables. Information about the level of missing data should also be declared in the results. It is important to check that the numbers add up in the tables and the text. If the study has recruited 100 participants, the tables and the text should include data about 100 participants. If not, the missing data should be clearly declared and the reason for its non-appearance explained.

Comprehensive Description of Results

It is important to check that all the methods described previously lead to data in the results section (question 16). Sometimes the results from all analyses are not described. If this is noted it will be unclear whether the researcher found non-significant results or just didn't describe what was found. If there are results missing that you would expect to find, there is a concern that these missing results may not have been what the researcher wanted to see and hence the authors have omitted them. It is also important that the significance level declared in the methods is adhered to. As the reader, it is important to watch out for phrases such as "tended towards significance" in the text, and if these are used to pay close attention to the results.

Discussion

The discussion of a paper should summarise key results of the study objectives. It should give an overall interpretation of the results of the study keeping in mind the limitations and the external validity of the document. The discussion section should also address both significant and non-significant findings of the study and make comparisons with other research, citing their sources [2,4].

Justified Discussions and Conclusions

In question 17 there is an expectation that the researcher gives an overall summary of the main findings of the study and discusses these in detail. It is important that the reader considers the study as a whole when reading the researcher's conclusion. If the researcher's conclusion is different or is more definitive than the study suggests it should be, it can be an indication that the researcher has misunderstood their own study or has other motives or interests for coming to that conclusion.

It is up to the reader to explore the discussion fully in order to answer question 17. The following points should be taken into account:

Aim

In the discussion section the researcher should discuss all results that pertain to the overall aim of the study, even if they are not significant. If some results are overlooked in the discussion it could suggest that the researcher either doesn't believe the results, or doesn't want to draw attention to controversial discoveries from the study and may therefore be giving a biased overview of the research conducted.

Selection Bias

There is an expectation that the researcher discusses selection biases and takes these into account when interpreting the results of the study. This also gives a clear view of whether the researcher has an overall understanding of the study design. (See notes on selection bias in the methods section).

Non-response

Was there an interpretation of the results that included non-response? This is particularly important if the response rate was low, as non-responders may be a specific group, and lead to a shift in the baseline data (See notes on non-response in the methods section).

Confounding

Confounding is a major threat to the validity of practical inferences made from statistical analyses about cause and effect. Confounding occurs when the outcome of interest is associated with two different independent variables and one of those variables is closely associated with the outcome only because it is closely associated with the other variable (confounder). This can sometimes be accounted for using statistical methods however sometimes these associations are missed because the confounder isn't measured or isn't considered to be a confounder in the analyses. What then happens is an erroneous conclusion is made; that the variable might have a causal relationship with the outcome. The researcher should consider confounding both in the analyses and in the interpretation of the results [18]. An example would be where in a study on cancer a researcher concludes that increased alcohol intake causes lung cancer; however there was confounding in the sample that the researcher didn't discover. People in the study that were inclined to drink more alcohol were also inclined to smoke more (the confounder) and smoking was the cause of lung cancer not increased alcohol intake. Similarly, a study was undertaken to examine surgical deaths in cats. The researcher concluded that cats that had gaseous anaesthesia were more likely to die during surgery than those that had just injectable anaesthesia. There was confounding in the sample: cats that underwent surgery using gaseous anaesthesia were more likely to be ill or undergoing major surgical procedures (the confounders) and this was the cause for cats being more likely to die during surgery and not the use of gaseous anaesthetics.

Non-significant Results

Discussing non-significant results is as important as discussing significant results and should also be included in the discussion, especially if they have a direct association with the aim being investigated. Non-significant results can be influenced by factors associated with study design and sample size. If there are biases introduced during the study design this can lead to non-significant results that in reality may be significant (this can work the other way around as well). If there are only small differences between groups, non-significant results may be apparent because the sample size is too small (see sample size justification). Again it is important that the researcher has a clear understanding of this and conveys that in the discussion.

Limitations

In question 18 we explore whether limitations are discussed. Unfortunately all forms of research have some limitations. The question here is whether the researcher has an understanding of the limitations involved in their study design. If this issue is not explored, this is cause for concern that the limitations don't stop at the design and that the researcher has a poor understanding of the study as a whole.

Other

Conflicts of Interest

It is very important that conflicts of interest or bodies involved in funding the study are declared in the text (question 19). This can give an impression as to background reasons for carrying out the study. Where studies are funded by a specific agency the researcher may unconsciously interpret in favour of the agencies' ideals; if the researcher has worked in a specific area their own ideas and beliefs may affect the interpretation of the results. It is up to the reader to identify these and come to the conclusion as to whether these conflicts of interest are relevant or not. This can be declared in different areas of the text and should be stated.

Ethical Approval

Question 20 deals with ethical approval and participant consent. It is important that these are sought before carrying out research on any animal or person.

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Section Two: Research Report

Exploratory study of carer perspectives of indicators of psychological distress and psychological wellbeing in people who have severe or profound intellectual disabilities: a qualitative study

Abstract

Objective: There is limited research into how those with severe or profound intellectual disabilities express their feelings of wellbeing or distress. The present study sought to explore carers' views on the emotions expressed by those with severe and profound intellectual disability.

Design: This study used a qualitative exploratory design.

Method: Semi-structured interviews were conducted with 18 participants. Participants were paired and included family members or paid carers of those with a severe or profound intellectual disability. The interview transcripts were analysed using template analysis.

Results: Analysis of transcripts yielded eight superordinate themes relating to: relationship; physical contact; body; vocalisation; things that affect their emotions; facial expression; unknown and telling their story. *Telling their story* emerged as a theme only related to family members.

Conclusions: Agreement between the dyads was fair. The theme of *unknown* is of particular interest and may be explained by unconscious communication. This study makes a contribution to a growing body of research which uses the expertise of family and other carers to understand and identify the inner emotional states of those who are not able to communicate their needs and wishes.

Practitioner points

Clinical implications:

- Those with severe or profound ID use their body, vocalisation, physical contact and facial expressions to communicate wellbeing or distress to their carers – with each having unique individual profiles.
- Medication side effects may impact on carers' ability to recognise emotions in those under their care.

- Consideration of unconscious communication may help to identify inner emotional states.

Limitations:

- The results were based on a sample that was not culturally diverse and also only included parents as the family participants.

Introduction

Intellectual disability (ID) is characterized by significant limitations in both intellectual functioning and in daily functioning, with these deficits originating before the age of 18 years. This study will focus upon individuals with severe or profound ID (IQ range <34; World Health Organisation, 1992).

Adults with Intellectual disability (ID) are at increased risk of mental health problems (Cooper, Smiley, Morrison, Williamson, & Allan, 2007), and there is some indication that those with severe and profound ID are at even greater risk (Cooper et al., 2007; Hove & Havik, 2010). Further to this, longitudinal studies suggest that while mental health problems for those with mild ID may decrease over time, for those with severe and profound ID they are more likely to persist (Einfield et al., 2006).

Mental health research in ID has mostly involved people with mild and moderate ID, largely ignoring people with severe and profound disabilities (Campo, Sharpton, Thompson, & Sexton, 1997). Little research has addressed the emotions experienced and expressed within this population, despite their perceived importance in other populations (Adams & Oliver, 2011; Arthur, 2003; Wagner, 1991).

Since the person with the disability is thought to be the best source of knowledge concerning their own emotional wellbeing, determining this often involves self-reports (Cummins, 1997; Schalock et al., 2002). However, using self-report measures is more difficult for people with severe or profound ID, due to their intellectual, sensory and motor impairments. Additionally, those with severe or profound ID would, according to the Mental Capacity Act (2005), be unable to provide informed consent to take part in research as they would not be able to directly communicate their own experiences with spoken language. This makes research in the emotions of those with severe and profound ID, difficult. As a result of these difficulties, there is a lack of explicit knowledge about the way individuals with profound disabilities express their feelings (Daelman, 2003; Roemer & Van Dam, 2004).

A systematic review (Hastings, Flynn, & Vereenooghe, 2017), reported there is also a paucity of tools measuring wellbeing for those with severe and profound ID. Psychological wellbeing is considered to comprise of three parts: evaluative wellbeing (life satisfaction); hedonic wellbeing (feelings of sadness and happiness); and eudemonic well-being (sense of purpose and meaning in life) (Kahneman, Diener, & Schwarz, 2003). The review highlighted there is only one measure that specifically aims to measure wellbeing for this group. The Physiological Measure of Subjective Well-Being (Vos, De Cock, Petry, Van Den Noortgate, & Maes, 2010) which is a physical measure of cardiovascular and skin responses that are associated with emotions. The above review noted this measure was not reported as being developed using any theoretical model basis, has only been evaluated once by the researcher that developed it, and there is no evidence of internal consistency and inter-rater reliability. The Mood, Interest and Pleasure Questionnaire (Ross & Oliver, 2010; MIPQ) was also assessed in the review. The MIPQ was developed using symptoms of mental health problems, as outlined in the Diagnostic and Statistical Manual of Mental Disorders (American Psychiatric Association, 2013; DSM), and was designed to track symptoms of distress.

A recent qualitative study by Griffiths and Smith (2016) observed carers and individuals with severe or profound ID to assess the communication process between them. Communication between those with severe or profound ID and their carers is not verbal and is often non-symbolic (McLean, Brady, McLean & Behrens, 1999). Non-symbolic communication include contact gestures, vocalisations, body movements, facial expressions (Stephenson & Dowrick, 2005; Grove, Bunning, Porter, & Olsson, 1999), eye gaze and body direction (Bradshaw, 2001). They found this group does have a functional ability to engage in meaningful communication with their carers, as they are empathically attuned to each other, and concluded more research is needed in to the ways in which people with complex disabilities communicate with their carers. Carer views on the emotions expressed and communicated by people with severe and profound disabilities are therefore a potential source of expertise that have not yet been explored.

The purpose of the present study is to explore carer and family perspectives of indicators of psychological wellbeing and distress in those who have severe or profound ID with limited means of expressive communication. The research questions are as follows:

- How do paid carers and family members know when the person they care for is experiencing or communicating psychological distress or wellbeing?
- Are there any common themes or differences between the responses of paid carers and family members?

It is hoped this will increase understanding, which may in turn inform new assessment tools and more effective interventions to improve well-being and reduce distress for this group.

Method

Design. The present study used a qualitative exploratory design. Template analysis (TA) was selected to analyse the data from semi-structured interviews. TA emphasises the use of hierarchical coding and balances a high degree of structure, with the flexibility to adapt it to the needs of a given study (Brooks, McCluskey, Turley, & King, 2015). A feature of TA is its dual deductive/inductive stance whereby a priori themes are developed, which allow the researcher to define themes ahead of the analysis stage. This enables the researcher to take a systematic and structured approach to data handling. TA is also particularly useful when perspectives are being investigated from different groups. In this case, the research aimed to compare perspectives of distress and wellbeing from paid carers as well as family members (King, 2004). Although TA does not come with a prescribed epistemology, it can be used with a range of positions (King, 2012). A critical realist position was taken, which acknowledges our understanding of material reality is mediated by cultural interpretation and assumes an 'inherent subjectivity in the production of knowledge' (Madill, Jordan, & Shirley, 2000; Sayer, 2000, p. 3).

Ethical considerations. Qualitative standards of best practice were used to guide all aspects of the research to ensure the study is valid, helpful and meets legal and ethical requirements of research (Thompson & Chambers, 2012). Ethical approval for this project was granted by the Department of Psychology Research Ethics Committee at the University of Sheffield (see appendix 1).

Confidentiality. All reasonable steps were taken to protect anonymity of third parties spoken about during interviews. Participants were asked to refrain from including any patient-identifiable information during the interview and asked to use pseudonyms at all times. Any information that could lead to patient identification was removed from the transcripts.

Recruitment. Participants were recruited through local authority day services and independent sector providers who support people who have severe and profound ID and comprised of paid staff carers and family members.

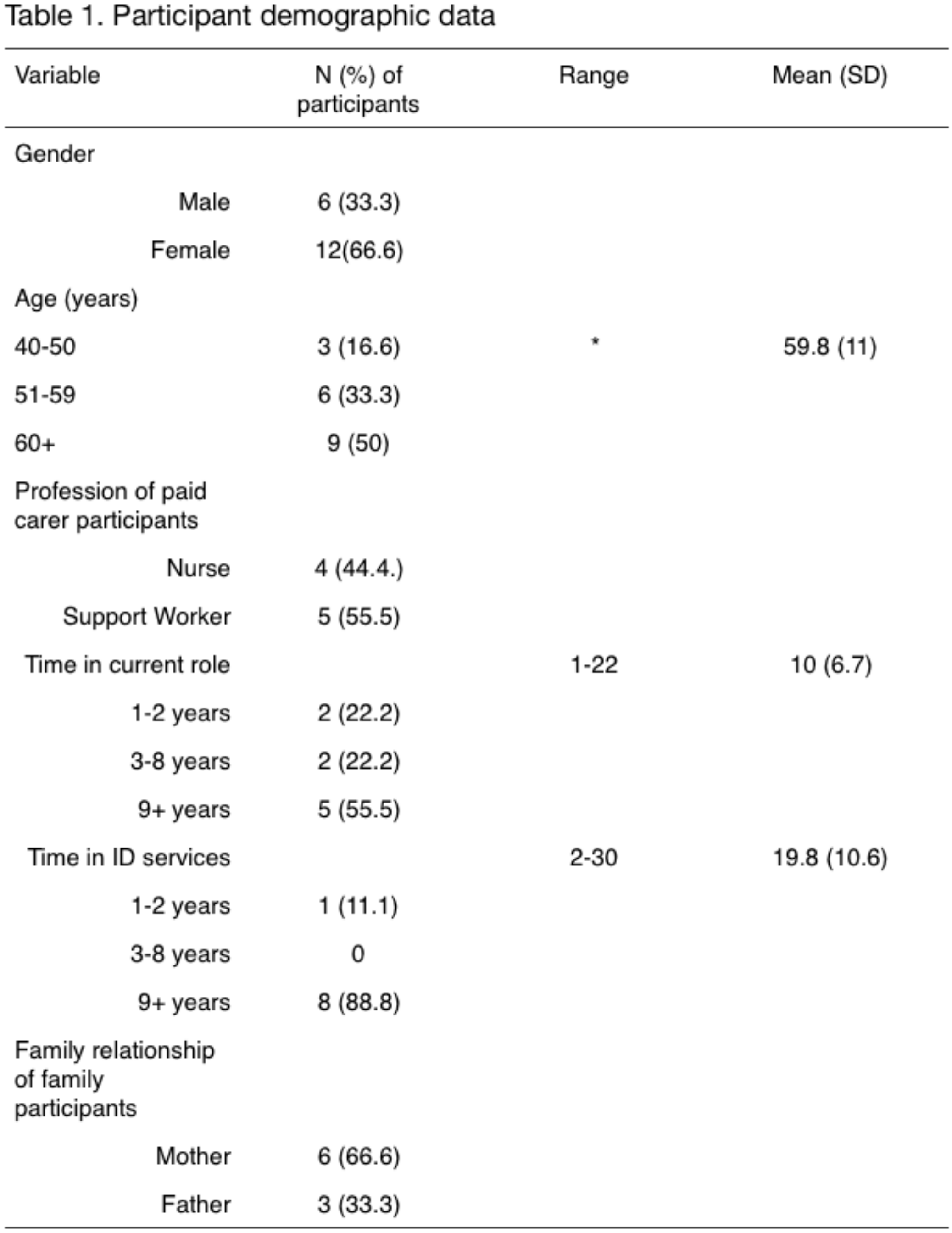
The lead researcher left some participant information sheets in the staff rooms and individuals were asked to contact the researcher should they wish to take part. Staff members who agreed to take part were asked to think of a person they care for with whom they have a good working relationship that they can discuss during the interview, and the service was asked to contact the person's family for permission to be contacted by the researcher. This was so the sample could be paired, meaning one paid carer and one family member were asked about the same service user.

Inclusion criteria. Participants were over the age of 18yrs. Paid carers were required to have worked in a learning disability service for a minimum of one year. Family members recruited were required to self-identify as the best person at being able to recognise the emotions of the person they care for.

Participants. In total, 18 participants were recruited and interviewed. This sample size is in line with other recent qualitative ID studies (Spassiani, Chacra, Sarah & Yona, 2017; Conder, Mirfin-Veitch, & Gates, 2015). Of these, 9 participants were paid carers and 9 were family members. Overall, 12 participants were female, and 6 were male. Their mean age was 59.8 (SD=11). Paid carers were either Nurses by profession (n=4) or Support Workers (n=5), and all family members were parents being either a Mother (n=6) or a Father (n=3) to the service user asked about at interview (see Table 1 for demographic data).

Informed consent. Participants were given a Participant Information Sheet (appendix 3), which contained information about the study and what taking part involved. Before proceeding with interviews, participants were given time to read through the Information sheet and the opportunity to ask questions, as in accordance with the BPS Code of Ethics and Conduct (2009). They were then asked to give written consent to participate in the study (appendix 4 for the consent form).

Table 1. Participant demographic data



Variable	N (%) of participants	Range	Mean (SD)
Gender			
Male	6 (33.3)		
Female	12(66.6)		
Age (years)			
40-50	3 (16.6)	*	59.8 (11)
51-59	6 (33.3)		
60+	9 (50)		
Profession of paid carer participants			
Nurse	4 (44.4.)		
Support Worker	5 (55.5)		
Time in current role		1-22	10 (6.7)
1-2 years	2 (22.2)		
3-8 years	2 (22.2)		
9+ years	5 (55.5)		
Time in ID services		2-30	19.8 (10.6)
1-2 years	1 (11.1)		
3-8 years	0		
9+ years	8 (88.8)		
Family relationship of family participants			
Mother	6 (66.6)		
Father	3 (33.3)		

Procedure. During the initial recruitment phase, three users of a learning disability day services centre, with a diagnosis of a mild or moderate ID, were consulted about the project before the main recruitment began, and any observations or comments they had were considered in the design of the project. A group meeting was held with six staff members of local authority day centre, to ask their perspective on the questions asked during the interview. This was used to further shape the interview schedule. Following this, a pilot interview was conducted with a member of the care staff to ensure familiarity of the researcher with the interview questions and to test the questions and structure.

Individuals who responded to invitations to take part were given the chance to ask questions before arranging the interview at their preferred location. Once an individual gave written consent, semi-structured interviews were conducted. Interviews were digitally recorded with participants' consent. Of the recorded interviews, 17 were transcribed verbatim by a third party, and 1 was transcribed by the researcher (see appendix 5 for interview schedule).

Data analysis

Analysis of transcripts were done in accordance with procedures outlined by King (2004). An initial template based on a priori themes was developed based on the MIPQ, discussions with clinicians and from consulting the wider literature. The MIPQ uses informants to score observable behaviours related to levels of mood, interest and pleasure and consists of 25 items with a five point Likert scale response format. It has good test-retest reliability coefficients (.87, total MIPQ score), good inter-rater reliability coefficients (.76, total MIPQ score).

This initial template had six higher-order themes of; body, physical contact, relationship, facial expression, and vocalisations. A conceptual map of priori themes can be found in appendix 6. The interviews were then read through twice, while listening to the recording. This is to ensure familiarity with the data. Initial coding of the data was then carried out on a sub set of four transcripts that the researcher felt were a good representation of the data. The initial template was applied to these transcripts and subsequently modified and adapted. This modified template was then applied to the rest of the transcripts and was again modified and adapted as necessary. No new codes emerged towards the end of the process and therefore saturation was assumed. The final template was reviewed and re-applied to the data set. All priori themes were retained and 3 new higher order themes emerged: things that affect their emotion; unknown; and telling their story.

The level of agreement between the paid carers and family members was analysed quantitatively using Cohen's Kappa, which measures inter-rater agreement for qualitative (categorical) items, and takes in to account the possibility of the agreement occurring by chance (Cohen, 1968). All themes except *Relationship* and *Telling their story* were used, as these were the only themes that did not speak directly to the signals of wellbeing or distress.

Quality control

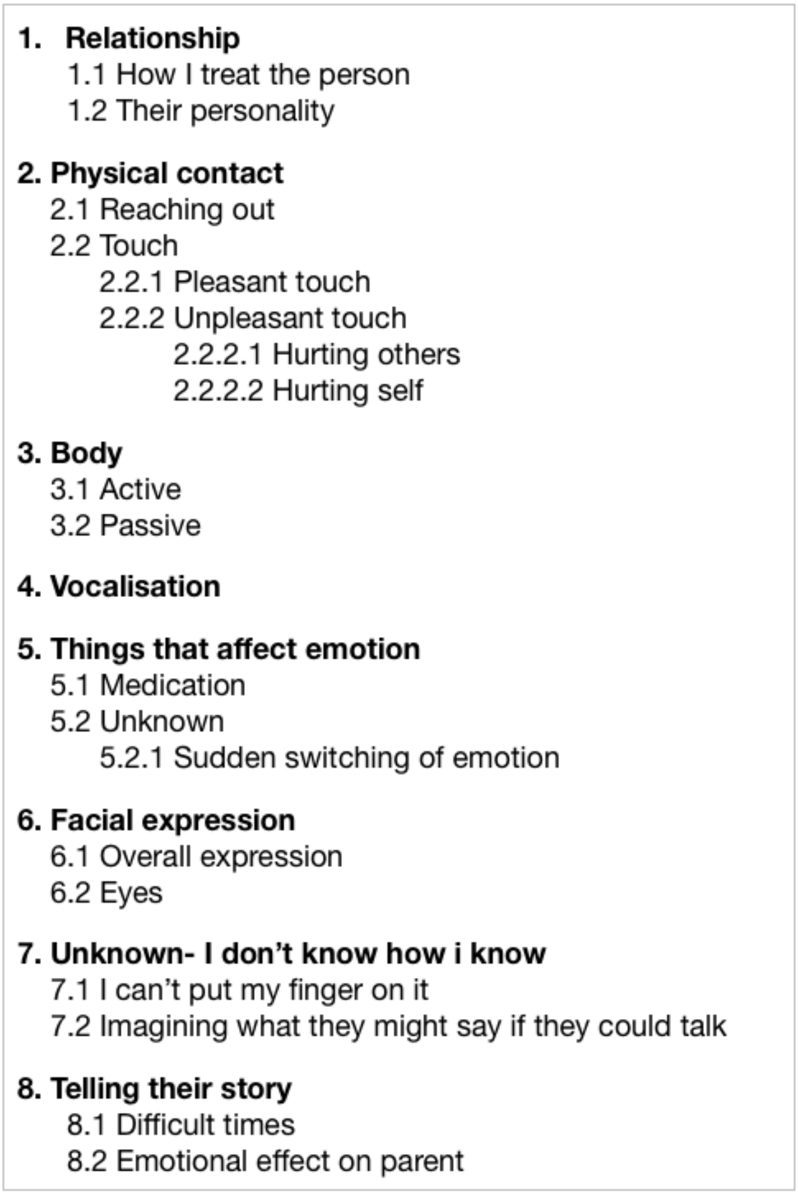
To ensure good research quality, transcripts were systematically checked against audio recordings and the QualSyst quality appraisal checklist (Kmet, Lee, & Cook, 2004) was referred to throughout all stages of the study. The credibility of the analysis was enhanced through a peer-review process whereby another Trainee Clinical Psychologist applied the template to three randomly selected transcripts. No changes were required to the template following this process.

Reflexivity

A reflexive log was kept throughout the whole research process, which included the researcher's thoughts, feelings and assumptions. This was to ensure the researcher's experiences and attitudes were acknowledged and did not unduly influence the findings (Elliot, Fischer, & Rennie, 1999; Shaw 2010). Consideration was given to the researcher's thoughts and emotional reactions to participants, and the role they might play in shaping the template. An issue of note occurred when one of the day services began the process of closing down, which was a surprise to many of the participants. During the last few interviews, this was brought up in conversation by participants. With one exception, most of the discussion with participants about this issue were conducted before or after the recorded interviews. The researcher’s feelings around this, informed discussion with the independent coder as part of the quality control process. The reflexive log was used to explore the researcher’s feelings and reactions around this event which assisted the consideration of this issue during the analysis. After listening to the audio recordings and reviewing the transcripts, potential hypothesis and themes were recorded in the log. Throughout the analytic process, a description of the researchers thought process, and rationale for codes, was noted in the log.

Results

Detailed analysis of 18 participant transcripts produced eight superordinate themes relating to: relationship; physical contact; body; vocalisation; things that affect their emotions; facial expression; unknown; and telling their story. Seven superordinate themes yielded second and/or third level themes and one yielded two fourth level themes (see figure 1). The theme of *telling their story* was specific to the family participants.



1. **Relationship**
    - 1.1 How I treat the person
    - 1.2 Their personality

2. **Physical contact**
    - 2.1 Reaching out
    - 2.2 Touch
        - 2.2.1 Pleasant touch
        - 2.2.2 Unpleasant touch
            - 2.2.2.1 Hurting others
            - 2.2.2.2 Hurting self

3. **Body**
    - 3.1 Active
    - 3.2 Passive

4. **Vocalisation**

5. **Things that affect emotion**
    - 5.1 Medication
    - 5.2 Unknown
        - 5.2.1 Sudden switching of emotion

6. **Facial expression**
    - 6.1 Overall expression
    - 6.2 Eyes

7. **Unknown- I don't know how i know**
    - 7.1 I can't put my finger on it
    - 7.2 Imagining what they might say if they could talk

8. **Telling their story**
    - 8.1 Difficult times
    - 8.2 Emotional effect on parent

*Figure 1.*Final template

Cohen's Kappa was run to determine if there was agreement between paid carer and family dyads on the indicators of distress or wellbeing. Themes 2-7 were used for this calculation, and scored as either present or absent in the participants' transcripts (see appendix 9 for themes for each participant). Agreement was mixed (see appendix 7), with one dyad having excellent agreement, two dyads having moderate agreement, four dyads having fair agreement, and two dyads having poor agreement.

Themes

1. Relationship. Participants spoke about their relationship with the person they care for, and how well they feel they connect with each other. Two second level themes emerged from the data whereby participants spoke about how they tried to treat the person and references made to their personality.

1.1 How I treat the person. Participants spoke of regarding the individual with respect, understanding and treating them as they would anyone else. One paid carer said she tries to keep in mind how she would feel if she were in their position:

> I: How do you feel that you're able to communicate with her, despite her not being able to speak?
>
> P10. "*You speak to that individual the same way- well I do- in the same way that you speak to anybody else. You don't know how much understanding that person's got, how much awareness they got, so you've got to assume that they're really aware of what's going on so that the premise that I work on- how would I feel if I was in that position? How would I like somebody to speak to me?"*

A parent spoke of treating the person with patience and understanding, but felt it important to voice the skills needed to communicate cannot be learned in books:

> P14: *"And you know the main thing? Patience and understanding. If you haven't got them things, you're not- you don't want to be in the job. […] You want to learn about them- not easy. You can't get that out of books. Because the books are written across the board, as if they're the same, and they're not. And this is where they go wrong in lots of things. They make these rules about.. for mentally handicapped kids. It doesn't work like that. No, they're all individual. What suits one doesn't suit another."*

1.2 Their personality. Participants also spoke of the individual's personality. One participant spoke about how strong willed their daughter is:

> I: You said that it's really obvious when she's angry?
>
> *P02: "Yes. Oh, that…she just fights you. She just doesn't.. if X doesn't want to do something, then she'll not do it. You can't make her! She has got her own personality and that, she has".*

Another participant spoke about their son's happy disposition:

> *P13. "He's a brilliant happy chappy for the situation that he's in, really he is. I mean, it must be holding him inside… if he'd have been…I suppose he'd have been as lively as me. I'm the same. He's my son, it's hereditary."*

In summary, analysis identified a distinction between how the individual is treated and the individual's personality. Many participants indicated individuals with an ID should be treated the same as anyone else, and they recognised they have their own personality and disposition and many felt the care they receive should reflect this individuality.

2. Physical contact. Physical contact seemed to be an important theme when individuals are trying to let their carers know how they are feeling. There were two level 2 themes; two level 3 themes; and two level 4 themes identified within the superordinate theme.

2.1. Reaching out. Participants spoke of individuals reaching out for physical contact when trying to convey emotion. One paid carer spoke of how one lady expresses distress:

> P10. *"If she's distressed, you'll walk up, she'll interact with you, she'll give you eye contact, she'll follow you and stuff, and as I said, she'll*

> *reach out as if to say 'I'm not quite happy today'. ..Well, that's how I interpret it anyway."*

2.2 Touch. Individuals were also described as seeking touch from others when they were trying to convey how they are feeling. A distinction emerged between 'pleasant' and 'unpleasant' touch.

2.2.1 Pleasant. Participants spoke of individuals holding hands or stroking and patting others, which the carers described as an expression of a need for comfort or an expression of wellbeing. One paid carer described an individual who, they believed, conveyed distress by seeking to be comforted by touch:

> P08. *" If he's feeling sad […] and if he wants a bit of comfort he'll grab your hand and want to hold your hand or he'll put your.. his hand on his head…to have a.. like a bit of comfort."*

2.2.2 Unpleasant. Conveying emotion could be shown through touch that was potentially painful, such as hitting, nipping or pulling hair. This third level theme was further split in to two level four themes, of *hurting others* and *hurting self.*

> **2.2.2.1 Hurting others.** Hurting others seemed to be a common theme that often, but not exclusively, was described as being a conveyance of distress of some kind. One mother spoke about how her son nips her when he is angry:
>
> P11. *"He gets mad quite easily […] and he will nip me, or he will nip himself, and he does that when he's really angry, yes."*
>
> **2.2.2.1 Hurting self.** Hurting themselves was also a common theme described, that seemed to be related to the conveyance of distress for

the individual. A paid carer described how an individual would bang their head when he was angry or frustrated:

P15. "*..he will start banging his head. He will bounce, really bad like that. Then he will start to bang his head backwards […] there is a cupboard where he bashed a big hole in the back of his head."*

To summarise, the analysis showed a distinction between *reaching out,* which could be interpreted as a request for physical contact, and actual *touch* which is further split in to *pleasant* and *unpleasant* touch. Physical touch is known to be important for emotional wellbeing, which may explain why this superordinate theme generated level four themes. The *hurting others* or *hurting self* themes highlight the difficulties individuals have in conveying their feelings and needs, when they are not able to communicate this verbally.

3. Body. Individuals were also perceived as using their body to convey emotional states. This consists of two level two themes of 'active' and 'passive'.

3.1 Active. Active is defined as the use of the body in an active and moving way such as by dancing, waving, and tensing. The following extracts demonstrate the various ways individuals use their body in an active way to convey their inner emotional state. One paid carer described one man she cares for who smells her shoulder when he is happy:

I: How do you know when he's feeling well or happy?

P12*. "Because he dances. He likes a jig, he likes a dance, he'll bop up and down. If he's happy with you he comes and smells your shoulder."*

One mother described how her daughter uses her fists and arms when she is frustrated:

> P06*. "If she's frustrated, her arms will go out and clench her fists and things like that….her arms flail out as though she's having a fit, and she's really frustrated."*

Another paid carer described how the lady she cares for uses her arms, and rocks, when she is feeling happy and content:

> P10. *"If she's happy and content, she rocks. She'll sort of sit forward and wave her arms about. I would say now, thinking about it, that perhaps when she has got excited, that that's more intense."*

3.2 Passive. Passive is defined as the use of the body in an inactive way such as turning away, refusing to cooperate or ignoring. A parent described how their son showed distress and anger by turning his body away from them:

> P05*. "If he doesn't like you he ignores you, ha! That's what he'll do..he'll just ..or he'll turn. I can remember when he was in XXX, and he was really distressed being there, and he'd turn his face to the wall.. lie in bed and turn his face to the wall and just ignore you. He used to do that when I said I was going, every time. It was really distressing."*

In summary, participants described the use of an individual's body to convey their inner state in two ways, either as *active* or *passive* use of the body. It is interesting to note within this theme there are examples of how inner states are expressed differently between people, with one action showing frustration for one individual and a similar action denoting happiness in another.

4. Vocalisation. Vocalisation is defined as when an individual makes an audible noise. Vocalisation could include things like: calling out, crying, laughing, screaming and singing. The following extracts demonstrate how individuals use various vocalisations to convey how they are feeling.

One paid carer spoke about how loud and varied a man's vocalisations become when he expresses happiness:

> I: What was he like when he was really happy then?
>
> P18*. "When he's happy, he was full on happy.. you can spot it a mile away! […] He's very, very loud […] and then he'll start squealing, you know what I mean? […] And then he starts clucking. He clucks with his tongue. He laughs out loud… proper loud *HAHA*."*

One mother said her daughter cries when she is upset:

> P06*. "She just makes a noise, a crying noise..if she's really upset you'll get tears, really upset.*

One father spoke about how infectious his son's laughter is when he expresses happiness:

> I: So how does he communicate to you that he's feeling well or happy?
>
> P14*. "Oh.. you can see it in his face..laughing..[…] he laughs so much, yes, he'll laugh so much that you'll come in because he's laughing… you're laughing! Staff will start laughing. They will say- 'he just starts that tummy laugh and then I'm laughing and he's laughing' she'll say 'we're all laughing…what are we laughing at?!"*

One paid carer spoke of a man who always sings 'Happy birthday' when he is expressing happiness:

> I: So, how does he communicate to you that he's feeling well or happy?

> P15. *“He’ll sing ‘Happy Birthday’! Yeah, yeah, he’ll um clap, sing Happy birthday and everybody's involved with him.”*

In summary, vocalisations to show emotion were really varied and so no lower level themes were delineated. It is interesting to note many of the participants described fairly extreme end examples of expressing emotion, for example the man who sings ‘happy birthday’ to express happiness and the man who laughs so much that everyone around him joins in without knowing why they are laughing. This is perhaps an indicator of how carers struggle to notice less intense moods.

5. Things that affect emotion. Participants spoke of things that seemed to affect the emotions of the people they care for. Two level two themes emerged; *Medication* and *Unknown*.

5.1 Medication. Participants identified medication having an effect on individual’s expression of emotions. The following extracts show examples of how participants spoke about medication affecting emotions. One carer spoke of how she wonders if she does not notice negative emotions in a lady she cares for because the medication she takes may be having an effect on them:

> P10*. “She is on anti-epileptic medication which is also a mood stabiliser, so obviously that might have some bearing on her emotions, in terms of suppressing her emotions. […]. You know, so maybe that’s why we’re not seeing the frustration, the anger and that kind of thing.”*

One mother spoke about how her son used to show expressions of more extreme emotion before he was medicated, and how she feels the change has been difficult to adjust to:

> P05*. "And I think he has about 1ml in the morning and 1ml at nights, and it's made a lot of.. he's got some middle.. because he was either really happy- over the top happy- or really distressed…and this now.. you'll still get his smile and get a chuckle now and then, but it's not over the top. It's more in context and it's more how anybody else would be, you know. He's got a lot of middle now, which he never had before. It's taken some getting used to, this new XXX."*

One father spoke of his daughter never expressing a 'temper' and how he thinks this is because of all the medication she takes:

> P02*. "No, she doesn't have a temper. I can't ever say XXX's ever had a temper. She's very placid and happy. Well…she's drugged up to the eyeballs, that's why!"*

5.2 Unknown. A theme emerged around the participants being aware of changes in emotional states in the individual but not knowing why these changes occur. A third level theme of Sudden switching of emotion emerged.

5.2.1 Sudden switching of emotion. Many participants described sudden shifts in emotional states that they found difficult to explain. One mother spoke about her son's behaviour that challenges and how he can suddenly switch to laughing:

> P11*. "Well, we just have to let him ride it out. You can't talk him out of it while he's doing it..while he's in it. So we just have to let him go through it, but then suddenly he might start laughing, and you think 'what were all that about?' […] He does have epilepsy, like something affects his mood for that short term, all of a sudden I can't do anything with him".*

Another paid carer spoke about a man who also switches from a negative emotion to expressions of happiness very quickly:

> P12. *"He brings his shoulders in. But other than that- very contained…blink of an eye, one minute he's sad, the next minute he'll start laughing."*

Another mother also spoke about her son's seeming extremes of emotion and how quickly these emotions can change:

> P05. *"He was either very, very happy or very, very upset, and people used to see one or the other when he was little, and he was like it from when he was very small, from being a couple of months old- more or less as soon as he were sent here. So, people would see him when he was very happy and say 'Oh, it's a shame, but he's no trouble'. Or see him when he was in a melt-down and say 'How do you cope?' and I would say, both times- 'He's not like that all the time'….but he could click from one to the other just like that!".*

To summarise, the themes which emerged were linked to things that effect an individual's emotions that were known (i.e. medication) and unknown. It is common for medical prescribers in the NHS to give medication to people with ID who display *behaviour that challenges*. Sometimes these medications can have an impact on a person's emotional affect. Again, it is also possible these sudden switches of emotion are actually a reflection of carers missing subtle cues that the individual's inner state is changing.

6. Facial expression. Participants identified a range of facial expressions that were important when trying to identify emotions in individuals.

6.1. Overall expression. This secondary theme is defined as changes in the face. For example: frowning, smiling, grimacing or 'pulling a face'. The following extracts demonstrate the variability of this theme. One mother described her son's facial expression when he is feeling distressed:

> P05*. " Um, he makes noises that are distressed and you can see by his face, he pulls faces, you know. Um, I know we've got…it's something…you know..he'll put his lip out, and he'll sort of sneer with his nose if someone's said something he's not fond of. […].. he's got things he does with his mouth, things he does with his nose.."*

A paid carer described one woman who grimaces or pulls faces when she is in distress:

> *P01. "..you would know. Um, her facial expressions as well as shouting out..the grimacing.. […] ..she would keep on with that demeanour of pulling faces."*

One father spoke about his daughter's emotions showing clearly in her facial expressions:

> P06*. "But it's all about contact..her face and facial expressions…being able to understand them. We say to people 'If she's smiling, she's happy, if she's sad and pulling faces, then find out what's wrong with her."*

6.2. Eyes. This secondary theme is defined as anything related to emotion and the eyes. For example, eye gaze, eye contact, and 'bright' eyes. The extracts below demonstrate the importance of eyes when inferring emotional states. One carer described how important eye contact for one woman when she's communicating happiness:

> I: How does she communicate to you that she's feeling well or happy?
>
> P07*. "Yes, eye contact! So if she's […] really interested in things that have been going off. She's distracted by whatever the problem is, so*

> *you don’t get really much eye contact and she's looking sad as well. But if she’s settled and everything’s fine and something happens that she likes she’ll look at it. She’s really interested in the thing that’s happening, she’ll make eye contact with it as well and respond to you.”*

One father described his son’s eyes as being the most important factor in determining his inner state:

> P14*. Oh, he’s angry you can see the angry look […]. Eyes and his mouth, you know…staring! […] You’ve got to …look at his eyes, to make sure everything’s alright.”*

One mother described her daughter, who is not able to vocalise, and how her feelings of happiness show in her eyes:

> I: How does she communicate to you that she’s feeling well or happy?
>
> P02. *“She smiles…her eyes. Yes, her eyes. A lovely smile, her eyes are bright.”*

To summarise, facial expression was an important theme that emerged during the analysis, which yielded two second level themes of *overall expression* and *eyes*. The theme of *eyes* was often a challenge to articulate for participants, who would often use phrases such as ‘you can tell by the eyes’, but when pressed could not specify what it was about the eyes that was so telling.

7. Unknown. This theme was identified as participants commonly found that, while they felt they knew how individuals were feeling, they found it difficult to articulate how they knew. Some participants described it as ‘getting a vibe’ or ‘I can’t put my finger on it’. There was also doubt expressed as to whether they were successful in correctly inferring the individual’s feelings.

7.1. I can't put my finger on it: One paid carer said she felt you had to know the woman she spoke of very well to be able to tell if she was angry or frustrated, but it was difficult to articulate how she knew how the woman was feeling:

> P01*. ".. it's very subtle, and I would say that you would have to be very familiar with her..to get to know her. [You] get vibes from her, like a mum does with a baby, do you know what I mean? But I would say that there will be differences, in how she….I can't put my finger on it."*

One participant used a metaphor to help explain how they knew their daughter had a feeling of wellbeing:

> P02*. " It's a bit like a flower. You know when a flower is in bud? ..And then it opens.. that's what happens. XXX opens! And we wish we could bottle it […] she blossoms, and we don't know…we don't know the real reason as to why…but she does!"*

Participants sometimes described not being sure whether they are successful in understanding the emotional states of the person they care for, as one mother demonstrates:

> P05*. "You've got to be very observant because lots of..very similar gestures and very similar sounds mean different things…and, um, you're never sure whether you've got it right."*

7.2. Imagining what they might say if they could talk. Some participants would try and put in to words what they think the person they care for would say if they could speak. The following extracts are examples of how participants imagined what the person they care for might be saying:

> P10*. ..She'll tense up and she'll sort of..withdraw a little bit. So, to me, that would suggest that it's her saying 'I'm not really interested in it now'.*
>
> P06*. "yes, I presume something is happening in her head..very similar going into a fit..but they're not fits- it's frustration! It's 'get me out of here! I don't like it!'- you can't mistake it."*
>
> P08*. Yes, same when he's upset. If he's upset, or angry or anxious, it gets louder…he's vocalising 'I'm not happy and you need to help me, or 'you need to sort me out', or 'I don't want to do this!'."*

In summary, the theme of *Unknown* had two level two themes of *I can't put my finger on it* and *Imagining what they might say if they could talk*. It is possible this theme is related to attunement that happens between the two people who know each other well, or unconscious communication.

8. Telling their story. This theme emerged during the analysis of family transcripts, as they seemed to want to tell 'their story' during the interview. This theme was not apparent for the paid carer participants. This theme yielded two level two themes: Difficult times; and emotional effect on parent.

8.1. Difficult times. A father spoke about him having a difficult time taking his son out on holidays and to restaurants because of his behaviour that challenges, but how he has persevered:

> P14. "*And like, you know, we've always taken him out. We've took him on holidays and we've said 'Never again!', but we've gone again. We've gone, we've taken him in restaurants, we've persevered […] We've tried everything".*

One father spoke of his worries about service providers for his son in the future:

> P13. *"He has his own set ways of being in his own place, and I've always said- God forbid if somebody ever stops this and they take him away..you know, not to be independent anymore. Because the way things are going with finances, it frightens you sometimes, because the social care funding for one is a shocking.. I mean, somebody wants to be on my side of having to understand things and now I'm getting older."*

8.2 Emotional impact on parent. Participants who were parents often spoke about how difficult things have been for them, and how certain events have felt for them. One mother spoke of the upset she felt at not having the kind of relationship with her daughter she had hoped for:

> I: Thinking about your daughter, can you tell me how you feel about your relationship with her?
>
> P16. *" Well, obviously I wish I had a better one, which I always thought when I did start a family, that I would have a daughter and I would have a mother daughter relationship. So, that upsets me."*

Another mother spoke about a time where she cared for her son full time at home and longed for another person to have a conversation with:

> P5. "*It took me a long time to start communicating with other people because I got to the point where I would think things instead of say them, because there was no feedback. No verbal feedback, and I got to the point with other people where I'd think I'd said something and I hadn't; I'd only thought it. Um, and when, once a week the milkman came to get his money, poor buggar, he only wanted to get paid and go on..and I'd collar him for the sound of another human voice for half an hour on the doorstep, just, you know ..'and another thing!'.."*

In summary, this theme of *Telling their story* has two level two themes of *Difficult Times* and *Emotional impact on parent* ,and neither themes were present for the paid carer. It is interesting that when interviewing parents, many of them spoke of their own emotions and told stories of some difficult times in the family in relation to their son or daughter. It could be said this is an example of parents using themselves, and their own emotions, to help explain the inner state of their loved one.

Discussion

Participants in this study believe people with severe and profound ID express psychological wellbeing and distress in the following ways, through; behavioural, and vocal repertoires; facial expressions; eye contact; touching of others and themselves; and via something unknown. Agreement was found in the themes of both paid carers and family, but an additional theme of *telling their story* emerged in the analysis of the family transcripts.

The theme of the *unknown* is of particular interest. This phenomena of 'I don't know how I know', could be explained by attunement. This has been explained as a state of highly empathic, "mutually pleasurable and symmetrical sociability", that is achieved by people who have known each other for some time (Firth, Berry & Irvine, 2010). This feeling of knowing without knowing how they know is also likely to be explained by unconscious communication, which is verbal or nonverbal communication that is not consciously transmitted between two people that influence the dynamics of their relationship (Roberts, 1994, p.110-121). Unconscious communication can also occur verbally, but as those with severe or profound ID can not communicate verbally, it is likely to occur non verbally in this case. Our understanding may be improved if unconscious processes are taken into consideration, as well as the more overt signals and visible behaviours.

In most cases, analysis of inter-rater reliability showed a fair agreement between each dyad. Previous research into the inter rater reliability of carers of those with severe and profound ID, has shown mixed results, with some research showing a lack of agreement between respondents when interpreting affective communication (Hogg, Reeves, Roberts & Mudford, 2001). Other research has reported a greater concordance (McVilly, Burton Smith, & Davidson, 2000). When there is no way of resolving ambiguity by asking the person concerned what they meant, validity of the interpretation remains a key issue (Grove, Bunning, Porter, & Olsson, 1999), and so further research is needed.

The results of this research showed expression of distress and wellbeing did not share a commonality among individuals with severe and profound ID. Although some wellbeing and distress cues were common between individuals, each person seemed to have their own distinct profile of how they showed wellbeing and distress. As examples shows, one individual would reach out for others when they were feeling distress, while another would reach out for others when they were thought to feel happy. This may be another reason for the paucity of tools measuring wellbeing and distress for this group.

Lyons (2003) suggested interpretation of feelings and emotions may be best validated by triangulating data on a person’s communicative behaviour profile that is obtained from two familiar observers and a third unfamiliar observer. Communicating with a person without verbal expression usually relies on the interpreter drawing on the full range of information available to them. This might involve using prior knowledge, as well as feelings, attitudes and the context in which they operate (Grove et al., 1999). While the familiar observers have the advantage of knowing the person and their context, the unfamiliar observer will be unprejudiced by past events and personal context, and so may provide a complimentary source of information alongside the familiar observers. Petry and Maes (2006) used parents, support workers and an external researcher to rate expressions of pleasure and displeasure for people with severe and profound ID. They concluded it is possible to draft meaningful individual profiles of affective expressions. Similar to the present research, they also found different respondents mention some of the same expressions of affect, but also some different expressions, and since there is no absolute way of knowing whether any of these identifications are correct, these differences can be seen as complementary to the overall profile of expressions of the individual (Petry & Maes, 2006). Other researchers have concluded interpretations should not be labeled as either right or wrong. Instead, they should be viewed as existing on a continuum where at one end they achieve a fully shared interpretation and at the other end a lack of any shared agreement (Griffiths & Smith, 2016).

The theme around medication emerged not as a result of direct questioning. It is common for medical prescribers in the NHS to give medication to people with ID who have a diagnosed mental health problem. Recent estimates indicate prescription rates are 2-4 times higher than the rate of diagnosed mental health problems for those with ID (Glover, 2015; Tsiouris, Kim, Brown, Pettinger, & Cohen, 2013), and these prescriptions are given to reduce behaviour that challenges. One of the possible side effects of prescribing psychotropic medication to a person without a diagnosed mental health problem can be changes in cognition (Deb, 2014), which could potentially make recognition of distress or wellbeing more difficult for carers to identify. It is unknown whether individuals spoke of in this study had a diagnosis of a mental health problem, but the results suggest medication impacts on the parent or carers ability to recognise feelings of wellbeing or distress.

Limitations and research implications

A strength of this study is the representation of both paid carer and family members. Replication of the findings with a larger sample size would be necessary to assess whether the themes identified are representative of a wider population. Although the sample was predominantly women, this may be considered representative of the wider carer population. Efforts were made to recruit other family members, such as siblings, so as to ensure maximum differentiation in the qualitative analysis. However, all family members who self-identified as the person best able to recognize emotions were parents. All participants were also of white British descent and so future research would benefit from a more familial and culturally diverse sample, which would make the results more generalisable to the wider population.

The researcher also acknowledges that qualitative analysis, while identifying useful trends in the data, is an interpretive process (King, 2004), and the researcher's interpretation will have been influenced by their knowledge and experiences. Future research designs that employ qualitative and quantitative methods are recommended.

Inter-rater reliability is particularly challenging when evaluating emotions and feelings in those with severe and profound ID, and since we have no way of asking the person how they feel, future research that aims try and understand and how to improve inter rater reliability, is vital. It is recommended further research on the *Unknown* theme yielded from this analysis would also be beneficial. Measurement of wellbeing in the real world is made difficult if there is an 'unknown' element in how carers identify emotions. Further qualitative research with a wider familial and culturally diverse sample may shed some clarification on this theme.

Clinical implications

Recognition that a person with severe and profound ID can, and does, communicate feelings of psychological distress and wellbeing is an important source of dignity and respect. Clinicians should consider those with severe or profound ID use their body, vocalisation, physical contact and facial expressions to communicate wellbeing or distress to their carers, but that each have unique individual profiles. Clinicians should be aware that medication side effects may impact on carer's ability to recognise emotions in those they care for. Clinicians also might consider the potential role unconscious communication, may have in identifying inner emotional states in those with severe and profound ID.

Conclusion

The present study sought to explore paid carer and family perspectives on how those with severe or profound ID express wellbeing and distress, and to compare themes between respondents . Analysis of participants' transcripts yielded eight superordinate themes relating to: relationship; physical contact; body; vocalisation; things that affect emotion; facial expression and the unknown. The eighth theme emerged only from the family transcripts: telling their story, with all other themes being present in both paid carer and family transcripts. The level of agreement was also analysed using Cohen's Kappa and found there was fair agreement between the dyads. The theme of *unknown* is of particular interest, and this may be explained by unconscious communication between the carer and individual. However, further research is needed.

This study makes a contribution to a small but growing body of research which recognises the importance of the inner emotional states of those who are not able to communicate their needs and wishes. Further research is needed with a more culturally diverse and a larger sample size to test these findings.

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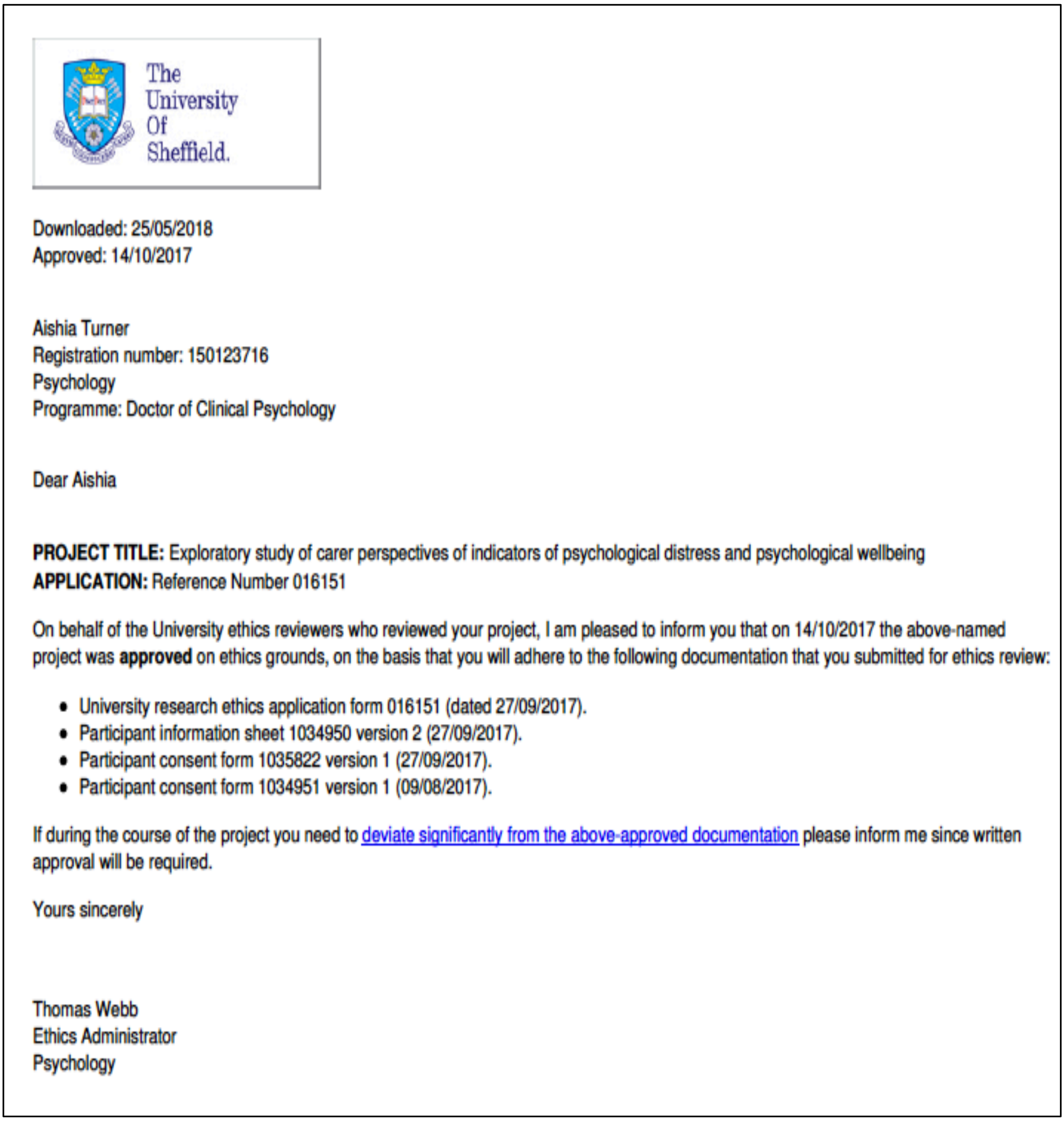
Appendix 1. University ethical approval



The
University
Of
Sheffield.

Approved: 14/10/2017

Aishia Turner
Registration number: 150123716
Psychology
Programme: Doctor of Clinical Psychology

Dear Aishia

PROJECT TITLE: Exploratory study of carer perspectives of indicators of psychological distress and psychological wellbeing
APPLICATION: Reference Number 016151

On behalf of the University ethics reviewers who reviewed your project, I am pleased to inform you that on 14/10/2017 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:

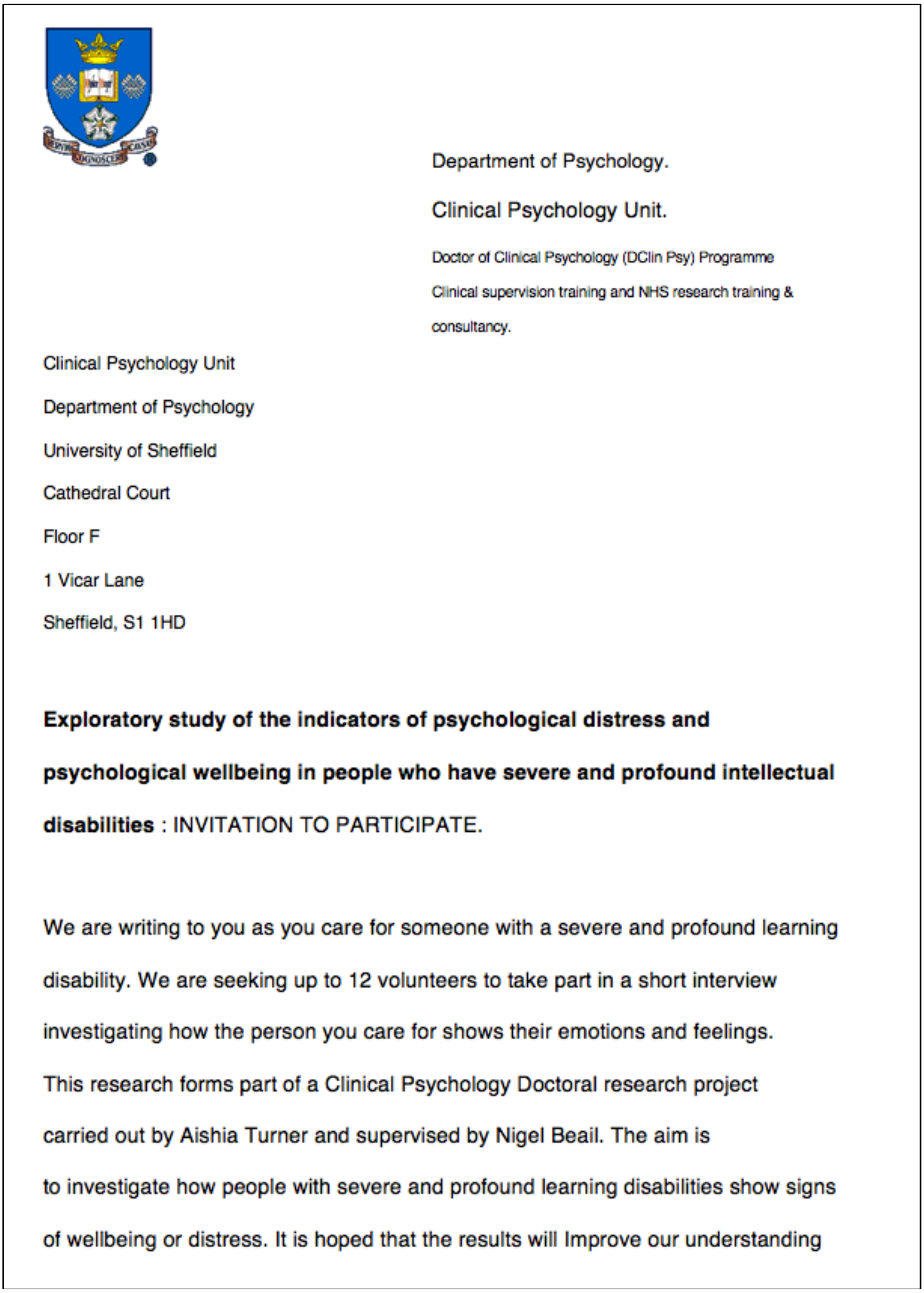
- University research ethics application form 016151 (dated 27/09/2017).
- Participant information sheet 1034950 version 2 (27/09/2017).
- Participant consent form 1035822 version 1 (27/09/2017).
- Participant consent form 1034951 version 1 (09/08/2017).

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

Thomas Webb
Ethics Administrator
Psychology

Appendix 2. Invitation letter to participate



Department of Psychology.

Clinical Psychology Unit.

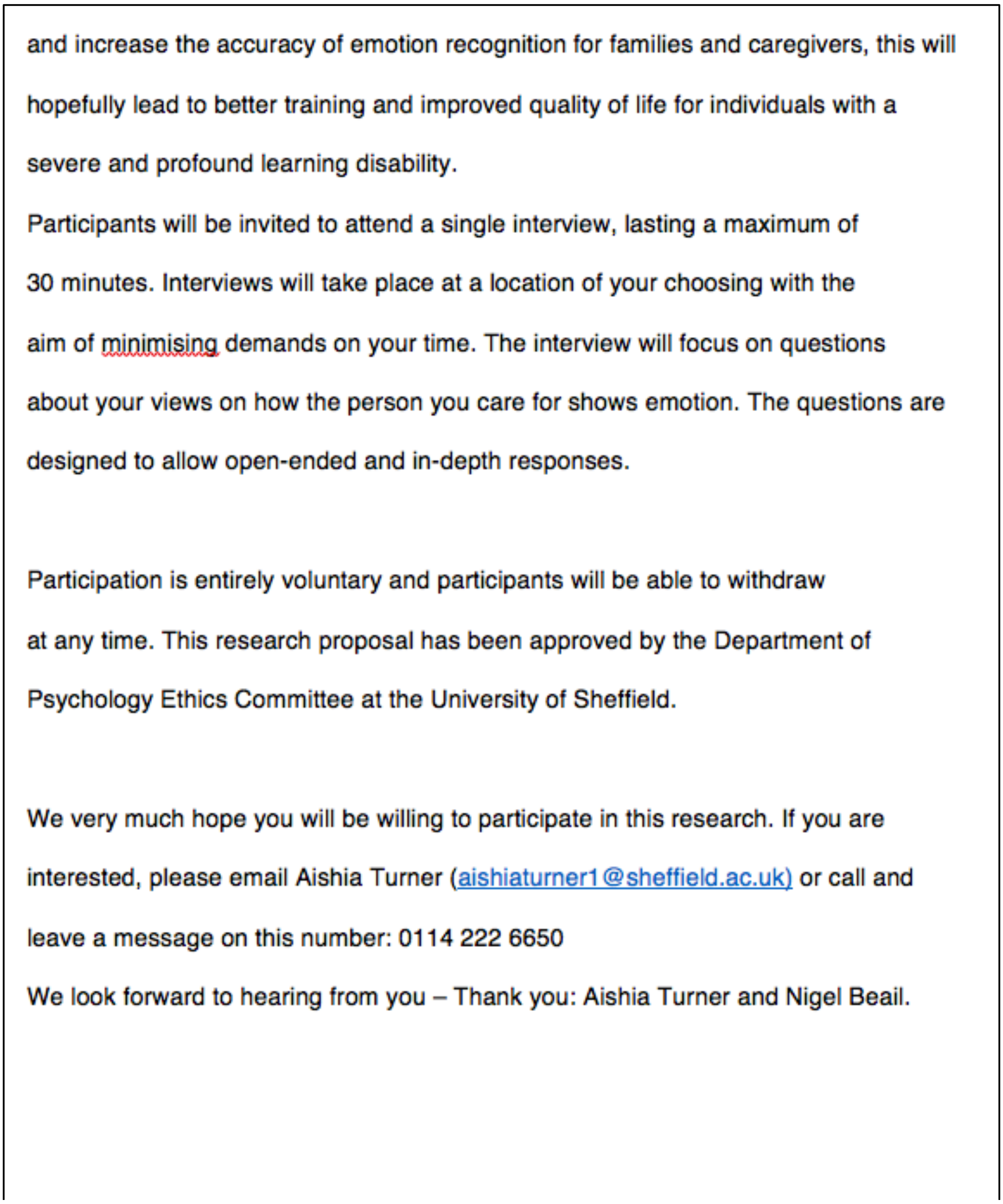
Doctor of Clinical Psychology (DClin Psy) Programme

Clinical supervision training and NHS research training & consultancy.

Clinical Psychology Unit
Department of Psychology
University of Sheffield
Cathedral Court
Floor F
1 Vicar Lane
Sheffield, S1 1HD

Exploratory study of the indicators of psychological distress and psychological wellbeing in people who have severe and profound intellectual disabilities : INVITATION TO PARTICIPATE.

We are writing to you as you care for someone with a severe and profound learning disability. We are seeking up to 12 volunteers to take part in a short interview investigating how the person you care for shows their emotions and feelings. This research forms part of a Clinical Psychology Doctoral research project carried out by Aishia Turner and supervised by Nigel Beail. The aim is to investigate how people with severe and profound learning disabilities show signs of wellbeing or distress. It is hoped that the results will Improve our understanding



and increase the accuracy of emotion recognition for families and caregivers, this will hopefully lead to better training and improved quality of life for individuals with a severe and profound learning disability.

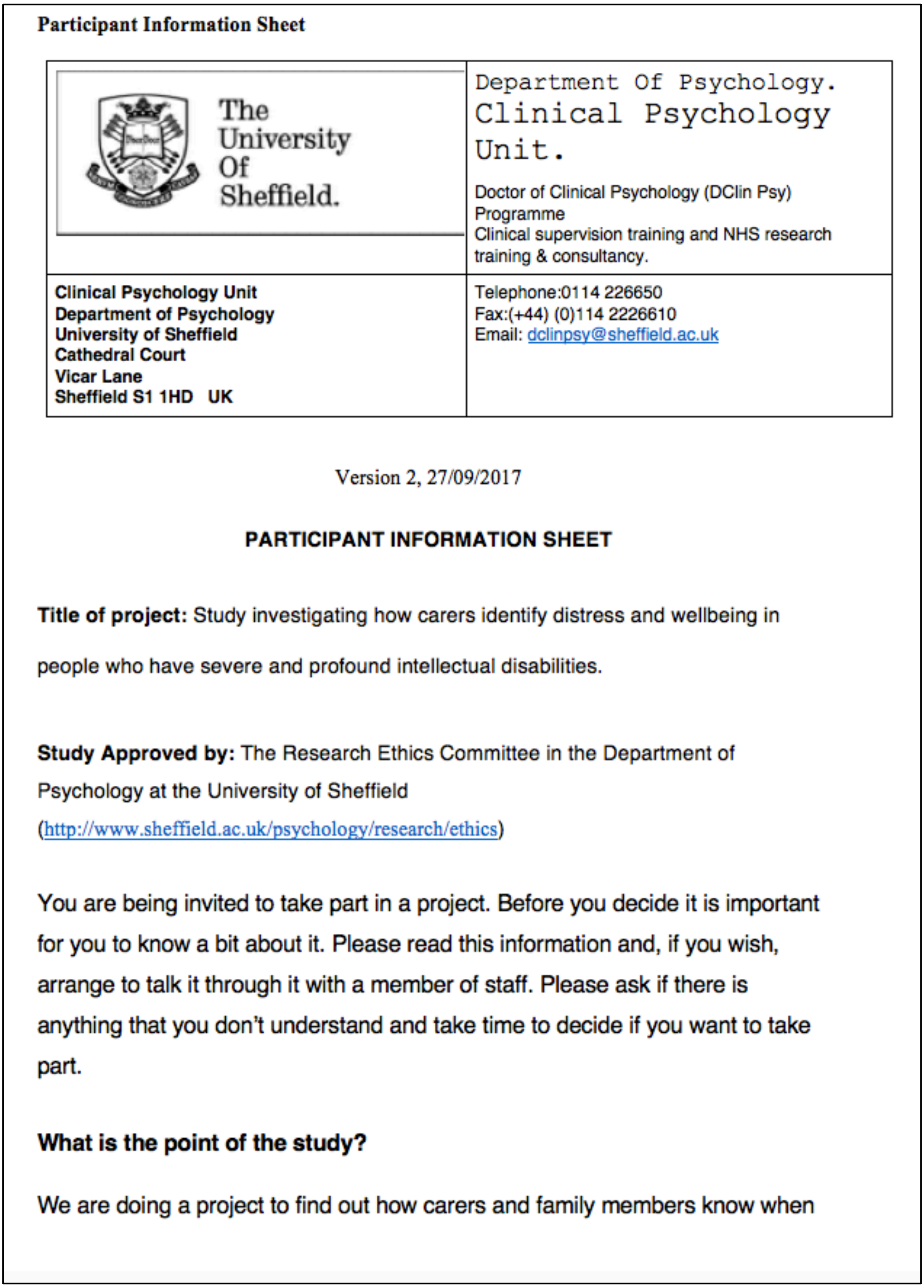
Participants will be invited to attend a single interview, lasting a maximum of 30 minutes. Interviews will take place at a location of your choosing with the aim of minimising demands on your time. The interview will focus on questions about your views on how the person you care for shows emotion. The questions are designed to allow open-ended and in-depth responses.

Participation is entirely voluntary and participants will be able to withdraw at any time. This research proposal has been approved by the Department of Psychology Ethics Committee at the University of Sheffield.

We very much hope you will be willing to participate in this research. If you are interested, please email Aishia Turner (aishiaturner1@sheffield.ac.uk) or call and leave a message on this number: 0114 222 6650

We look forward to hearing from you – Thank you: Aishia Turner and Nigel Beail.

Appendix 3. Participant information sheet



Participant Information Sheet

The University Of Sheffield.	Department Of Psychology. Clinical Psychology Unit. Doctor of Clinical Psychology (DClin Psy) Programme Clinical supervision training and NHS research training & consultancy.
Clinical Psychology Unit **Department of Psychology** **University of Sheffield** **Cathedral Court** **Vicar Lane** **Sheffield S1 1HD UK**	Telephone:0114 226650 Fax:(+44) (0)114 2226610 Email: dclinpsy@sheffield.ac.uk

Version 2, 27/09/2017

PARTICIPANT INFORMATION SHEET

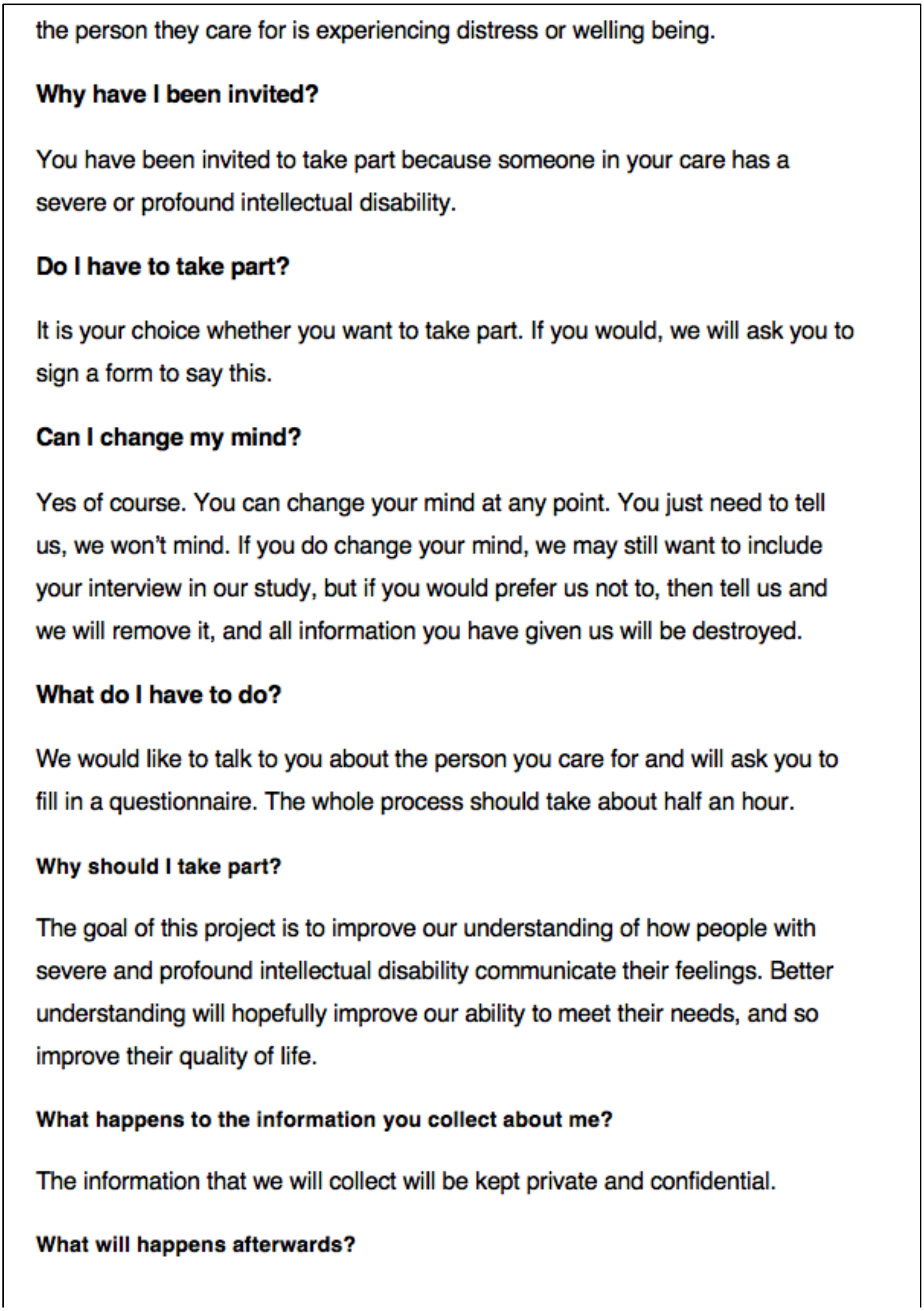
Title of project: Study investigating how carers identify distress and wellbeing in people who have severe and profound intellectual disabilities.

Study Approved by: The Research Ethics Committee in the Department of Psychology at the University of Sheffield (http://www.sheffield.ac.uk/psychology/research/ethics)

You are being invited to take part in a project. Before you decide it is important for you to know a bit about it. Please read this information and, if you wish, arrange to talk it through it with a member of staff. Please ask if there is anything that you don't understand and take time to decide if you want to take part.

What is the point of the study?

We are doing a project to find out how carers and family members know when



the person they care for is experiencing distress or welling being.

Why have I been invited?

You have been invited to take part because someone in your care has a severe or profound intellectual disability.

Do I have to take part?

It is your choice whether you want to take part. If you would, we will ask you to sign a form to say this.

Can I change my mind?

Yes of course. You can change your mind at any point. You just need to tell us, we won't mind. If you do change your mind, we may still want to include your interview in our study, but if you would prefer us not to, then tell us and we will remove it, and all information you have given us will be destroyed.

What do I have to do?

We would like to talk to you about the person you care for and will ask you to fill in a questionnaire. The whole process should take about half an hour.

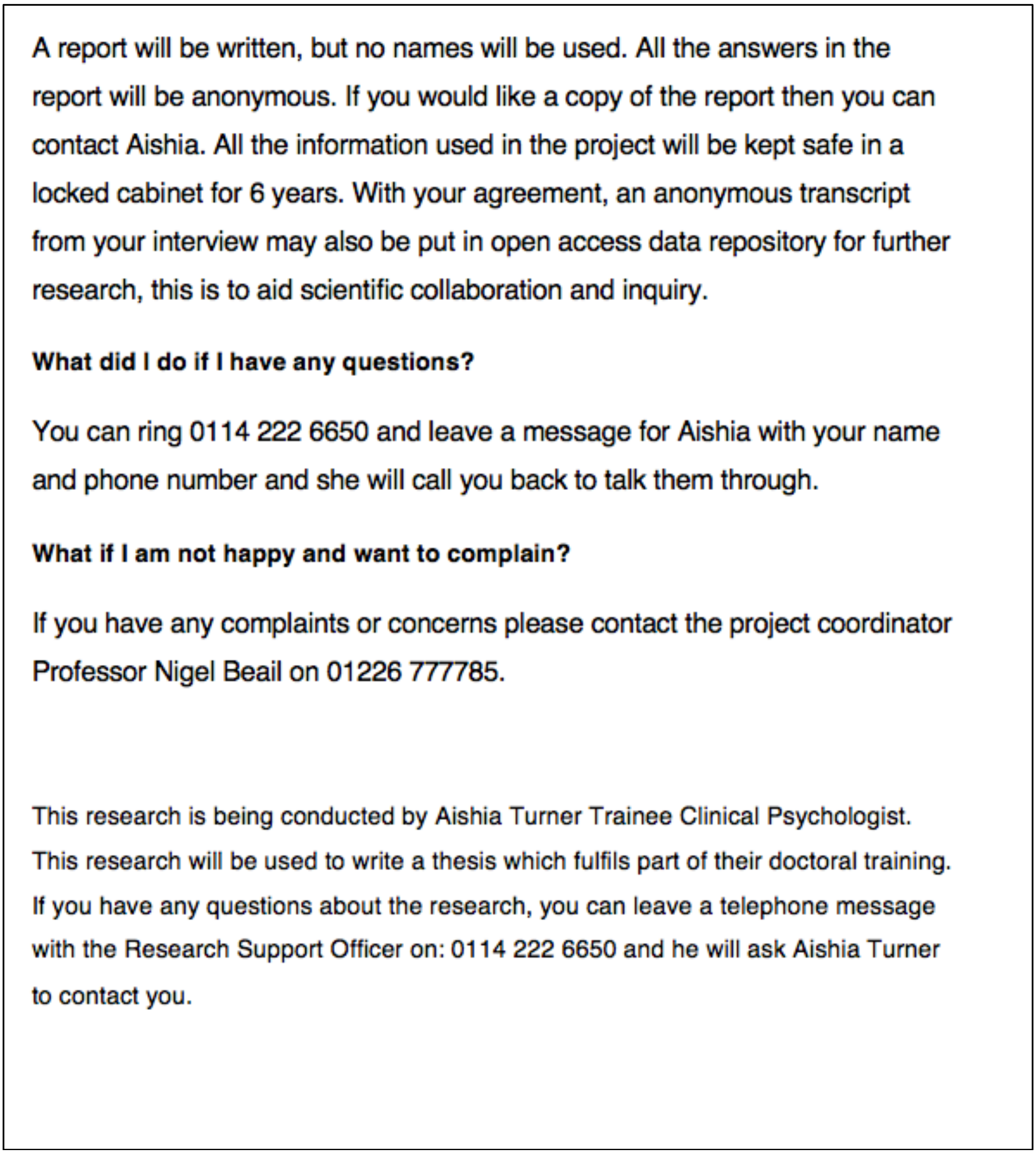
Why should I take part?

The goal of this project is to improve our understanding of how people with severe and profound intellectual disability communicate their feelings. Better understanding will hopefully improve our ability to meet their needs, and so improve their quality of life.

What happens to the information you collect about me?

The information that we will collect will be kept private and confidential.

What will happens afterwards?



A report will be written, but no names will be used. All the answers in the report will be anonymous. If you would like a copy of the report then you can contact Aishia. All the information used in the project will be kept safe in a locked cabinet for 6 years. With your agreement, an anonymous transcript from your interview may also be put in open access data repository for further research, this is to aid scientific collaboration and inquiry.

What did I do if I have any questions?

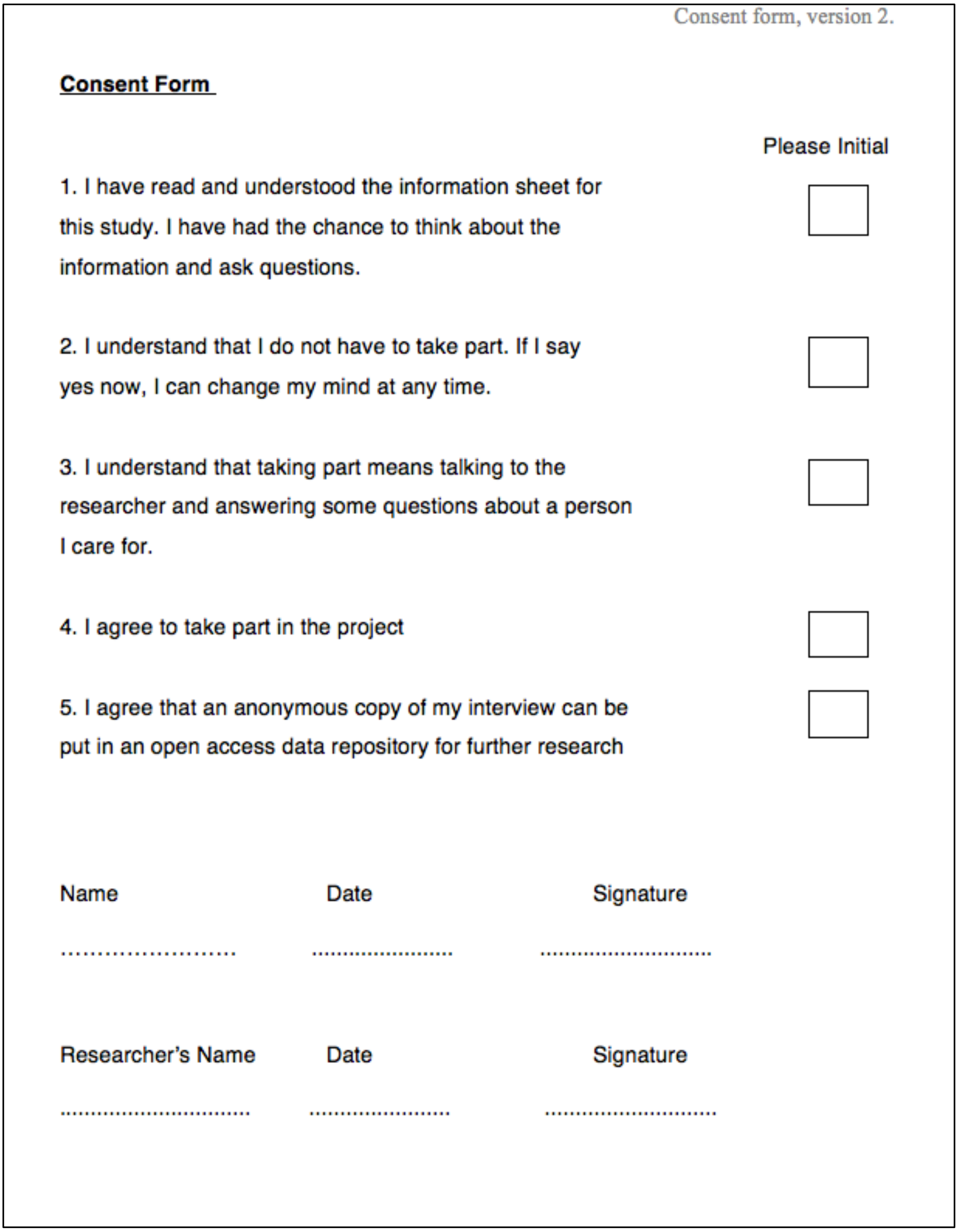
You can ring 0114 222 6650 and leave a message for Aishia with your name and phone number and she will call you back to talk them through.

What if I am not happy and want to complain?

If you have any complaints or concerns please contact the project coordinator Professor Nigel Beail on 01226 777785.

This research is being conducted by Aishia Turner Trainee Clinical Psychologist. This research will be used to write a thesis which fulfils part of their doctoral training. If you have any questions about the research, you can leave a telephone message with the Research Support Officer on: 0114 222 6650 and he will ask Aishia Turner to contact you.

Appendix 4. Participant consent form



Consent form, version 2.

Consent Form

Please Initial

1. I have read and understood the information sheet for this study. I have had the chance to think about the information and ask questions. ☐

2. I understand that I do not have to take part. If I say yes now, I can change my mind at any time. ☐

3. I understand that taking part means talking to the researcher and answering some questions about a person I care for. ☐

4. I agree to take part in the project ☐

5. I agree that an anonymous copy of my interview can be put in an open access data repository for further research ☐

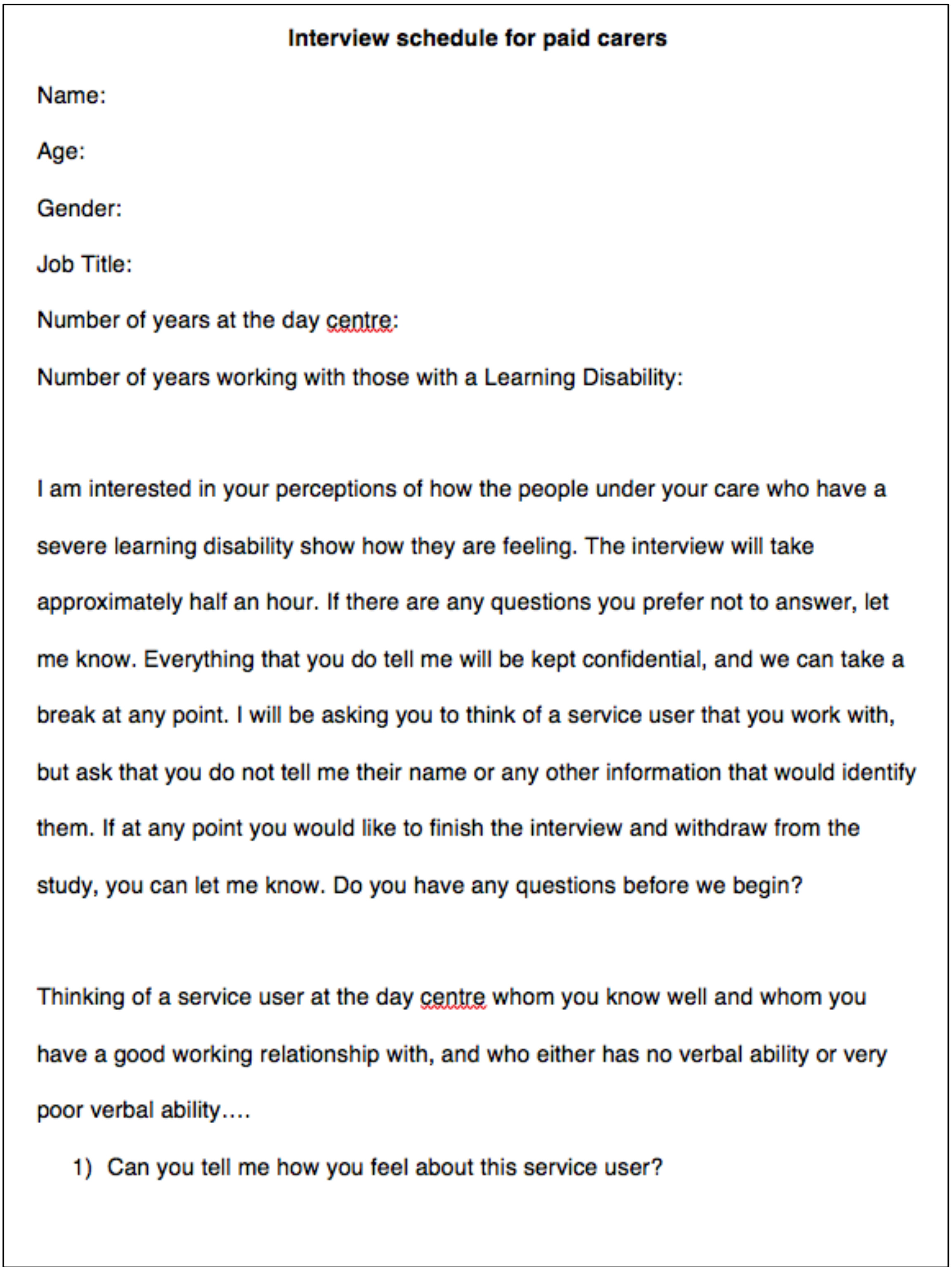
Name Date Signature

…………………… …………………… …………………………

Researcher's Name Date Signature

………………………… …………………… …………………………

Appendix 5. Interview Schedule



Interview schedule for paid carers

Name:

Age:

Gender:

Job Title:

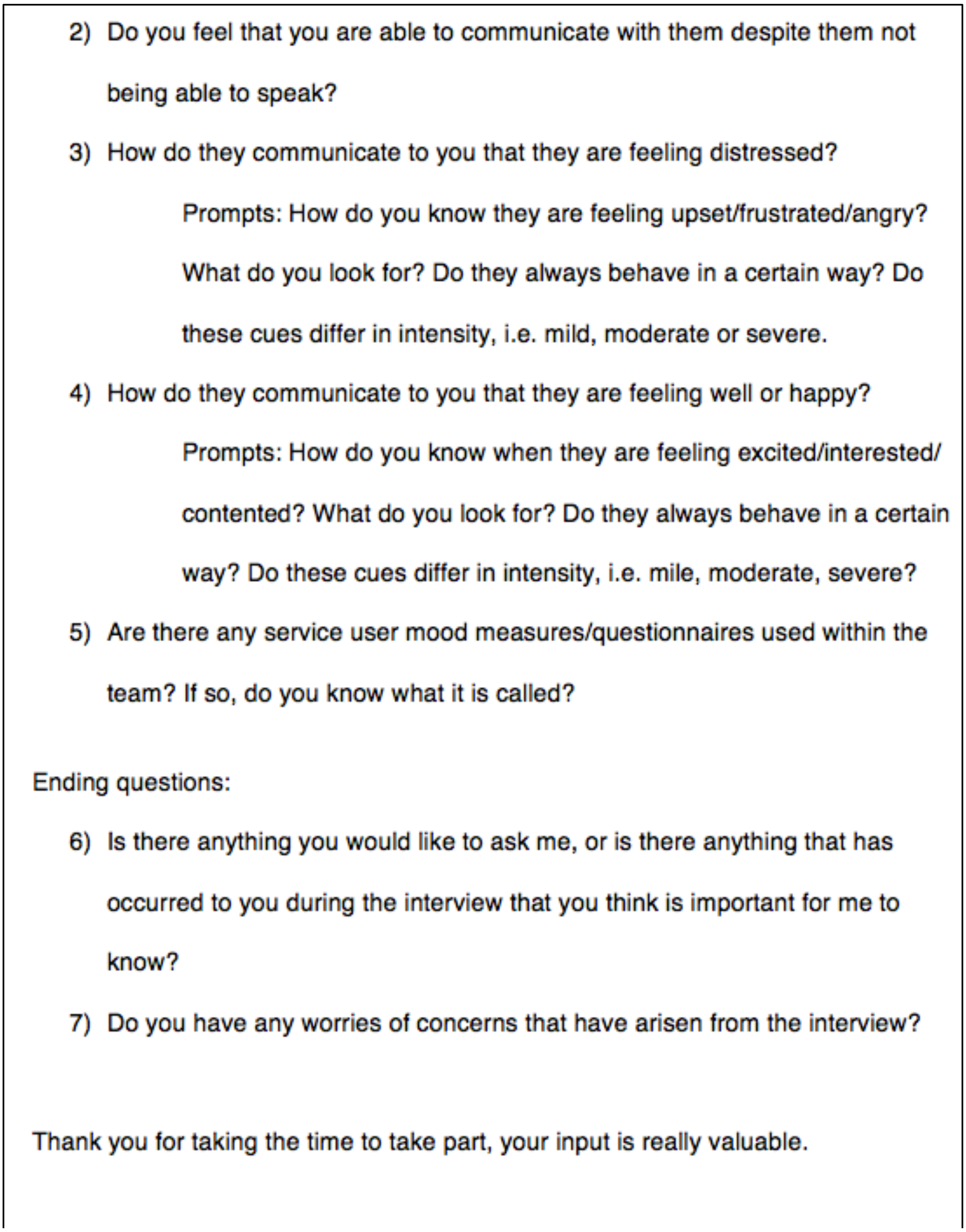
Number of years at the day centre:

Number of years working with those with a Learning Disability:

I am interested in your perceptions of how the people under your care who have a severe learning disability show how they are feeling. The interview will take approximately half an hour. If there are any questions you prefer not to answer, let me know. Everything that you do tell me will be kept confidential, and we can take a break at any point. I will be asking you to think of a service user that you work with, but ask that you do not tell me their name or any other information that would identify them. If at any point you would like to finish the interview and withdraw from the study, you can let me know. Do you have any questions before we begin?

Thinking of a service user at the day centre whom you know well and whom you have a good working relationship with, and who either has no verbal ability or very poor verbal ability….

1) Can you tell me how you feel about this service user?



2) Do you feel that you are able to communicate with them despite them not being able to speak?
3) How do they communicate to you that they are feeling distressed?

   Prompts: How do you know they are feeling upset/frustrated/angry? What do you look for? Do they always behave in a certain way? Do these cues differ in intensity, i.e. mild, moderate or severe.

4) How do they communicate to you that they are feeling well or happy?

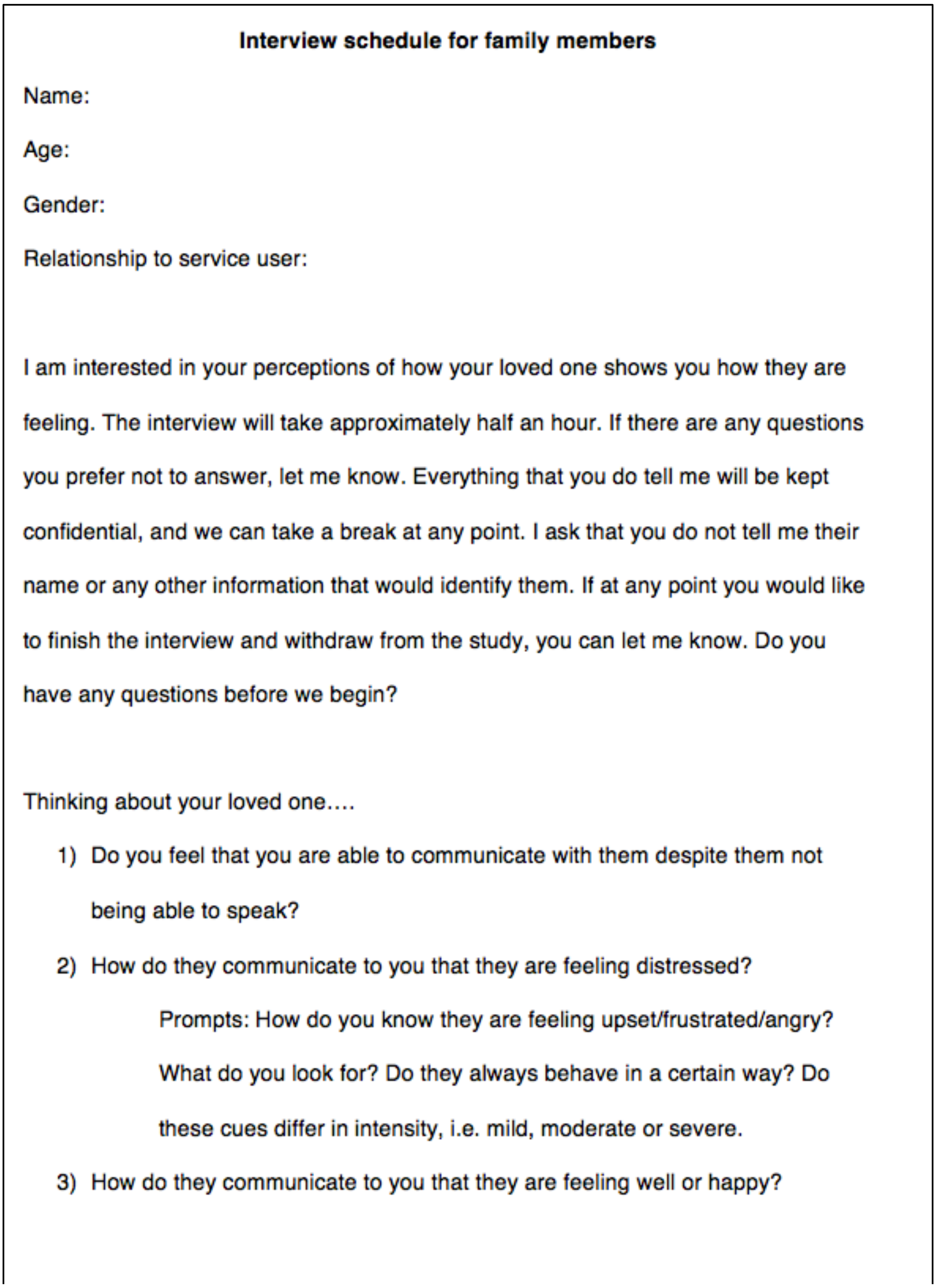
   Prompts: How do you know when they are feeling excited/interested/ contented? What do you look for? Do they always behave in a certain way? Do these cues differ in intensity, i.e. mile, moderate, severe?

5) Are there any service user mood measures/questionnaires used within the team? If so, do you know what it is called?

Ending questions:

6) Is there anything you would like to ask me, or is there anything that has occurred to you during the interview that you think is important for me to know?
7) Do you have any worries of concerns that have arisen from the interview?

Thank you for taking the time to take part, your input is really valuable.



Interview schedule for family members

Name:

Age:

Gender:

Relationship to service user:

I am interested in your perceptions of how your loved one shows you how they are feeling. The interview will take approximately half an hour. If there are any questions you prefer not to answer, let me know. Everything that you do tell me will be kept confidential, and we can take a break at any point. I ask that you do not tell me their name or any other information that would identify them. If at any point you would like to finish the interview and withdraw from the study, you can let me know. Do you have any questions before we begin?

Thinking about your loved one….

1) Do you feel that you are able to communicate with them despite them not being able to speak?
2) How do they communicate to you that they are feeling distressed?

    Prompts: How do you know they are feeling upset/frustrated/angry? What do you look for? Do they always behave in a certain way? Do these cues differ in intensity, i.e. mild, moderate or severe.

3) How do they communicate to you that they are feeling well or happy?

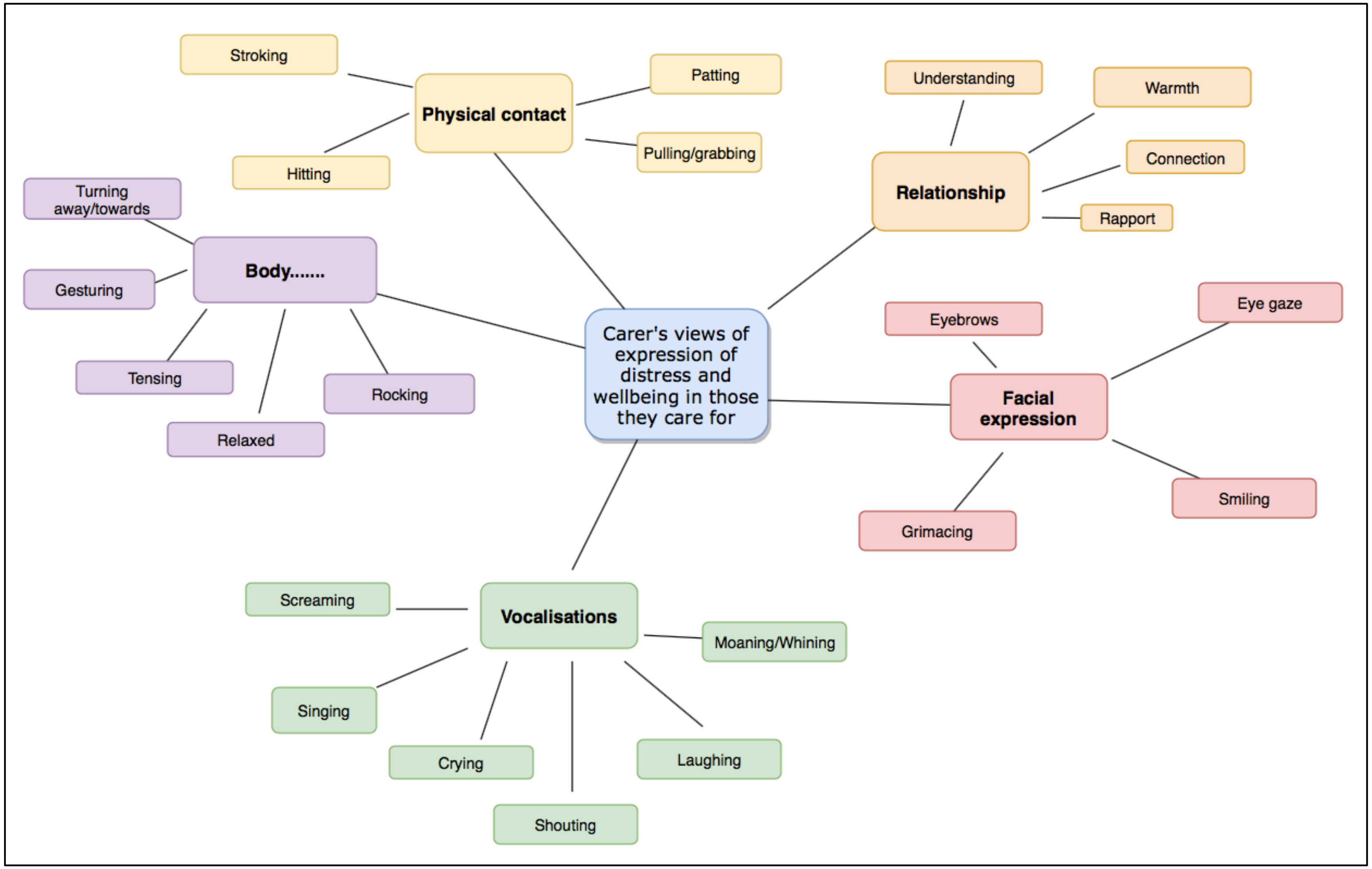
Prompts: How do you know when they are feeling excited/interested/ contented? What do you look for? Do they always behave in a certain way? Do these cues differ in intensity, i.e. mile, moderate, severe?

Ending questions:

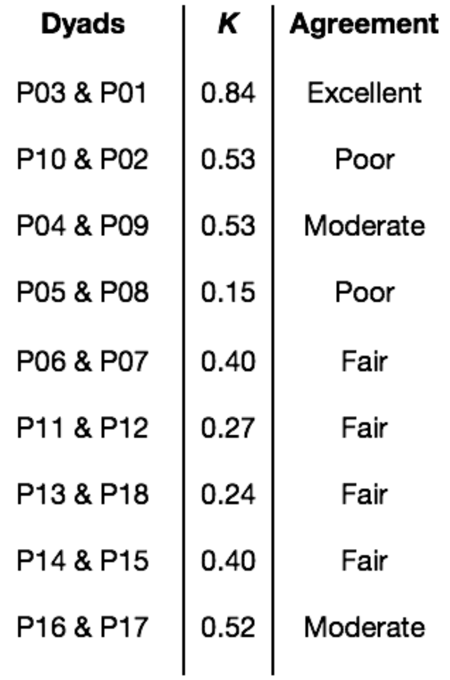
4) Is there anything you would like to ask me, or is there anything that has occurred to you during the interview that you think is important for me to know?
5) Do you have any worries of concerns that have arisen from the interview?

Thank you for taking the time to take part, your input is really valuable.

Appendix 6. Conceptual map of priori themes

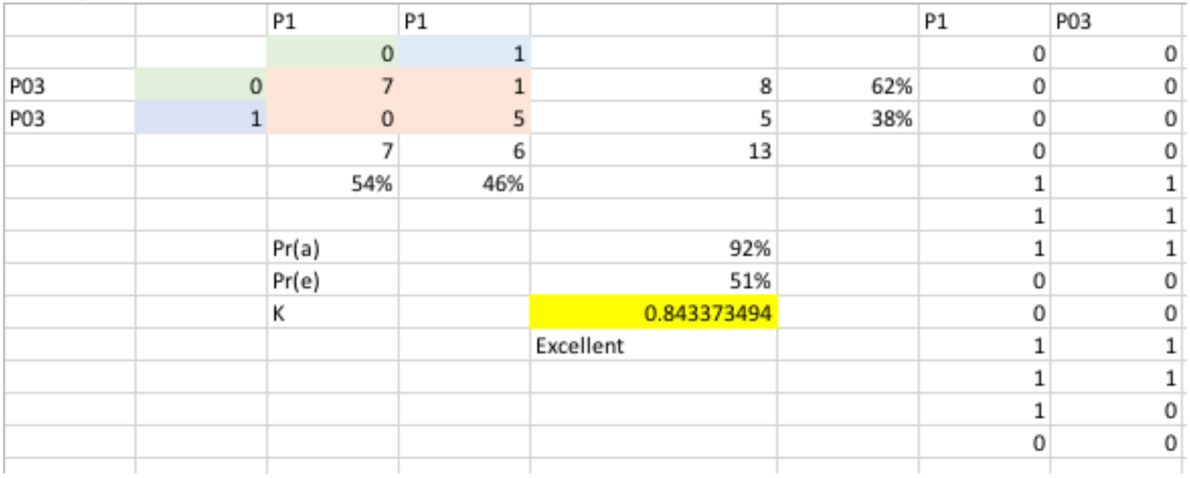


Appendix 7. Cohens cappa calculation



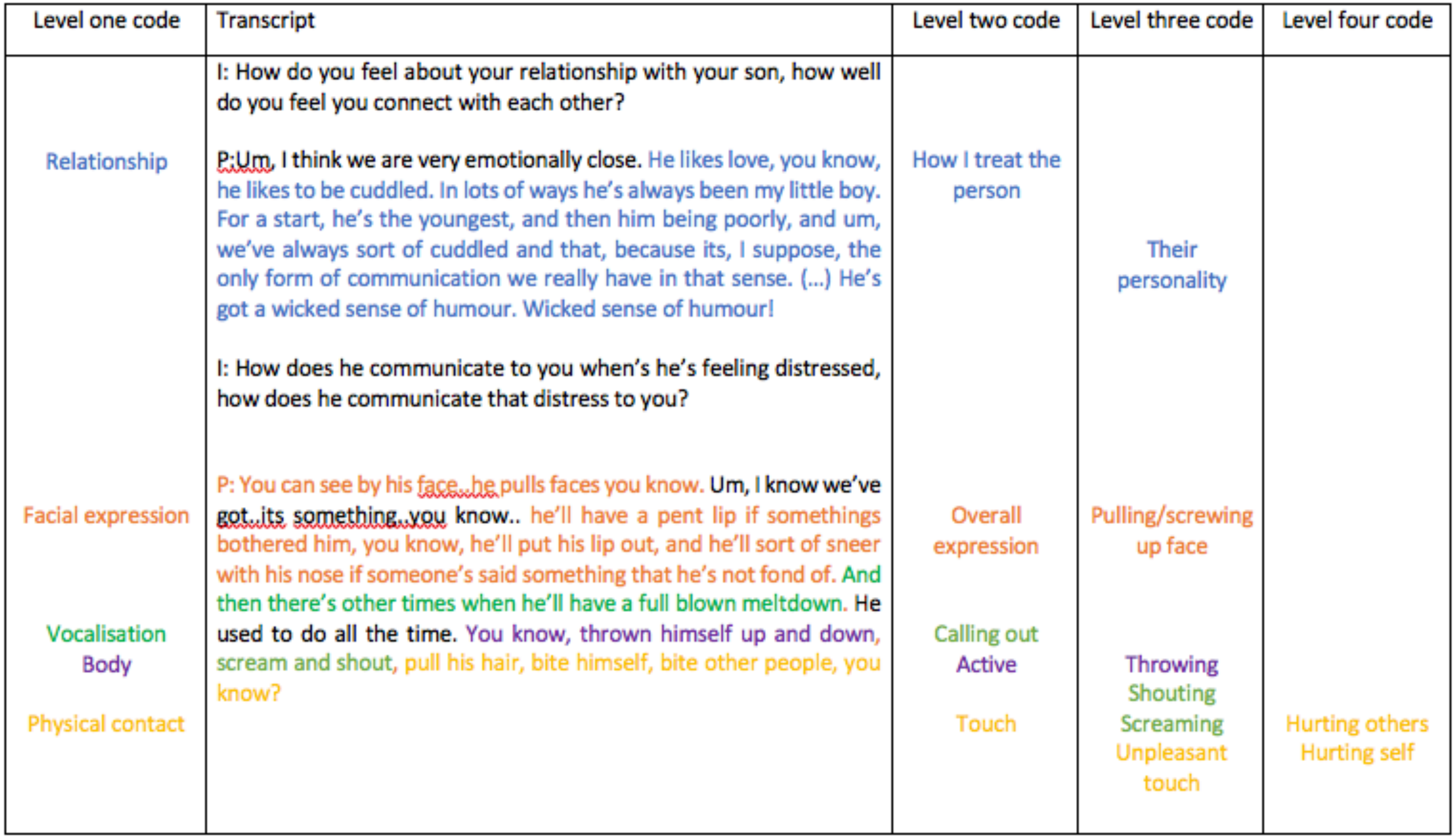
Dyads	*K*	Agreement
P03 & P01	0.84	Excellent
P10 & P02	0.53	Poor
P04 & P09	0.53	Moderate
P05 & P08	0.15	Poor
P06 & P07	0.40	Fair
P11 & P12	0.27	Fair
P13 & P18	0.24	Fair
P14 & P15	0.40	Fair
P16 & P17	0.52	Moderate

Example of calculation



		P1	P1			P1	P03
		0	1			0	0
P03	0	7	1	8	62%	0	0
P03	1	0	5	5	38%	0	0
		7	6	13		0	0
		54%	46%			1	1
						1	1
		Pr(a)		92%		1	1
		Pr(e)		51%		0	0
		K		0.843373494		0	0
				Excellent		1	1
						1	1
						1	0
						0	0

Appendix 8. Transcript illustrating code

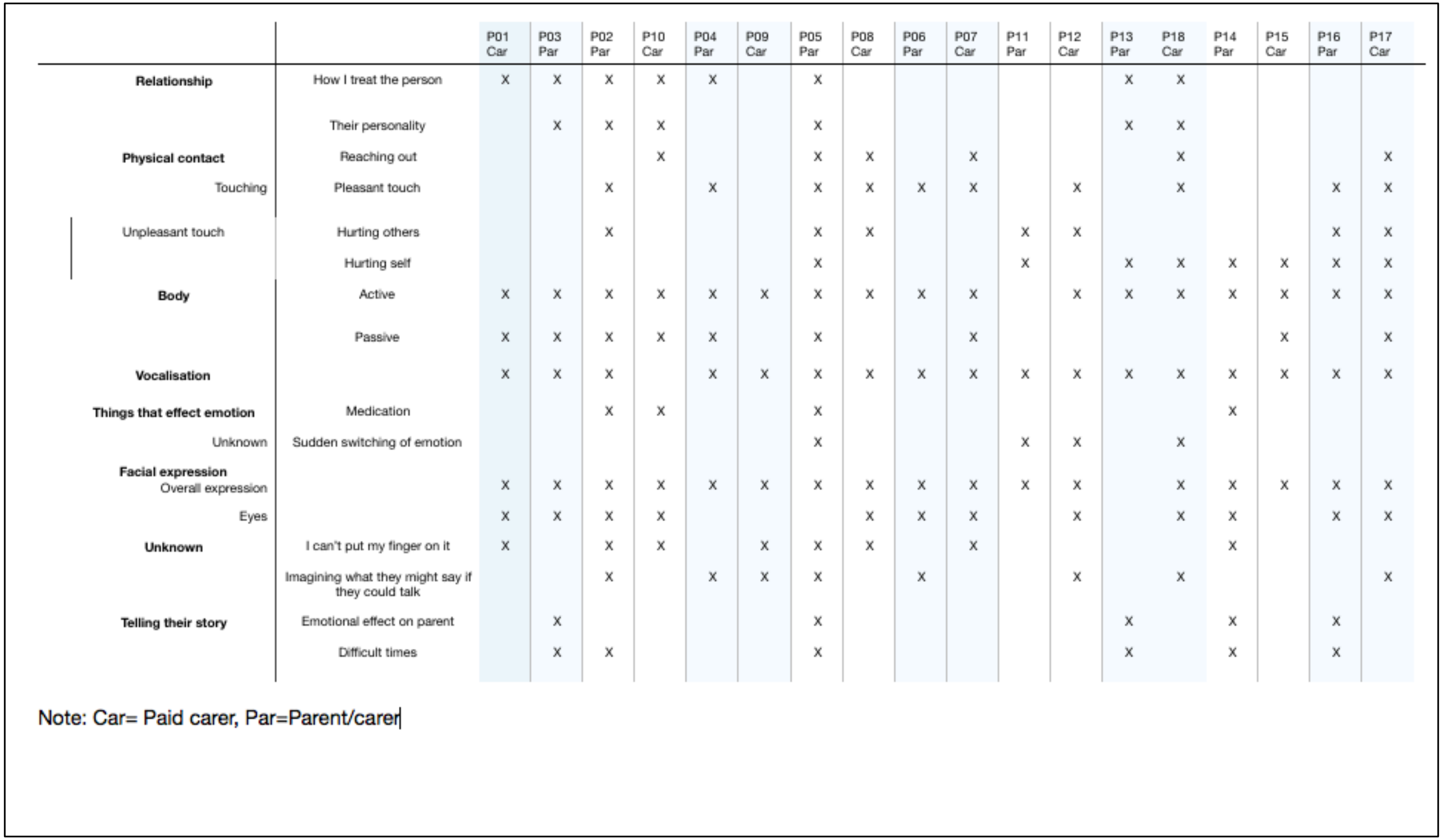


Level one code	Transcript	Level two code	Level three code	Level four code
	I: How do you feel about your relationship with your son, how well do you feel you connect with each other?			
Relationship	P:Um, I think we are very emotionally close. He likes love, you know, he likes to be cuddled. In lots of ways he's always been my little boy. For a start, he's the youngest, and then him being poorly, and um, we've always sort of cuddled and that, because its, I suppose, the only form of communication we really have in that sense. (...) He's got a wicked sense of humour. Wicked sense of humour!	How I treat the person	Their personality	
	I: How does he communicate to you when's he's feeling distressed, how does he communicate that distress to you?			
Facial expression Vocalisation Body Physical contact	P: You can see by his face..he pulls faces you know. Um, I know we've got..its something..you know.. he'll have a pent lip if somethings bothered him, you know, he'll put his lip out, and he'll sort of sneer with his nose if someone's said something that he's not fond of. And then there's other times when he'll have a full blown meltdown. He used to do all the time. You know, thrown himself up and down, scream and shout, pull his hair, bite himself, bite other people, you know?	Overall expression Calling out Active Touch	Pulling/screwing up face Throwing Shouting Screaming Unpleasant touch	Hurting others Hurting self

Level one	Transcript	Level two code	Level three code	Level four code
	I: How can you tell that he likes listening to people and music?			
Facial expression Physical contact	P: Well, he's altered now, so its more difficult, but he used to..in fact...it is.. when I go see him on a Saturday he will smile and he'll reach out, you know. When we come in, and when staff come in and change shift, you know, he will hear the doors open like that.. and somebody will talk to him and his face will light up and he will reach- he reaches out if he...um..if he doesn't like you he ignores you, ha!	Overall expression Reaching out	Smiling/grinning	
Body	You know, that's what he'll do, he'll just... or he'll turn away. [...] I remember one afternoon that I went and we'd take him out for a meal and they got this new walker that they wanted him to try that they were coming to fetch and so we didn't go out and he wouldn't cooperate with that and he went ape-shit. And then he laid on his bed and he looked at the wall and ignored me for 4 hours. Completely ignored me and I laid on the bed next to him and he just would not...you know...tried to put my arm and he just would not...	Passive	Refusing to cooperate or ignoring	
Unknown	it was..you know... "you didn't take me out- I hate you!", oh it was awful..Ha!	Imagining what they might say if they could talk		
	I: Do you find that you do that? That you interpret the way he's acting and thinking?			
Unknown	P: Yes. I mean the thing is, you're guessing, so you don't know if you're getting it right. But I do think people do make..you do make assumptions. I think it's the only... I think you've got to in a way ,	Guessing		
Telling their story	because otherwise you- I know when he was living at home and after the kids, his brothers were at work, and it was just me and him most of the time...um... I got to the point where I was thinking things. It	Emotional effect on parent		

Telling their story	took me a long time to start communicating with other people because I got to the point where I would think things instead of saying them because there was no feedback…no verbal feedback, and I got to the point with other people where I'd think I'd said something and I hadn't…I'd only thought it. Um…and when once a week when the milkman came to get his money, poor bugger, he only wanted to get paid and go on and I'd collar him for the sound of another human voice for half an hour on the doorstep, just… you know…"and another thing.."	Emotional effect on parent		
	I: Is there anything that occurred to you while we've been talking that you think is important for me to know?			
Unknown Things that affect their emotion	P: I think you've got to ..it takes a lot of effort to communicate with I'm and with people like that. You've got to be very observant because lots off…very similar gestures and very similar sounds mean different things. And..um, youre never sure whether you've got it right. That is the one thing that I think communicating emotions or anything..unless I mean X communicated his emotions when he wasn't on medication quite well, because he had such extreme…such extremes! Whereas now he's got the middle. I'd say if anything, it's harder..to distinguish the graduations because they're quite minute and if um.. you've got to be sort of next to him or you could miss quite a lot.	Guessing Medication		

Appendix 9. Distribution of participants across themes



		P01 Car	P03 Par	P02 Par	P10 Car	P04 Par	P09 Car	P05 Par	P08 Car	P06 Par	P07 Car	P11 Par	P12 Car	P13 Par	P18 Car	P14 Par	P15 Car	P16 Par	P17 Car
Relationship	How I treat the person	X	X	X	X	X		X						X	X				
	Their personality		X	X	X			X						X	X				
Physical contact	Reaching out				X			X	X		X				X				X
Touching	Pleasant touch			X		X		X	X	X	X		X		X			X	X
Unpleasant touch	Hurting others			X				X	X			X	X					X	X
	Hurting self							X				X		X	X	X	X	X	X
Body	Active	X	X	X	X	X	X	X	X	X	X		X	X	X	X	X	X	X
	Passive	X	X	X	X	X		X			X						X		X
Vocalisation		X	X	X		X	X	X	X	X	X	X	X	X	X	X	X	X	X
Things that effect emotion	Medication			X	X			X								X			
Unknown	Sudden switching of emotion							X				X	X		X				
Facial expression Overall expression		X	X	X	X	X	X	X	X	X	X	X	X		X	X	X	X	X
Eyes		X	X	X	X				X	X	X		X		X	X		X	X
Unknown	I can't put my finger on it	X		X	X		X	X	X		X					X			
	Imagining what they might say if they could talk			X		X	X	X		X			X		X				X
Telling their story	Emotional effect on parent		X					X						X		X		X	
	Difficult times		X	X				X						X		X		X	

Note: Car= Paid carer, Par=Parent/carer

Initial Template (01/02/18)

1. <u>Relationship</u>
    - 1.1 Understanding
    - 1.2 Warmth
    - 1.3 Connection
    - 1.4 Rapport
2. <u>Physical contact</u>
    - 2.1 Stroking
    - 2.2 Patting
    - 2.3 Pulling/grabbing
    - 2.4 Hitting
3. <u>Body</u>
    - 3.1 Turning away/towards
    - 3.2 Gesturing
    - 3.3 Tensing
    - 3.4 Relaxed
    - 3.5 Rocking
4. <u>Vocalisations</u>
    - 4.1 Screaming
    - 4.2 Singing
    - 4.3 Crying
    - 4.4 Shouting
    - 4.5 Laughing
    - 4.6 Moaning/whining
5. <u>Facial expression</u>
    - 5.1 Eyebrowns
    - 5.2 Eye gaze
    - 5.3 Grimacing
    - 5.4 Smiling

First template revisions (02/03/18)

1. <u>Relationship</u>

- 1.1 Understanding
- 1.2 Respect
- 1.3 Connection
- 1.4 “reading them”

2. <u>Physical contact</u>
    - 2.1 Touching
    - 2.2 Reaching out
    - 2.3 Kissing
    - 2.4 Hurting
    - 2.5 Self-harm
        - 2.5.1 scratches
        - 2.5.2 pulls hair
        - 2.5.3 bites
        - 2.5.4 throws self on floor
3. <u>Body</u>
    - 3.1 Turning towards/away
    - 3.2 Refusing to cooperate/ignoring
    - 3.3 Waving arms
    - 3.4 Tensing
    - 3.5 Exaggerated movements
4. <u>Vocalisations</u>
    - 4.1 Screaming/squealing
    - 4.2 Moaning/whining
    - 4.3 Laughing
    - 4.4 Singing
    - 4.5 Crying
    - 4.6 Lack of vocalisation/being quiet
5. <u>Facial expressions</u>
    - 5.1 Face lights up
    - 5.2 Gnashing teeth
    - 5.3 Pulls face

                - 5.3.1 grimacing
            - 5.4 Eye gaze
                - 5.4.1 You can tell by the eyes
                - 5.4.2 Eye contact
        6. <u>Guessing</u>
            - 6.1 I can't put my finger on it
            - 6.2 I get vibes
            - 6.3 Imagining what they might say

Second template revisions (15/03/18)

1. <u>Relationship</u>
    - 1.1 Understanding
    - 1.2 Respect
    - 1.3 Personality
2. <u>Physical contact</u>
    - 2.1 Touching
    - 2.2 Reaching out
    - 2.3 Kissing
    - 2.4 Hurting others
    - 2.5 Self harm
3. <u>Body</u>
    - 2.6 Turning towards/away
    - 2.7 Refusing to cooperate/ignoring
    - 3.2 Active movements
4. <u>Vocalisations</u>
    - 4.1 Screaming/squealing
    - 4.2 Moaning/whining
    - 4.3 Crying
    - 4.4 Lack of vocalisation/being quiet
    - 4.5 Happy vocalisations
5. <u>Facial expressions</u>

    5.1 Face lights up
    5.2 Pulls face
    5.3 Eye gaze
        5.3.1 You can tell by the eyes
        5.3.2 Eye contact

6 <u>Guessing</u>
    6.1 I can't put my finger on it/ unknown
    6.2 Imagining what they might say

7 <u>Emotions of carer</u>
    7.1 Difficult times

Third template revisions 18/03/18

1. <u>Relationship</u>
    1.1 How I treat the person
    1.2 Their personality

2<u>. Physical contact</u>
    2.1 Reaching out
        2.1.1 Hurting others
        2.1.2 Hurting self

3. <u>Body</u>
    3.1 Active
    3.2 Passive

5. <u>Medication</u>
    5.2 Sudden switching of emotion

6. <u>Facial expression</u>
    6.1 Overall expression
    6.2 Eyes

7. <u>Unknown- I don't know how I know</u>
    7.1 I can't put my finger on it
    7.2 Imagining what they might say if they could talk

8. <u>Telling their story (family participants only)</u>
    8.1 Difficult times

        8.2 emotional effect on parent

Fourth template revision (24/03/18)

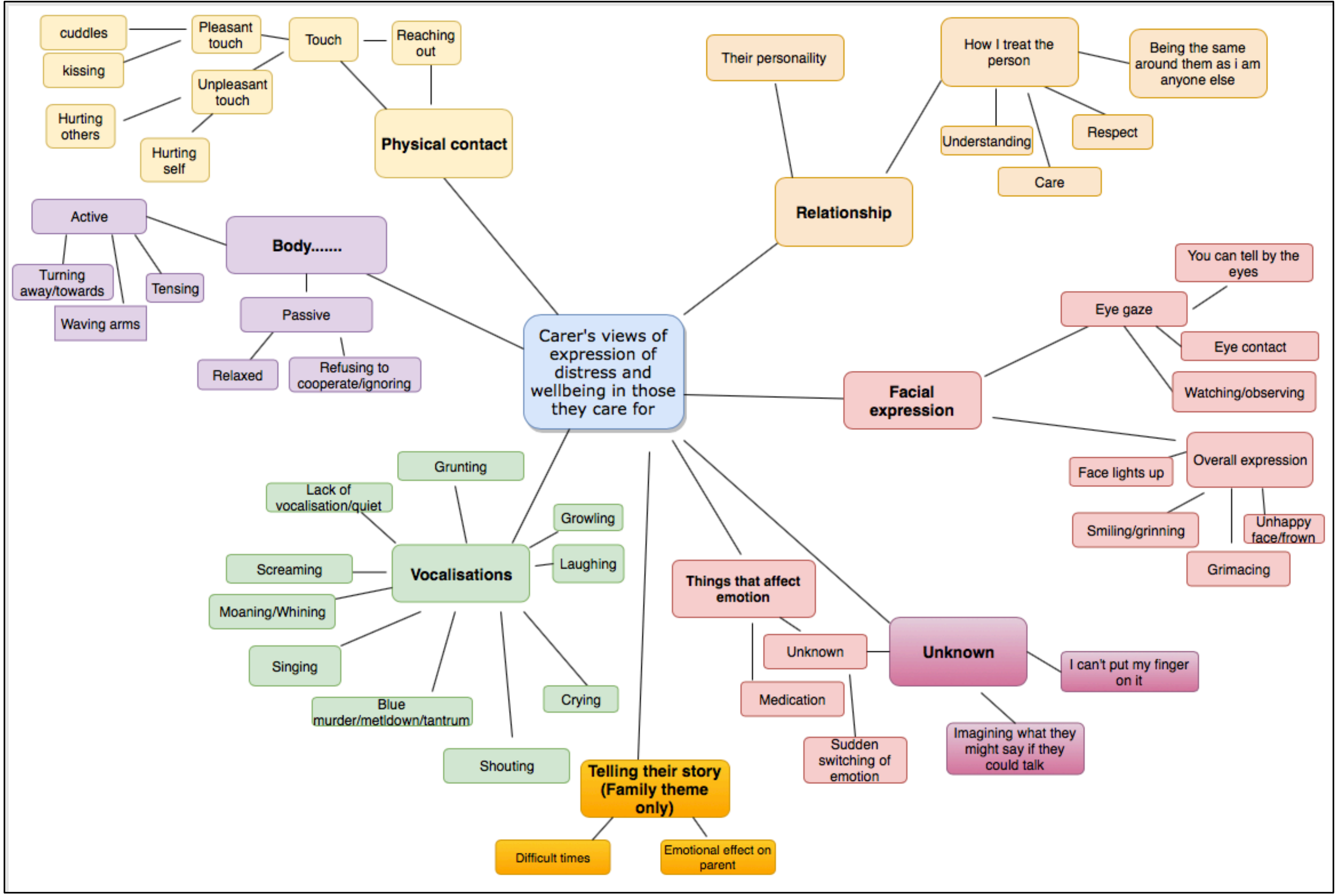
1. <u>Relationship</u>
    - 1.1 How I treat the person
    - 1.2 Their personality
2. <u>Physical contact</u>
    - 2.1 Reaching out
    - 2.2 Touch
        - 2.2.1 Pleasant touch
        - 2.2.2 Unpleasant touch
            - 2.2.2.1 Hurting others
            - 2.2.2.2 Hurting self
3. <u>Body</u>
    - 3.1 Active
    - 3.2 Passive
4. <u>Vocalisation</u>
5. <u>Things that affect emotion</u>
    - 5.1 Medication
    - 5.2 Unknown
        - 5.3.1 Sudden switching of emotion

6. <u>Facial expression</u>
    - 6.1 Overall expression
    - 6.2 Eyes

7. <u>Unknown- I don’t know how I know</u>
    - 7.1 I can’t put my finger on it
    - 7.2 Imagining what they might say if they could talk

8. <u>Telling their story</u>
    - 8.1 Difficult times
    - 8.2 Emotional effect on parent

Appendix 11. Conceptual map of final template



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