Stigma Experienced by the Families of Individuals with Intellectual Disabilities

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Thesis declaration form

I confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

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Date: 24th October 2017

Overview

This thesis is presented in three parts, with an overall focus on the stigma experienced by family members of individuals with intellectual disabilities (ID). This phenomenon warrants further exploration because families not only face the often challenging task of managing the needs of the person with ID, but they also have to cope with the emotional demands of caregiving, including the accompanying stigma of being undervalued by others.

The first part presents a review of literature on courtesy and affiliate stigma experienced by family carers of individuals with ID. It summarises evidence which shows that carers experience stigma and its consequences, and concludes that our understanding of courtesy and affiliate stigma is limited by a lack of clarity in distinguishing these concepts.

In the second part, an empirical paper is presented, documenting a quantitative study of stigma experienced by family members of persons with ID in the UK. The study focused on the development of a psychometrically sound new measure of family stigma and proceeded to examine the relationships between family stigma and various sociodemographic and contextual/psychological variables. The findings indicate that carers perceive family stigma before potentially internalising it, and that they face emotional and behavioural consequences due to its internalisation. These processes are influenced by a number of sociodemographic and contextual variables. The study suggested that family carers may benefit from more support and intervention strategies which target family stigma in order to reduce caregiving burden and improve subjective wellbeing.

Part three presents a critical appraisal of the study. The rationale for this research and the process of measure development are discussed. This is followed by an exploration of themes that emerged from qualitative feedback provided by caregivers who took part in the study in part 2, and their implications for future research on family stigma. In closing, a conclusion and personal reflections are provided.

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Acknowledgements

To all Harry Potter fans, you would understand what I mean when I say that this entire research process has been like the journey to facing Voldemort. At times it indeed did feel like I was battling proverbial villains, with statistics frequently rearing its head like a Dementor and sucking all happiness out of me (but nevertheless so useful!). There are so many people who encouraged me along the way. I would like to thank my supervisor, Dr Katrina Scior (my very own Professor Dumbledore), for her invaluable guidance and enthusiasm throughout the research process. I will fondly remember sitting in her office, staring at her in awe and admiration as she suggested idea after idea and cited existing research as if she were the Hogwarts Library herself. I'm also thankful to my external supervisor, Dr Afia Ali, for her guidance and generous support, particularly during the conception and design of the work presented. She gave me direction as I lumbered through the mystifying Forbidden Forest of definitions, concepts and methodology. I am also very grateful to the many people who helped with recruitment and participated in this research. Their dedication to helping those with intellectual disabilities as well as their carers reinforced my belief in this project time and again. I would especially like to thank my family; Pops, Mum, Navin and Nikhail. Even miles away, your words mean the world to me and continue to inspire me. Not forgetting my friends, who put up with my whining and dragged me out occasionally to ensure I did not turn into a hermit. And above all, my Seeta Masi, for keeping me in check with her humour, encouragement and patience. Seets, you tolerated my crankiness, Thesis-induced squalor and occasional breakdowns with your selflessness and the tough love of McGonagall, even at times when I was downright insufferable. Thank you for being one of my pillars in this journey.

Part 1: Literature Review

Stigma experienced by families of individuals with intellectual disabilities: A Systematic Review

1. Abstract

Aims: Studies have investigated the experiences of courtesy stigma and affiliate stigma in family members of individuals with intellectual disabilities without a clear distinction between the two. This systematic literature review aimed to evaluate the findings of studies that examined the experiences of stigma in families of individuals with intellectual disabilities.

Method: A systematic search of PsycINFO and Scopus identified relevant articles published between 2012 and 2016.

Results: Ten articles pertaining to eight studies were identified. They revealed that family carers experienced stigma and various consequences related to these, with family culture influencing these experiences. There is a lack of clarity in distinguishing the concepts of courtesy stigma and affiliate stigma in family members.

Conclusion: A number of psychosocial variables have been associated with the development of courtesy stigma, affiliate stigma and their consequences. Family carers also reported a number of coping strategies and protective factors. Further research would help gain a better understanding of stigma in family members of individuals with intellectual disabilities.

2. Introduction

Individuals with intellectual disabilities (ID) are one of the most ostracised groups and face stigma, prejudice and significant obstacles that restrict their human rights (Kôvágó, 2005). Stereotypes, prejudice and discrimination result from misconceptions about the individual's attributes by the dominant cultural group, and perpetuated by biased social structures (Corrigan, 2000). Stigma has been conceptualised as a mark of social disgrace in which the target individual is discredited based on attributes such as ethnicity, mental health problems, disability or drug-use. It describes the effect of negative attitudes and behaviours on the target individual and often leads to negative psychological health for the individual (Goffman, 1963).

2.1 Forms of Stigma

Five forms of stigma have been identified in the literature. The first, public stigma, refers to attitudes held within society about members of stigmatised groups (Bos, Reeder & Stutterheim, 2013). The second type, institutional stigma, occurs when policies reduce the choice of the stigmatised person (Heflinger & Hinshaw, 2010). The third type, self-stigma, occurs when the stigmatised person becomes aware of *and* internalises public stigma (Bos et al., 2013). Typically, studies in the ID field have focused on self-stigma as experienced by the individual with ID. Often however, persons associated with the stigmatised person, particularly family members, are also subjected to stigmatisation. This fourth type of stigma has been referred to as family stigma (Phelan, Bromet & Link, 1998), courtesy stigma (Goffman, 1963) or associative stigma (Mehta & Farina, 1988). The fifth type, affiliate stigma, involves the process of internalisation of courtesy or associative stigma by the stigmatised individual's affiliates, most likely family members (Ali, Hassiotis, Strydom & King, 2012).

Although caregiving can often be a rewarding experience, the family caregivers of individuals with ID also face considerable responsibility and accompanying stress (Baxter, Cummins & Yiolitis, 2000). They have to cope with both the physical and emotional needs accompanying the affected individual's disability. In addition, the stigma of being undervalued by others and often continuous and/or repeated battles with services adversely affect their quality of life (Chou & Palley, 1998; Chou, Pu & Lee, 2009). Consequently, they may feel dejected and helpless about their association with the stigmatised individual. This may result in lowered self-esteem and impaired family relationships (Wahl & Harman, 1989). For instance, an individual with ID exhibiting challenging behaviour in public can be an extremely distressing experience for family caregivers and may result in increased discrimination from the public and feelings of perceived stigma, social isolation and powerlessness (Cantwell, Muldoon & Gallagher, 2015). Mothers of children with ID have been found to be especially susceptible to poor mental health, reporting more depressive symptoms, higher levels of malaise, depression and anxiety than do mothers of typically developing children (Andersson, 1993; Blacher & Mink, 2004). Affiliate stigma may contribute to the negative impact of being a parent of an individual with ID. As a means of coping with some of the effects of affiliate stigma, caregivers may react by withdrawing socially, or even distancing themselves from their relative with ID to avoid association (Mak & Cheung, 2008).

To date, there has only been one published systematic review by Ali et al. (2012) examining 20 studies on stigma in family members of individuals with ID. Most of these studies were qualitative and cross-sectional in design with small sample sizes and mainly focused on the mothers' views. The authors found that family carers, especially those in non-Western cultures, were treated negatively by the community as well as other family members, which led to lack of social support, isolation, stress, increased burden and an overall lower quality of like. Protective factors included social support and the use of disclosure and education.

Since the review, the experience of stigma in family members of individuals with ID is an area that has received growing attention, although research is still somewhat lacking (Werner & Shulman, 2013). The present systematic review set out to summarise the findings of research into courtesy and affiliate stigma in family members of people with ID carried out since Ali et al.'s review. In particular, this review will focus on whether the experience of stigma has changed for family members, and if so, the factors that influence the experience.

3. Aims and Objectives

The review aims to answer the following questions:

1. What is known about the experience of stigma associated with ID in family members of individuals with ID?
2. To what extent is there evidence that this results in the internalisation of stigma (i.e. affiliate stigma)?
3. What are the documented consequences of affiliate stigma?

4. Method

4.1 Search Strategy

Study reports published over the period March 2012 to July 2016 were identified by searching the electronic databases PsycINFO and Scopus. The terms "mental retardation, ''intellectual disability'', 'learning disability'', "autism", "autism spectrum disorder",

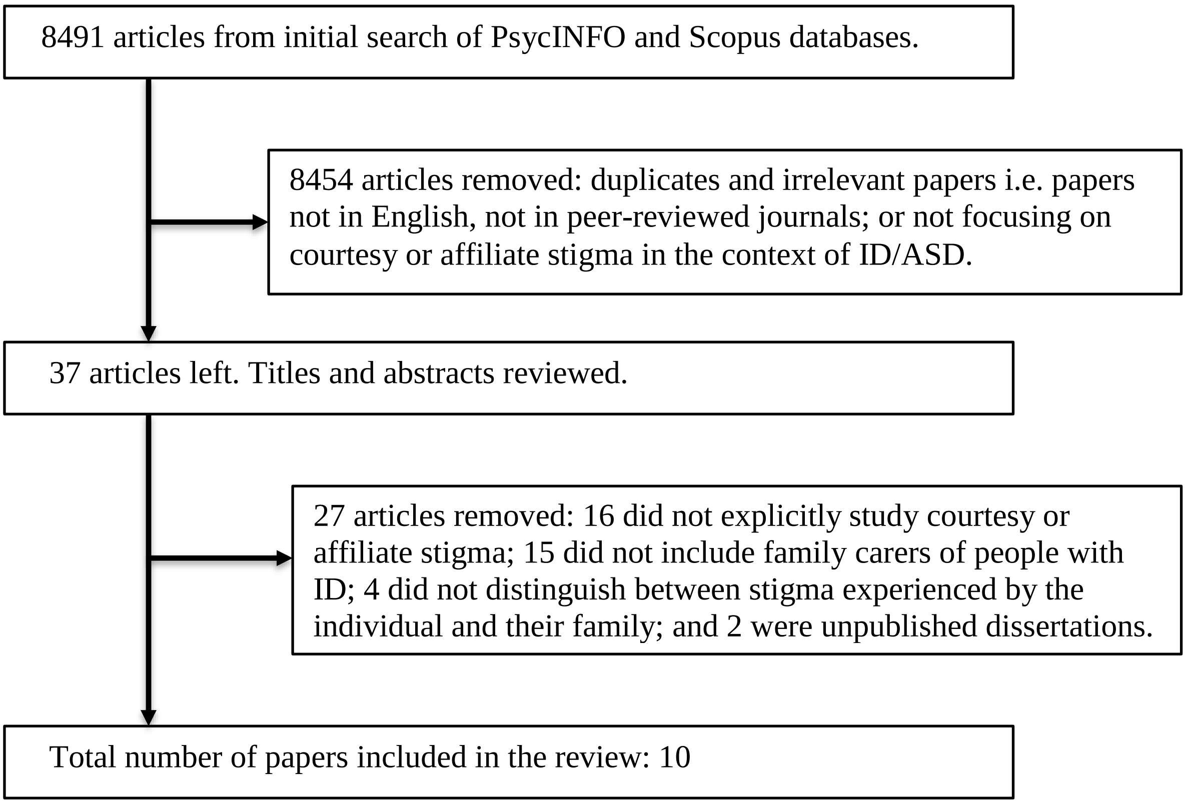

Figure 1. Flow diagram of search process.

"developmental disorder" and "developmental disability*" were combined separately (using AND as the Boolean operator) with the terms ‘‘stigma’’, ‘‘discrimination’’, "prejudice" and "attitude*". These terms were also combined with the search terms ‘‘carer’’, ‘‘family’’ and "relative". For the studies identified, titles of articles and abstracts were first screened and duplicates and those that were not relevant removed. The abstracts of the remaining studies were read and irrelevant ones were removed. Full papers were acquired for the remaining studies. These were read in full to identify whether they met the inclusion criteria. References of the included articles were also searched to identify further relevant articles not picked up in the database searches.

4.2 Inclusion and Exclusion Criteria

For an article to be included, it had to be a peer reviewed paper in English on a qualitative or quantitative study published between March 2012 and July 2016examining the experience of affiliate stigma in family members of people with ID or ASD. Family members included mothers, fathers, siblings and other relatives of the individual with ID or ASD.

Case studies, meta-analyses, and systematic reviews were excluded. Studies that did not specifically refer to individuals with ID, ASD in the absence of ID, or other developmental and neurological disorders were also excluded (e.g. epilepsy).

4.3 Quality Assessment and Analysis

A structured questionnaire developed by Hawker, Payne, Kerr, Hardey and Powell (2002) was used for quality assessment (see Appendix A). For each study, information was extracted about the design, sample size, selection of participants, type of measures or interviews utilised and their reliability and validity (if appropriate), the quality of reporting of the findings, their generalisability and any methodological limitations. Each paper was rated on these criteria and given a total score, with a possible score range of eight to 36 and higher scores indicating better quality.

5. Results of Search Strategy

A total of ten articles met the inclusion criteria and were included in this review. Table 1 provides an overview of these studies. The ten articles reported on eight different studies: two articles by Chiu, Yang, Wong, Li and Li (2013) and Chiu, Yang, Wong and Li (2015) reported on the same Chinese study and two articles by Werner and Shulman (2013, 2015) reported on the same Israeli study (see Table 1). Studies were conducted mainly in Asia and Israel, with only one paper from the UK and one from the African subcontinent. Studies focused on the experiences and internalisation of stigma, as well as factors associated with these, such as caregiver burden, social support and parental mental health. One study examined only courtesy stigma, four focused on affiliate stigma and three on both.

5.1 Methodological Considerations

Each study was evaluated in terms of its strengths and limitations using the aforementioned quality appraisal tool. Overall the studies were of a fair to good quality, see Table 2.

The total score of the studies ranged between 23 and 34 out of a possible 36. The strengths of the literature reviewed included thoroughness of presentation of results. In addition, most studies provided useful contributions to research on stigma in ID with important implications on policy and practice. However, these strengths were offset by some limitations. First, there was no mention on how the authors addressed the issues of sensitivity and researcher bias. Secondly, there were large variations in scores on the scale question pertaining to data analysis. While some studies, such as Werner and Shulman (2013) and Kwok, Leung and Wong (2014) gave a clear description of their analysis, others like Ngo, Shin, Nhan and Yang. (2012) and Chiu et al. (2015) provided less details. Furthermore, most studies used measures of courtesy and affiliate stigma that had not been validated in the respective cultural setting. However, despite these methodological considerations, overall the studies were rated fair to very good. Most studies received scores of three and four, suggesting that their findings were reliable.

Table 1

Overview of the studies examining courtesy or affiliate stigma in family members.

Study & Location	**Design**	**Sample**	**Method**	**Key Findings**	**Limitations**
Ngo et al. (2012); Vietnam	Mixed methods	70 parents (37 mothers, 33 fathers) of 37 children with ID who were concurrently being recruited for an early intervention programme in Hue City.	Developed Restriction of Social Life Scale to measure familial stigma, specifically limitations on social experiences of family members. Also assessed functional ability of child.	Cultural norms closely linked to stigma, which was positively associated with child's disability and with negative emotional reaction, and negative social life concerns and experiences.	Children with severe ID and/or challenging behaviour excluded. No standardised diagnostic system. Small size, use of non-validated measure.
Chiu et al. (2013); China	Cross-sectional	211 family members (163 parents, 46 other) of individuals with ID/ASD from registries of two urban cities.	*Affiliate Stigma Scale* (Mak & Cheung, 2008). Also measured face concern, mental health problems, anxiety level and empowerment.	Older caregivers showed higher level of behavioural affiliate stigma. Social resources and status likely to affect stigma. Positive association between face concern, stigma and mental health problems.	Overlooked stigma in grandparents and siblings.
Werner & Shulman (2013); Israel	Cross-sectional	170 parents (129 mothers, 13 fathers and 11 others) of individuals with ID, ASD and physical disabilities (PD).	19/22 items of *Affiliate Stigma Scale*. Also measured subjective wellbeing, positive meaning in caregiver, caregiving burden, self-esteem and social support.	Greater levels of affiliate stigma associated with lower ratings of subjective wellbeing in caregivers of ASD but not ID and PD. Relative contribution of affiliate stigma decreased when other fxs entered, especially self-esteem and social support.	Small sample. Difficult to distinguish between different disorders as some individuals had multiple primary diagnoses. Diagnosis based on caregivers' report.

Study & Location	**Design**	**Sample**	**Method**	**Key Findings**	**Limitations**
Kwok et al. (2014); Hong Kong	Cross-sectional	160 mothers of pre-school children with ASD and/or ID aged 2-6yo	Stigma measured using Devaluation of Consumer Families scale. Also measure caregiving burden and marital satisfaction (MS).	Negative associations between stigma and marital satisfaction. Burden mediates relationship between stigma and marital satisfaction. Mothers of ASD greater stigma than ID.	Data only from mothers.
Cantwell et al. (2015); Republic of Ireland	Cross-sectional	115 parents (92% female) of children with ID/ASD and 58 control typically developing control children	Caregiver stigma measured with 3 items adapted from Phelan et al. (2011). Also measured depressive symptoms, self-esteem, social support, child challenging behaviours and caregiver identification.	Self-esteem mediated the association between stigma and depressive symptoms. The path between stigma and depressive symptomology through self-esteem varies as a function of emotional support.	Measure of stigma very brief. Conflated depressive symptoms and self-esteem.
Chiu et al. (2015); China	Cross-sectional	211 family members (163 parents, 46 other) of individuals with ID/ASD from registries of two urban cities.	*Affiliate Stigma Scale* (Mak & Cheung, 2008). Also measured face concern. Also measured face concern, mental health problems, anxiety level and empowerment.	Face concern not as strong a determinant of mental health as compared to affective stigma. Mediator effect of affective stigma explained more variance in face concern when anxiety present.	Sample from urban areas but culture tends to be more diverse in rural areas. Unable to examine changes in stigma across age groups or disability types.
Werner & Shulman (2015); Israel	Cross-sectional	170 parents (129 mothers, 13 fathers and 11 others) of individuals with ID, ASD and physical	*Affiliate Stigma Scale*. Also collected demographic and background information.	Relatively low reported affiliate stigma compared to Hong Kong samples. Confirmed a one-factor solution of the *Affiliate Stigma Scale*. Significant differences in affiliate	Coping mechanisms not examined. Small convenience sample. More than one primary diagnosis for most and

Study & Location	Design	Sample	Method	Key Findings	Limitations
		disabilities (PD).		stigma according to diagnosis. Affiliate stigma not related to caregiver variables, but to child-related variables in ASD.	based on caregiver reports.
Yang (2015); China	Mixed methods	120 family members (102 parents, 18 others) of children with ID. Of these, 10 (5 mothers, 1 grandmother, 1 siste rand 3 fathers) participated in sequential interviews.	Affiliate Stigma Scale and semi-structured interviews. Also measured social face and caregiver mental health.	Low socioeconomic status group and individuals with ID that developed prenatally particularly vulnerable to stigma. Strategies for coping with affiliate stigma include social withdrawal, compensation/ overprotection, self-compassion and development of multiple identities.	Small sample and limited generalisability.
Tilahun et al. (2016); Ethiopia	Mixed methods	102 caregivers of children with ID from a hospital outpatient child mental health service.	Structured questionnaire for stigma, explanatory model of ID, type of intervention used or desired and coping strategies. Stigma also measured using the Family Interview Schedule (FIS). Also assessed caregiver's support needs.	Caregivers experienced courtesy and affiliate stigma. Those seeking help from traditional institutions experienced significantly higher levels of stigma.	Study facility-based so biased towards caregivers with higher education levels and ability to access specialist care. ASD sample small so limited power in comparing to ID. Possible social desirability bias.

Wong et al. (2016); Hong Kong	Cross-sectional	180 parents (149 mothers, 28 fathers, 3 missing data) of children with ASD through NGOs & special education schools.	*Affiliate Stigma Scale*. Also measured psychological distress, self-compassion, social support, professional support and positive parental perceptions.	Self-compassion moderated relationship between affiliate stigma and distress and was a powerful protective source above and beyond effect of social support.	Limited causality and generalisability.

Table 2
Component ratings of the quality assessment tool for the reviewed studies

Author(s) & date	**Methodological items (0-4)**									**Overall score (0-36)**
	Abstract & title (Q1)	Intro & aims (Q2)	Method & data (Q3)	Sampling (Q4)	Data Analysis (Q5)	Ethics & bias (Q6)	Findings & results (Q7)	Transferability/ generalisability(Q8)	Implications & usefulness (Q9)	
Ngo et al. (2012)	3	3	3	2	2	1	3	2	4	23
Chui et al. (2013)	4	3	4	3	4	2	4	3	3	30
Werner & Shulman (2013)	4	4	4	4	4	2	4	4	4	34
Kwok et al. (2014)	3	4	3	4	4	2	4	3	4	31
Chiu et al. (2015)	4	2	4	3	2	1	4	3	3	26
Yang (2015)	3	3	3	3	2	1	3	3	3	24
Werner & Shulman (2015)	4	3	3	3	4	2	4	4	3	30
Cantwell et al. (2015)	4	3	3	3	4	3	4	3	4	31
Tilahun et al. (2016)	4	2	2	3	3	4	3	3	4	28
Wong et al. (2016)	4	3	4	4	4	2	3	3	4	31

5.1.1 Study design. Four studies used the Affiliate Stigma Scale (Mak & Cheung, 2008) to measure affiliate stigma, whereas one each used the Devaluation of Consumer Families Scale (DCFS; Struening, Perlick, Link, Hellman, Herman, Sirey, 2001) and the Family Interview Schedule (FIS; Sartorius & Janca, 1996). One study used three items adapted based on questions from the Social Adjustment Scale (SAS II; Schooler. Hogarty & Weissman 1979), and one used the Restriction of Social Life Scale (Ngo, Shin, Nhan & Yang, 2012). In half of the studies, these scales were translated into the languages spoken by the respective samples. Most used measures of courtesy and affiliate stigma that had been modified or adapted but not been validated in the respective cultural setting or for family caregivers of people with ID or ASD. For example, the Family Interview Schedule (Tilahun, Hanlon, Abebaw, Tekola, Baheretibe & Hoekstra, 2016) was originally developed to measure experiences of stigma in the community among relatives of people with schizophrenia. The authors had adapted the tool for use in the study to focus on caregivers of children with developmental disorders. Similarly, the Devaluation of Consumer Families Scale (Kwok, Leung & Wong, 2014) was modified to measure the stress brought on by perceived stigma from having one or more children with disabilities and was then used without determining the psychometric properties of the modified version. Only Werner and Shulman (2013, 2015) examined the psychometric properties of their translated version of the Affiliate Stigma Scale, while the reliability and validity of the other measures within the respective study’s cultural context is uncertain. Most quantitative studies used t-tests to determine differences between groups (e.g. Kwok et al., 2014; Werner & Shulman, 2013). All the mixed methods studies used face-to-face interviews (Tilahun et al., 2016; Ngo et al., 2012; Yang, 2015).

The methodologies of the studies reviewed lacked robustness. The cross-sectional design of most of the studies limited any inference of causality and generalisability. Although the psychometric properties of the Affiliate Stigma Scale have been thoroughly examined in Asia and Israel, recent studies have questioned the unidimensional factor structure of the scale described by the original authors (Chang, Su, Tsai, Liu & Lin., 2015).

5.1.2 Sample characteristics and recruitment. Participants were recruited from various sections of the community and from different sources, including special schools (n = 6), non-governmental organisations (NGOs) focused on ID (n = 2), official registries (n = 1), and outpatient child mental health clinics (n = 2). Some recruited multiple populations. Out of the eight studies, only five included siblings and other relatives in their samples (Chiu, Yang, Wong, Li and Li, 2013, Chiu et al., 2015; Werner & Shulman, 2013, 2015; Yang, 2015); the remainder focused on parents, primarily mothers.

Random sampling was used in one study (Yang, 2015). One study employed cluster sampling by randomly selecting participants from three government funded special schools in a city in China. Another study used a mixed data source (Chiu et al., 2013, 2015) and the rest used convenience sampling, therefore the results are unlikely to be representative of the cultures or countries that were examined which limited generalisability.

The majority of participants in Hong Kong and Israel were recruited from special schools. According to a recent review of ID stigma globally (Scior et al., 2015), special schools in Hong Kong and Israel tend to cater for children with more severe ID and from more affluent families. It is possible that the experience of caregivers of children with mild to moderate ID and from lower socio-economic backgrounds could have been missed.

The studies reported on experiences of middle-aged family members between the mean ages of 35 and 43.15 caring for children whose mean ages ranged from two to 12.69 years of age. The experiences of young carers or those of adults with ID might differ. For instance, Sarkar (2010) reported that younger parents of adult children experienced stigma differently from older parents, particularly in terms of how stigma affected their interactions with other family members and their family’s quality of life. Furthermore, the individuals with ID in four studies were male; gender characteristics for the remaining four were not reported. The behavioural manifestations of ID for females have been found to be distinct from males, who tend to exhibit more abnormal behaviour patterns and externalising behaviour (Bell, Foster, & Mash, 2005; Maulik & Harbour, 2010).

5.2 Courtesy and Affiliate Stigma

The studies reviewed provided evidence for both the experience and internalisation of stigma. While all studies reported caregivers experiencing stigma based on various cognitive, emotional and behavioural responses, most studies did not differentiate between courtesy stigma and affiliate stigma. Three studies explicitly stated that they examined affiliate stigma (Chiu et al., 2013; Chiu et al., 2015; Werner & Shulman, 2013; Werner & Shulman, 2015; Wong, Mak & Liao, 2016). The other five studies examined some aspect of stigma, but used terms such as 'familial/family stigma' (Ngo et al., 2012; Tilahun et al., 2016), 'perceived stigma' (Cantwell et al., 2015, Kwok et al., 2014), and 'internalised self-stigma among family caregivers' (Yang, 2015) to describe both the experience as well as internalisation of stigma. As previously mentioned, for the purposes of this review, 'courtesy stigma' is used to describe findings related to the perception of stigmatisation due to the association with individuals with ID, while 'affiliate stigma' illustrates the internalisation of stigma. Findings from the studies reviewed will therefore be discussed in terms of the extent to which they advance our understanding of these two processes, regardless of the terms adopted in the original study.

In examining the psychometric properties of the scale, Werner and Shulman (2015) reported a lack of clarity between the items from the scale's cognitive, affective and behavioural components of affiliate stigma, with items from one component possibly representing another. For example, some of the items under the scale's cognitive dimension, such as 'my reputation is damaged because I have a child with a disability at home', arguably have an affective component to them. While Mak and Cheung (2008) suggested that the scale had a one-factor solution, a recent study employing principle component and Rasch analyses contradicted this notion, proposing instead that the affective, behavioural, and cognitive dimensions were unidimensional underlying constructs with three domains (Chang et al., 2015). The other studies used various different scales in order to measure courtesy and affiliate stigma. There was no common measure of stigma experiences and clear overlap between the two concepts, indicating inherent difficulties with the conceptualisation of courtesy and affiliate stigma in caregivers.

Relatively lower levels of stigma were reported in Israel (Werner & Shulman, 2015) compared to caregivers from Hong Kong, Taiwan and China, whereas in Vietnam caregivers experienced higher levels of stigma compared to China (Ngo et al., 2012). This could indicate that while some experiences, including self-blame and social withdrawal are universal, they might vary in different cultural settings, and therefore be different for individual caregivers. Indeed, of the studies reviewed, findings linking courtesy and affiliate stigma to cultural variables emerged more from low and middle income, collectivist countries (Chiu et al., 2013; Chiu et al., 2015; Ngo et al., 2012; Tilahun et al., 2016; Yang, 2015), where cultural beliefs are frequently subscribed to and interwoven into daily narratives and practices. In Vietnam, the endorsement of traditional Confucian values of karma and atonement for past sins is particularly relevant to stigma (Ngo et al., 2012). The studies from China predominantly focused on the traditional Chinese concept of ‘face concern’ (Chiu et al., 2013; Chiu et al., 2015; Yang, 2015), which refers to one’s sense of dignity in a social context, and the study from Ethiopia explored the role of religious beliefs (Tilahun et al., 2016).

5.3 The Experience of Courtesy Stigma

Based on the studies reviewed, family caregivers experienced courtesy stigma both within their communities and in public as well as within the family. Most of the studies appeared to describe courtesy stigma in relation to caregivers’ experiences of marginalisation by their communities and families. Two variables appear to be particularly relevant in this process of marginalisation.

5.3.1 The impact of culture on courtesy stigma. The initiation of courtesy stigma usually occurred when the child’s stigmatisation was “transferred” to carers through cultural mechanisms. For instance, caregivers in Ngo et al.’s (2012) study reported that their child with ID would never be able to wed or be employed in future. In Vietnam, the violation of cultural norms initiated the cognitive and behavioural processes of courtesy stigma, such as labelling and social withdrawal (Ngo et al., 2012). In China meanwhile, merely having a child with ID led to concerns about ‘saving face’ in the community (Chiu et al., 2013, 2015; Yang, 2015). People from Asian cultures may have a tendency to highlight the genetic origins of ID (Kung, 2001) and may be more inclined to considering children with disabilities as “bad seeds” who bring shame to their families (Sue & Zane, 1987). The child’s disability is seen as threatening cultural norms, for example, by the child viewed as being unable to fulfil social obligations such as marriage. Furthermore, traditional beliefs about karma and disability as some form of retribution for past sins compound the problem (Ngo et al., 2012). Given such cultural beliefs, family carers are implicated in the genesis of ID and may therefore be exposed to courtesy stigma.

Similar findings were reported in Ethiopia by Tilahun et al. (2016), who found that caregivers of the Orthodox Christian faith, and those who sought help from traditional sources and provided supernatural explanations for their child’s disability experienced more stigma. Research in African communities has indicated that conditions such as mental illnesses and disability are perceived as a curse from God (Ababa, 2014) or the result of other supernatural causes including witchcraft and angry ancestral spirits (Amuyunzu-Nyamongo, 2013). Members of the community may also consider the ID of the individual as culturally unacceptable, thereby subjecting the affected individual to stigma and extending this to the family as well. Moreover, indigenous medical traditions and healing rituals are common practices in the Ethiopian Orthodox Church (Anderson, 2007). Seeking help from such traditional sources could possibly result in further stigmatisation of the family.

5.3.2 Challenging behaviour and courtesy stigma. Only one study (Cantwell et al., 2015) directly examined the association between any challenging behaviour of the individual with ID and courtesy stigma. Two other studies referred to challenging behaviour in the context of disability type and findings on such behaviours related to the Affiliate Stigma Scale (Kwok et al., 2014; Werner & Shulman, 2013; Werner & Shulman, 2015).

Cantwell, Muddon & Gallagher (2015) found no association between child problem behaviours and courtesy stigma. This lack of association was explained by the authors as a result of the choice of measure of challenging behaviours, which were the conduct disorder and hyperactivity subscales of the Strengths and Difficulties Questionnaire (SDQ; Goodman, 2001). Although the psychometric properties of the original SDQ have been well researched (Goodman, 2001; Stone, 2010), it has been suggested that the extended version of the SDQ, which assesses the level of distress, and impact and level of difficulty presented by the child’s problems, might be a more valid tool for use with family carers of children with ID (Emerson, 2005).

Kwok et al. (2014) found that mothers of children with ASD reported greater levels of courtesy stigma than mothers of children with ID. Similarly, Werner & Shulman (2013, 2015) reported stigma to be higher in caregivers of individuals with ASD as compared to ID and physical disabilities. Moreover, caregivers of children with ASD were found to rate the item “The behaviour of my child with a disability makes me feel embarrassed” higher than caregivers of the other groups (Werner & Shulman, 2015). In comparing individuals with ID to those with ID in addition to ASD, McCarthy et al. (2009) found that challenging behaviours were four times more likely in the latter group compared to the former. The disabilities of an individual with ASD may not be as visible as those of someone with ID, which may lead others to deem the problematic behaviour as the outcome of poor parenting (Gray, 2002; Kwok et al., 2014). As such, the carer may face discrimination by others and later come to internalise feelings of shame.

Kinnear, Link, Ballan, & Fischbach, (2015) developed and found support for a model testing social aspects of the experience of stigma for parents of children with ASD. According to this model, the initiation of the stigma process was characterised by the child's ASD related behaviours, such as becoming visibly upset with a change in routine, or repetitive behaviours noticeable to others. The responses of others carried assumptions about poor parenting and about the capabilities of the individual with challenging behaviours, which increased the likelihood of social exclusion. The studies reviewed show support for this conceptual model, adding a dimension of culture that underlies this experience.

5.4 The Experience of Affiliate Stigma

Through repeated exposure to courtesy stigma, some family carers may eventually internalise stigma based on the subjective evaluation of these experiences (Chiu et al., 2013). Numerous psychosocial mediating and moderating variables have been linked to this internalisation process and the reviewed studies have mainly examined their effects.

5.4.1 Sociodemographic variables and affiliate stigma. All of the studies collected sociodemographic information from family carers. Their role in the process of internalisation was examined by six studies (Cantwell et al., 2015; Chiu et al., 2013; Chiu et al., 2015; Ngo et al., 2012; Tilahun et al., 2016; Werner & Shulman, 2015; Yang, 2015). Of these, two studies reported no association between affiliate stigma and caregiver sociodemographic variables (Werner & Shulman, 2015; Cantwell et al., 2015).

Socioeconomic status. Three of the articles reviewed examined affiliate stigma in relation to socioeconomic status (Chiu et al., 2013; Chiu et al., 2015; Ngo et al., 2012;

Yang, 2015). In line with some findings from the West, Ngo et al. (2012) found that socioeconomic status was positively related to social exclusion. Qualitative interviews with carers led the authors to conclude that this group perceived and internalised more stigma than those of lower socioeconomic status, consequently placing restrictions on their social lives. In the context of mental illness, Phelan, Bromet and Link (1998) suggested two explanations for greater concern with stigma among families of higher status; the first is related to the anxiety of the family’s reputation being damaged/diminished as a consequence of having mental illness in the family, and the second is the awareness of the family’s alleged role in the aetiology of mental illness thus fearing blame from others. These explanations were supported by Yang’s (2015) findings; family caregivers of individuals whose ID was of prenatal aetiology were more uncertain of the causes of the ID, and were consequently more likely to blame themselves as they made personal attributions (e.g. bad genes) to the causes. This increased their susceptibility to stigma. On the other hand, the post-natal aetiology group tended to attribute the ID to external conditions (e.g. illness), protecting themselves from potential blame (Yang, 2015).

Chiu et al. (2013) described contrary findings, with family carers of higher socioeconomic status generally reporting lower level of face concern and affiliate stigma than carers from low and middle socioeconomic status. These carers had more resources to cope with the effects of stigma, reporting reduced psychological distress, less anxiety and increased personal empowerment. A possible explanation for differences in findings from China and Vietnam could be linked to the level of development of these countries. In terms of the Human Development Index, an indicator of life expectancy, education, and per capita income (Human Development Report, 2015), China ranked 90th while Vietnam ranked 116th in 2015. Therefore, it is likely that people in less developed Vietnam hold on to traditional beliefs more than people in China. A survey conducted in 2009 showed that religiosity is highest in poorest countries, with 33% of the 1000 Vietnamese people interviewed reporting religion to be important in their lives compared to 18% in China (Crabtree, 2010). Findings by Ngo et al. (2012) on education levels of Vietnamese family carers provide further evidence for this; those with higher education perceived less restriction on their social life when they experienced affiliate stigma. The authors suggested that better educated carers may not endorse traditional Vietnamese beliefs of karma and past sins which implicate carers in the aetiology of the child's ID, thus reducing effects of stigma on their social lives. Equally likely is that family carers from China could afford to engage with services and social support that mitigate the effects of affiliate stigma; this is supported by Chiu et al. (2013), whereby families of lower socioeconomic status were more vulnerable to affiliate stigma compared to those of higher socioeconomic status.

Carer age. While Werner and Shulman (2015) reported no significant associations between affiliate stigma and age, Chiu et al. (2013) found older caregivers, compared to younger ones, had higher levels of affiliate stigma on the behavioural component of the Affiliate Stigma Scale. These caregivers also experienced lower empowerment. The authors concluded that due to increasing emotional and behavioural problems of individuals with ID as they age, older caregivers have been subjected to the affective and cognitive components of affiliate stigma for a longer duration and are therefore more likely to internalise stigma (Chiu et al., 2013). This was supported by recent findings indicating that as the individual with ID becomes bigger and older, parents face an increased threat of being assaulted by the individual and to encounter difficulties coping (Mckenzie & McConkey, 2016).

It should be noted, however, that all of the reviewed studies sampled younger carers, ranging between the mean ages of 35 and 48. The mean age of carers in Chiu et al. (2013) was 47.49 years, with a standard deviation of 11.94 years. The internalisation of stigma may be different for older/elderly family carers. For example, Chou et al. (2009) and Sarkar (2010) reported a strong effect of stigma on the quality of life of carers who were aged 55 years and older of adults with ID. The lack of research with such carers is disconcerting, especially given that most individuals with ID live with their families for their entire lives (McConkey, 2005).

5.4.2 Face concern and affiliate stigma. Face concern was examined in two of the Chinese studies (Chiu et al., 2013, 2015; Yang 2015), most likely due to it being a traditional Chinese value. Chiu et al. (2013) found face concern to be positively related to affiliate stigma and its affective, cognitive and behavioural dimensions. Overall psychological health was also related to the affective dimension of affiliate stigma, face concern and the level of anxiety (Chiu et al., 2013). In a related article on the same study, Chiu et al. (2015) showed that face concern operated through the mediating effects of the affective dimension of affiliate stigma to affect one's mental health, specifically by producing anxiety in situations of real or anticipated stigma. This implies that having a child with ID results in increased fear of losing face when both the child and carer are in a stigmatising environment (Chiu et al., 2015).

Related to this, Yang (2015) showed that face concern was related to the dimensions of behavioural and affective affiliate stigma but not the cognitive component of affiliate stigma. Specifically, parents who were concerned about face had poorer mental health and were more likely to experience feelings of shame, self-blame and powerlessness. Shame resulting in negative self-evaluations can arguably give rise to the affect regulating strategy of self-blame (Balzarotti, Biassoni, Villani, Pruna & Velotti, 2016; Leidner, Sheikh & Ginges, 2012; Lutwak & Ferrari, 1997). This means that the three experiences of shame, self-blame and powerlessness have emotional aspects to them, indicating that the process of internalisation of stigma might be related more to unconscious emotional underpinnings rather than conscious cognitive processes. The findings on affective stigma operating through face concern indeed support the notion that affiliate stigma may affect one without conscious awareness, exhausting one's internal resources and making one feel powerless, especially for carers who experience anxiety in light of stigma and those more concerned with face (Chiu et al., 2015). Moreover, these studies collectively demonstrate that the internalisation of stigma does not happen in a silo; rather it occurs within a cultural context.

5.4.3 Self-esteem and social support as buffers. Two studies examined the positive effects of self-esteem on affiliate stigma (Cantwell et al., 2015; Werner & Shulman, 2013). The latter (Werner & Shulman, 2013) assessed the moderating effects of psychosocial protective factors on the association between affiliate stigma and subjective wellbeing. Self-esteem, social support, positive meaning in caregiving and affiliate stigma emerged as the strongest predictors of subjective wellbeing in caregivers. This indicates that positive psychosocial resources, including self-esteem and social support, were more important to carers' subjective wellbeing than the negative influence of internalised stigma. Self-esteem is therefore protective of subjective wellbeing against the impact of stigma.

In examining the influence of self-esteem and social support on the relationship between stigma and depressive symptomology in parents caring for children with ID, Cantwell et al. (2015) found an association between caregiver stigma social support, self-esteem, depressive symptomology. Further, the relationship between stigma and depressive symptoms was mediated by self-esteem; those with higher levels of stigma and low self-esteem reported more depressive symptoms. Additionally, emotional support moderated this pathway. This meant that while perceived emotional support had a protective effect against depression from caregiver stigma at low to moderate levels of self-esteem, high levels of self-esteem alone protected psychological health whether perceived emotional support was low or high (Cantwell et al., 2015). These studies highlight the importance of positive psychosocial variables, self-esteem in particular, as potential buffers against the internalisation of stigma.

5.5 The Consequences of Affiliate Stigma

The studies reviewed generally found that the internalisation of stigma affected various aspects of caregivers' lives. They have shown that family members can experience considerable emotional distress, burden of care and social isolation because of stigma and develop coping strategies in an attempt to manage these consequences.

Findings by Chiu et al. (2013) support the idea that affiliate stigma reinforces the initial internalisation of stigma, creating a cycle of escalating internalisation and negative consequences. In the study, behavioural stigma, measured by items on the Affiliate Stigma Scale related to the behavioural consequences of affiliate stigma, was higher for older caregivers. The authors suggested that the longer the time spent giving care, the longer the caregiver was exposed to the affective and cognitive components of stigma, and the more likely they were to endorse and internalise stigma.

5.5.1 Caregiver mental health. Four studies involving a total of 806 participants investigated the relationship between caregiver mental health and affiliate stigma, and found more mental health difficulties in carers who experienced higher levels of stigma (Cantwell et al., 2015; Chiu et al., 2013, 2015; Yang, 2015; Wong et al., 2016). These studies mostly looked at the relationship between affiliate stigma, mental health and associated psychological variables and found positive correlations between affiliate stigma and mental health problems. The definition of mental health included here were psychological distress and mental health problems such as anxiety and depressive symptomology. Two other studies considered negative emotional reactions that were experienced by caregivers as a result of stigma (Ngo et al., 2012; Tilahun et al., 2016). These included sadness, embarrassment, shame, guilt and fear.

Chiu and colleagues (2013) reported 60.6% of their sample of family caregivers experienced evident mental health issues that required further professional attention. Each dimension of affiliate stigma was positively associated with anxiety and depressive symptoms, which were lowest in carers of middle or higher socioeconomic status. Mental health had a stronger association with the affective component of affiliate stigma and was subsequently found to mediate the relationship between face concern and general mental health (Chiu et al., 2015), with the affective component explaining more variance in general mental health when anxiety was present. This indicated that affiliate stigma could unconsciously affect the carer by draining internal coping resources and making them feel helpless (Chiu et al., 2015). Similarly, Yang (2015) showed that parents with higher face concern were more likely to experience affiliate stigma, resulting in poorer mental health. Overall, these findings suggest that there is a cultural dimension to the way in which negative affect is experienced in affiliate stigma that has far-reaching effects on caregivers' mental health. Though affiliate stigma consists of cognitive and behavioural aspects as well, it may be these interactions between the affective component and cultural factors that lead to mental health problems in caregivers, particularly in collectivist cultures. In Chinese cultures, this could be face concern, where family members of individuals with ID may overlook their own mental wellbeing in order to avoid social situations where they may "lose face" (Chiu et al. 2013; Mak & Cheung, 2012). The effects of affiliate stigma may be more acute for family members in such cultures due to concerns about social disgrace that is brought onto the family due to the individual with ID and subsequent self-blame (Geva & Wiener, 2015). Guilt becomes a manifestation of the internalization of stigma for family caregivers (Fink & Tasman, 1992; Turnbull & Turnbull, 1979). Indeed, the caregivers in Yang's (2015) study reported powerlessness and self-blame that resulted from affiliate stigma to have the most adverse effect on mental health, and Tilahun et al. (2016) reported caregivers blaming themselves and feeling depressed and ashamed about their child's condition. Ngo et al. (2012) and Chiu et al. (2015) also highlighted other negative emotional reactions experienced by family carers due to anticipated stigma. Similar experiences have been reported by parents from other collectivist cultures. For instance, Narayan (2014) found that South Asian parents raising a child with ID felt stigmatised by their communities due to cultural beliefs and experienced negative emotions such as anger and depression. Such negative emotions associated with affiliate stigma may be the mechanisms by which the caregiver develops mental health difficulties.

Cantwell et al. (2015) reported 33% of caregivers in their study met criteria for depression, and that the relationship between stigma and depressive symptomology was stronger in caregivers of individuals with ID compared to their control group of typically developing children. Affiliate stigma was one of the psychosocial pathways influencing high depressive symptomology in carers, with self-esteem mediating this association. The path through self-esteem varied as a function of emotional support, which meant that for parents with higher self-esteem, the mediation of stigma and depressive symptoms was not dependent on emotional support. In the same vein, Wong, Mak & Liao (2016) found an association between affiliate stigma and psychological distress, with self-compassion moderating this relationship. These findings indicate the importance of internal as well as external coping resources in reducing the psychological impact of affiliate stigma. Chiu et al. (2013) found that family carers of higher socioeconomic status reported better mental health and less affiliate stigma. It was likely that such family carers were able to afford better social resources, allowing them to protect their social image and self-esteem from the ill-effects of affiliate stigma (Chiu et al., 2013).

5.5.2 Burden of Care. Two studies reported findings on the consequences of affiliate stigma in terms of caregiver burden (Kwok et al., 2014; Werner & Shulman, 2013). Kwok et al. (2014) found that mothers of children with ASD showed higher levels of stigma, higher levels of perceived burden, emotional burden in particular, and lower marital satisfaction than mothers of children with ID. Perceived burden mediated the negative association between stigma and marital satisfaction. Werner and Shulman (2013) similarly found affiliate stigma and burden to be higher among caregivers of individuals with ASD when compared with caregivers of individuals with ID or physical disabilities (PD). Affiliate stigma and burden were also negatively correlated with caregivers' subjective wellbeing, the subjective dimension of quality of life.

Werner and Shulman (2015) found that caregivers reported the highest level of affiliate stigma on items of the Affiliate Stigma Scale that reflected negative internalised emotions associated with the daily strains and challenges of having a child with developmental disabilities. Previous research has indicated that emotional burden was linked to anxiety and depression in mothers, which affected their overall quality of life (Zhang & Yi, 2011). Tackling the embarrassment caused by the child's misbehaviour in addition to the consequent stigma may result in emotional and psychological distress in carers (Green, 2003), leading to reduced overall quality of life. Family carers often have to care for the individual with ID under the pressure of such a discriminatory environment, which adds to their burden of caregiving. Moreover, Narayan (2014) found an increase in marital conflicts after the birth of a child with ID, resulting in significant problems in the relationship between couples. Therefore, it is possible that the negative feelings associated with caregiving burden could have transferred to the spousal relationship, lowering marital satisfaction.

5.5.3 The use of coping strategies. The four studies that evaluated coping strategies reported the use of both positive and negative strategies by family members to cope with the impact of affiliate stigma.

Maladaptive strategies. Four studies reported the use of maladaptive strategies by family members. Efforts to conceal the individual's condition were common, as was social withdrawal (Ngo et al., 2012; Tilahun et al., 2016; Werner & Shulman, 2015; Yang, 2015). This was in line with Ali et al.'s (2012) conclusion that maladaptive coping approaches appear widespread among family carers of people with ID in Asian populations.

Yang (2015) found the use of an additional negative strategy among carers termed compensation/overprotection, where the anticipation of stigma and associated feelings of self-blame led to overprotection of the individual with ID from potential harm. For example, parents may insist on being around and intervening when the child is trying to play with other children. Previous research has indicated that long-term use of such strategies by parents can lead to lowered self-esteem and further discrimination of the individual with ID (Sanders, 2006), which could paradoxically increase affiliate stigma.

Positive means of coping. Tilahun et al. (2016) reported more than half of their sample of carers (57.8%) found support in their religious beliefs and rituals by using prayer as a means of coping with the negative effects of affiliate stigma. Cinnirella and Loewenthal (1999) found that prayers offered family carers of individuals with mental illness an opportunity to turn to God to unburden their worries and to maintain self-efficacy and feelings of empowerment. The same study found that prayer was especially beneficial for Afro-Caribbean families as it helped family members deal with stigma by keeping the mental illness of the individual concealed from the community and extended family (Cinnirella & Lowenthal, 1999).

As reported by Yang (2015), another way family carers may manage the effects of affiliate stigma is through defining themselves in relation to multiple identities. Instead of defining themselves solely in relation to the individual with ID, carers adopted other affirmative social identities, including in terms of their career or their caregiving role. A recent study of affiliate stigma among caregivers of individuals with mental illness found that the greater the extent of internalisation of stigma by carers, the more they experienced an unstable social identity and became socially isolated (Farzand & Abidi, 2013). This could apply to family carers of individuals with ID; an unintended consequence of internalisation could be that their social identity becomes disturbed and stigmatised.

Studies have found that emphasising other identities may act as a buffer against psychological distress by compensating for the problematic identity and minimising its effects (Thoits, 1983; Perkins, Holburn Deaux, Flory & Vietze 2002). Moreover, previous research has shown that mothers of individuals with ID who volunteer as advocates and activists considered this an important aspect of their identity, and experienced less shame, earning them the respect of other family members (Chang, 2009). Establishing multiple identities and switching between these seemed to protect carers' self-esteem and psychological wellbeing from the stigmatised identity, thereby preventing them from internalising stigma further. In this way, they may be able to protect their self-concept by acquiring a sense of self-efficacy in the face of their achievements (Crocker & Major, 1989). The emphasising of multiple identities could therefore be an important coping strategy for family carers of individuals with ID.

5.5.4 Social and community support. Tilahun et al. (2016) found that while talking to health professionals, family and friends was the most common strategy employed among carers, increased stigma was associated with seeking help from traditional sources, including religious centres and traditional healers. Indeed, Croot (2012) showed that Pakistani family carers resorted to biological explanations for ID in order to decrease stigma that had developed from the community's traditional ideas about ID. This could provide an explanation for the finding that the sample of caregivers in Tilahun et al.'s (2016) study sought help from biomedical sources as a coping strategy.

In the same study (Tilahun et al., 2016), prayer was the second most common coping mechanism among carers. Previous research using qualitative interviews with family caregivers of individuals with mental illness found that the community stigma associated with mental illness led to an inclination for private coping strategies such as prayer, lending some support these findings (Cinnirella & Lowenthal, 1999).

While prayer is a more personal and private experience, there is a social element attached to seeking help from traditional sources, exposing the carer to the risk of further stigmatisation. In most countries in Africa, where religious ideologies dominate, the stigma attached to mental illness is often a result of family members denying the existence of the illness (Ravello, 2015). The caregivers in Tilahun et al.'s (2016) study indeed cited such reasons as spirit possession and punishment from God as the perceived causes of developmental disorder in their child, themes which are strongly embedded in religious beliefs.

5.5.5 Self-compassion. Self-compassion, defined as 'a caring and compassionate attitude toward oneself in the face of hardship or perceived inadequacy' (Wong et al., 2016, p.2), was examined as a unique coping strategy in two of the more recent Chinese studies (Yang, 2015; Wong et al., 2016). In response to affiliate stigma, Yang (2015) found that parents occasionally used self-compassion to regulate the emotional and cognitive anxiety arising as a result of internalisation. Similarly, Wong et al. (2016) found that while psychological distress was positively correlated with affiliate stigma, self-compassion was negatively correlated with both affiliate stigma and psychological distress. Even after controlling for the effects of social support, self-compassion moderated the association between affiliate stigma and distress. This association was significant at low levels but not at high levels of self-compassion, leading the authors to infer that self-compassion was a protective factor against affiliate stigma. Furthermore, self-compassion can serve as an internal coping resource without having the family carer rely on external sources of help.

Wong et al. (2016) also reported that self-compassion explained the additional variance in psychological distress beyond that explained by positive parental perception of caregiving. Self-compassion is therefore distinct from positive meaning in caregiving, which involves the cognitive process of meaning-making (Frankl, 1984) and has been found to mitigate the negative effects of affiliate stigma (Werner & Shulman, 2013).

Indeed, self-compassion has been described as an emotional regulation strategy that may allow parents to acknowledge and understand negative emotional reactions implicated in affiliate stigma (Wong, et al., 2016). This is an important finding as it indicates that aside from building good networks of social support, family carers may need to develop a repertoire of internal coping resources in order to address the different dimensions (i.e. cognitive, affective and behavioural) of affiliate stigma.

6. Discussion

6.1 Summary of Findings

This systematic review summarises the key findings from research on the experience and internalisation of stigma by family members of individuals with intellectual disabilities, also referred to as courtesy and affiliate stigma. As such, it provides a deeper understanding of the experience of stigma for family members involved in the care of an individual with ID.

Eight studies were reviewed, most of which were from Asia and Israel and used cross-sectional and mixed methods designs. Higher levels of stigma were reported in Chinese countries and Vietnam, and experiences of stigma varied across cultural settings. Various sociodemographic and contextual factors affected the experiences of stigma, including challenging behaviours of the individual with ID, socioeconomic status, carer's age and culture. Culture appeared to have an impact throughout the process of stigma, from its initiation to its eventual internalisation and the consequences of this internalisation. In order to minimise its effects, family carers adopted both positive coping strategies, such as developing self-compassion and multiple identities, as well as maladaptive ones, including social withdrawal and compensation/overprotection.

The conclusions drawn concur with the findings of Ali et al. (2012) to some extent. As in Ali et al.'s (2012) review, most family caregivers experienced courtesy and affiliate stigma, but its extent and form varied across cultures. This review went beyond Ali et al.'s conclusions by highlighting that experiences of courtesy stigma depend on carers' setting and the behaviours the individual with ID exhibits in others' presence. Repeated exposure to others' stigmatising attitudes and behaviours resulted in the internalisation of stigma. The extent of internalisation was contingent on factors associated with carers' backgrounds and internal coping resources. Consequently, as reported in Ali et al. (2012), affiliate stigma had an effect on carers' burden of care and mental health, leading to the use of positive as well as negative coping strategies.

The current review differed and extended findings from Ali et al. (2012) in several ways. The studies presented here employed mainly cross-sectional and mixed method designs with comparably larger sample sizes compared to the previous review. Though most of the studies in the current review showed the same problems of small sample size and limited generalisability, there is definitely growing interest in the field which could spur more large scale, representative studies. The key difference to Ali et al.'s (2012) review is that findings from the current review indicated a shift from the complete pathologisation of ID to an increased focus on positive meanings in caregiving. Furthermore, recent research has emphasised family members' positive coping resources that can be drawn on in countering the effects of stigma.

Overall however, the concept of stigma among caregivers still remains unclear. Moreover, the studies reviewed mainly focused on the emotional underpinnings of stigma experiences, while the cognitive and behavioural dimensions of courtesy and affiliate stigma remain under-researched. This review has attempted to capture and synthesise different conceptualisations of stigma and arrive at a more holistic understanding of the process of stigmatisation for caregivers, see Figure 2.

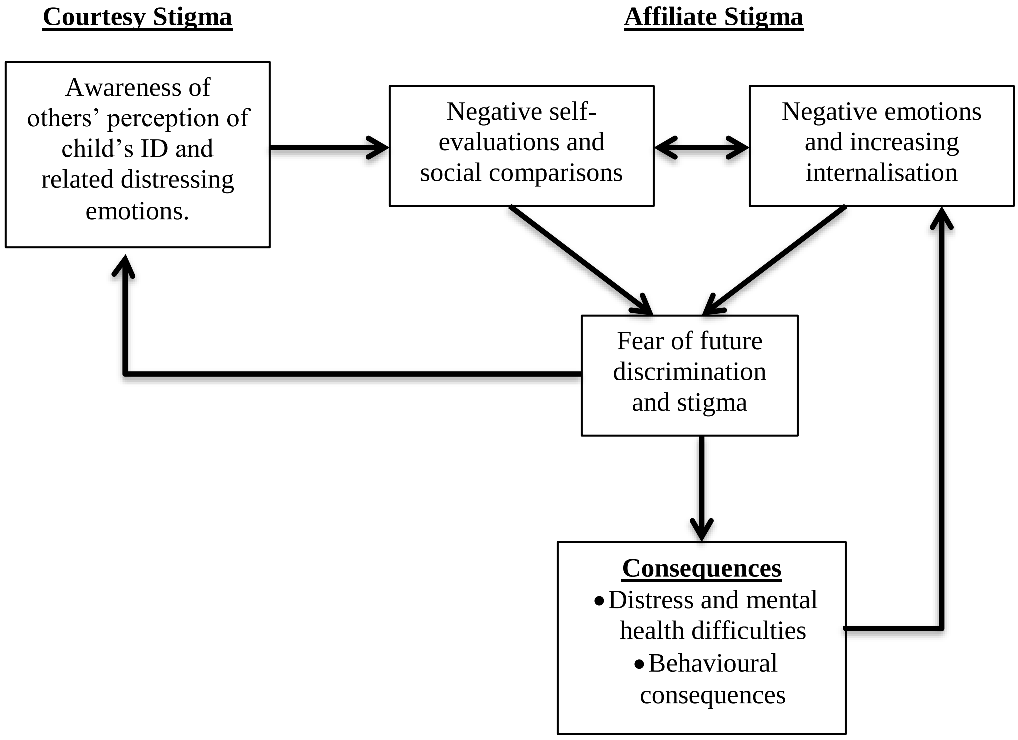

Figure 2. Synthesis of findings – The circle of stigma.

6.2 The Circle of Stigma

From Figure 2, we can see that courtesy stigma and related experiences precede the internalisation of negative evaluations and eventually lead to the development of affiliate stigma. As a consequence of this internalisation, carers may withdraw socially or conceal having a son/daughter with ID or ASD, which may in turn negatively influence access to social support, self-esteem and psychological wellbeing. Culture underlies this entire process.

A child with ID exhibiting problematic behaviour in public may attract the attention of others. Depending on the social and cultural context, an awareness of others' negative perceptions and responses may induce negative emotions for parents. Such repeated encounters may gradually be associated with increased feelings of courtesy stigma, and feelings of embarrassment and powerlessness (Cantwell et al., 2015).

Courtesy stigma may then prompt negative self-evaluations and negative social comparisons (Ali et al. 2012), and thus initiate the internalisation of stigma by caregivers.

Werner and Shulman (2015) found that the highest level of affiliate stigma was reported by carers who experienced negative internalised emotions associated with the daily challenges of having a child with developmental disabilities. Other studies in this review also reported on some of these emotional experiences of carers, namely shame, guilt and embarrassment (Chiu et al., 2013, 2015; Tilahun et al., 2016; Yang, 2015). Emotions have been theorised to be a response to cognitive activity, specifically that it involves a primarily unconscious process that results in the evaluation of the impact of an occurrence for one's own welfare (Lazarus, 1991; Sylwester, 2001). The pathway to internalisation itself could be through emotions and therefore unconscious. Indeed, this is reflected in findings in the previously mentioned study by Chiu et al. (2015) where the authors found an association between anxiety, face concern and the affective component of affiliate stigma in particular.

The degree of this internalisation is contingent on the some of aforementioned psychosocial variables, such as the socioeconomic status, culture and psychological health of the caregiver. With increasing affiliate stigma, family caregivers may come to fear future discrimination and stigma (Chiu et al. 2015; Ngo et al. 2012; Yang, 2015), resulting in more negative emotional reactions and poorer mental health. In order to cope with these, family carers may resort to such maladaptive coping strategies as concealment and social exclusion, which then reinforce affiliate stigma.

The findings of this review lend some support to the notion that phases of courtesy stigma, affiliate stigma and its consequences are not discrete events. The feedback between them ultimately creates a vicious circle of increasing levels of courtesy stigma and affiliate stigma. Therefore, the same way the negative consequences of affiliate stigma reinforce its initial internalisation, positive coping strategies and internal resources may lessen the degree of affiliate stigma and foster more positive emotions. However, some carers caught in the spiral of stigma might not be able to seek help, gradually isolating and excluding themselves from the community.

6.3 The Role of Culture

Findings from this review highlight the importance of the cultural context in the processes of courtesy stigma and affiliate stigma for caregivers. The Chinese concept of face concern is useful in illustrating how culture plays a role in the stigma circle presented in Figure 2. Due to concerns about saving face, Chinese family caregivers are more likely to be susceptible to courtesy stigma and the accompanying powerful feelings of shame, and anticipation of stigma through social contamination. This preoccupation with face may lead them to develop psychological difficulties for which they are reluctant be seek help due to more worries involving face (Chiu et al., 2013, 2015; Yang, 2015).

Previous research has found that collectivist cultures are more likely to stigmatise those who deviate from social norms as compared to individualist cultures (Papadopoulos, Foster & Caldwell, 2013). The role of culture in stigma could therefore be especially pertinent in collectivist cultures, where having a child with ID is in itself stigmatising (Chiu et al., 2013). In Western cultures, where individualistic beliefs dominate, other variables, such as child problematic behaviours or the type of ID may have a larger influence on the process. This was reflected in Ngo et al.’s (2012) study, whereby parents with higher education levels who are less likely to prescribe to traditional cultural beliefs were less likely to experience stigma. Indeed, while Cantwell et al. (2015) reported 33% of the sample of family caregivers in their study met criteria for depression, Chiu et al. (2013) found that 60.6% of carers in their sample experienced mental health difficulties. The former study was conducted in the UK, the latter in China, a collectivist country.

6.4 Implications

The findings from this review have a number of implications for the provision of interventions and support for family members caring for an individual with ID. Stigma has far-reaching effects on multiple aspects of their lives, therefore it is necessary that interventions that seek to reduce stigma are designed specifically for them, taking into account the context of culture.

At the level of the individual, family members should be encouraged to develop internal coping resources such as self-compassion and establishing or emphasising/valuing other social identities to bolster their self-esteem (Wong et al., 2015; Yang, 2015). Greater self-esteem in parents of children with disabilities has been found to be predictive of resilience and better psychological health, acting as a coping mechanism that alleviates the impact of stress on psychological health (Bekhet, Johnson & Zauszniewski, 2012; Marcussen, Ritter & Safron, 2004; Raina et al. 2005). This is especially important in highly stigmatising communities where families may not be able to depend on others for support. Additionally, regardless of their cognitive self-perceptions, experiences that impinge carers' cultural beliefs promptly evoke emotional responses (Yang, 2015). Culturally sensitive interventions that enhance coping skills to self-stigmatised feelings may thus be more effective than attempting to change stigmatising cognitions (Mittal, Sullivan, Chekuri, Allee & Corrigan, 2012).

At the familial level, consideration should be given to the primary caregiver in order to reduce caregiving burden, which was found to be a chief contributor of affiliate stigma (Werner & Shulman, 2015). The sharing of household tasks could be encouraged to facilitate time and energy for self-care, as well as enable the caregiver to establish and retain a social network. Having open and regular communication within families can also reduce feelings of powerlessness and isolation (Yang, 2015).

Based on the findings on this review and previous research, affiliate stigma is less likely to occur if courtesy stigma is minimised (Papadopoulos, 2016). This is an important consideration when implementing strategies at the community and societal level. First, improving public awareness of developmental disabilities and challenging related stigmatising beliefs could foster more positive community attitudes which may in turn reduce affiliate stigma. Secondly, collaborations with traditional sources of support such as religious institutions and faith healers may be necessary for public awareness and stigma tackling efforts. Thirdly, as noted above, families from lower socioeconomic backgrounds may encounter difficulties in seeking support due to financial constraints and lack of knowledge about professional services. Outreach programmes that target rural communities where families with lower socioeconomic status tend to live may be useful to provide support for them.

Lastly, given that affiliate stigma can affect one without conscious awareness (Chiu et al., 2015), the identification of parents susceptible to it is critical. The ability of professionals to recognise risk factors associated with affiliate stigma, such as significant child behaviour problems, low self-esteem and lack of social support should be improved through staff training and regular contact with families. Furthermore, offering culturally sensitive counselling and psychotherapy services to families deemed at risk of affiliate stigma and establishing support groups for them to provide emotional support through the exchange of shared experiences and reflections could serve as important early intervention strategies.

6.5 Limitations

Limitations of this review have to be acknowledged. Relatively narrow search parameters were applied. Specifically, using other potential sources of information such as dissertations or unpublished and non-English publications would possibly uncover other relevant literature. In addition, the review only focused on stigma-related experiences of relatives of individuals with ID and ASD in the context of ID. The experiences of family members caring for individuals with other types of developmental disabilities were not included and might differ. Finally, only two databases were searched. Using additional databases may have yielded other types of studies.

7. Potential Areas for Future Research

The findings of the review identify shortcomings in the existing literature, and several questions still remain. First, future studies need to differentiate between the different components of courtesy and affiliate stigma in order to establish an accurate representation of the multidimensional theoretical construct. The ways in which various psychosocial variables relate to these different components may also be useful in this regard, and could additionally serve to elucidate the underlying mechanisms of courtesy and affiliate stigma. As with other areas of research in ID in the field, more accurate measures need to be developed for this, especially if we want to test the outcomes of interventions for family caregivers.

Secondly, a majority of the studies reviewed focused on the internalised emotional basis of stigma experienced by caregivers, as well as factors that exacerbate or buffer against this. Cognitive aspects of stigma, for instance existing stigmatising beliefs held by carers and negative self-evaluations, are underrepresented in research and require more attention. There is also a need for larger scale population based prevalence studies in this area. Longitudinal studies could potentially clarify how affiliate stigma affect caregivers' social and emotional wellbeing over time.

Research around family dynamics that influence stigmatising beliefs held by carers is still lacking. The impact of stigma on family members other than parents may vary, but is rarely studied. Research into positive coping tactics and protective factors used by primary caregivers as well as other members of the family is also required. This is crucial in order to ensure carers are well supported in their caregiving needs.

Lastly, given that most of the research on stigma experiences relating to culture has been conducted in collectivist societies, there is a necessity for more cross-cultural comparisons; specifically, these should aim to investigate the experiences of caregivers who originate from collectivist cultures but have settled in an individualistic society, and vice versa.

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Part 2: Empirical Paper

Stigma Experienced by the Families of Individuals with Intellectual Disabilities

9. Abstract

Aims: This study set out to develop a scale measuring stigma in the families of individuals with intellectual disabilities and to examine the associations between family stigma and various sociodemographic, psychological and contextual variables.

Method: Four hundred and seven family carers from the United Kingdom completed a survey that contained an item pool measuring family stigma and positive aspects of caregiving, and questionnaires related to other variables, including subjective wellbeing, caregiver burden, positive meaning in caregiving, self-esteem and social support. Data from the new measure underwent exploratory factor analysis, reliability analyses, and validity tests. Data were further analysed using regression analyses.

Results: The new measure, the Family Stigma Instrument (FAMSI), yielded a five-factor structure. Perceived family stigma and positive aspects of caregiving were each represented by one factor. Affiliate stigma was represented by three factors: affective, cognitive and behavioural affiliate stigma. These corresponded to the three component model of attitudes. The FAMSI had good internal consistency with Cronbach's alpha coefficients ranging from 0.78 to 0.91 for the five factors and of 0.84 for the overall questionnaire. Correlations for test-retest reliability ranged from 0.62 to 0.83 for the five factors. The strongest predictor of affiliate stigma was perceived family stigma. Caregiver burden was predictive of all aspects of affiliate stigma except the cognitive aspect.

Conclusions: Families of individuals with intellectual disabilities perceive and internalise stigma which affects various aspects of their lives. Consideration of their experiences of stigma should take into account sociodemographic, psychosocial and contextual variables. This could also inform support and intervention strategies to alleviate the negative impact of family stigma.

10. Introduction

Recently published reports have found individuals with intellectual disabilities (ID) to be one of the most socially excluded groups and confront stigma, prejudice and significant challenges that limit their rights (European Union Monitoring and Advocacy Programme, 2005). Stigma has been conceptualised as the process of marginalisation and devaluation of certain groups, such as those with ID, based on attributes that differ from the dominant cultural group, including ethnicity, mental health problems, disability or drug-use (Goffman, 1963; Rusch, Angermeyer, & Corrigan, 2005). Stereotypes, prejudice and discrimination result from misconceptions about these attributes, and are perpetuated by biased social structures (Corringan, 2000).

10.1 Forms of Stigma

Five forms of stigma have been identified in the literature. The first, public stigma, refers to the attitudes held by society about the stigmatised individual (Bos, Pryor, Reeder & Stutterheim, 2013). The second type, institutional stigma, occurs when policies reduce the choice of the stigmatised person (Heflinger & Hinshaw, 2010). Self-stigma, the third type, occurs when the stigmatised person becomes aware of, endorses and internalises public stigma (Bos et al., 2013). Typically, studies have focused on self-stigma as experienced by the individual with ID. Often however, people associated with the target individual are also subjected to stigmatisation. This fourth type of stigma has been referred to as family stigma, courtesy stigma (Goffman, 1963) or associative stigma (Mehta & Farina, 1988).

Related to these is the construct of affiliate stigma, the fifth form, which involves the internalisation of stigma by associates of targeted individuals, such as caregivers of individuals with ID. It encompasses self-stigma and the consequent psychological responses of the caregiver.

Similar to self-stigma, it has been conceptualised as having three components: affect, cognitions and behavioural responses (Mak & Cheung, 2008). This differentiates it from constructs such as courtesy stigma or associative stigma, which focus mainly on others' perceptions of associates, but not on how the latter respond to being viewed negatively.

The caregivers of individuals with ID usually face a lifetime of responsibility with considerable stress (Baxter, Cummins & Yiolitis, 2000). They have to cope with both the physical and emotional needs accompanying the affected individual's disabilities. In addition, the stigma of being undervalued by others can adversely affect their quality of life (Chou & Palley 1998; Chou, Pu & Lee, 2009). Consequently, they may develop affiliate stigma, whereby they feel dejected and helpless about their association with the stigmatised individual, perceiving a negative impact on themselves. This may result in lowered self-esteem and impaired family relationships (Wahl & Harman, 1989). Mothers have been found to be especially susceptible to poor mental health, reporting higher levels of depression and anxiety than mothers of typically developing children (Cantwell, Muldoon & Gallagher, 2015; Chiu, Yang, Wong & Li, 2015). As a means of coping with these effects of affiliate stigma, caregivers may react by withdrawing socially, or even distancing themselves from the targeted individuals to avoid association (Mak & Cheung, 2008).

The aforementioned terms describing the experience of stigma by caregivers have often been used interchangeably in the literature. In general, there is a lack of clarity surrounding the effects of stigma on the family. Specifically, it is unclear whether family members actually perceive stigma, to what extent they internalise this and whether there is a clear relationship between perception of stigma and its internalisation or conversely, what factors determine whether family members perceive themselves to be stigmatised by others but reject this and assert their value and rights.

10.2 Measuring Stigma in Family Members

In most studies focusing on caregivers, stigma has been assumed to be a contributing stressor in caregiver burden. Research has mainly focused on caregiving for individuals with mental illness. For example, Szmukler, Burgess, Herrman, Benson, Colusa and Bloch (1996) used a 5-item stigma scale as part of an inventory measuring caregiving experiences of families of individuals with mental illness and found stigma to be one of the factors that made up the multidimensional construct of ‘caregiving experiences’.

One tool the Affiliate Stigma Scale, however, was developed in Hong Kong and designed specifically to measure stigma experienced by the family members of individuals with ID (Mak & Cheung, 2008). Though the authors presented the scale as a unidimensional one, a recent study in Taiwan by Chang, Su, Tsai, Yen, Liu & Lin (2015) using Rasch analysis found three separate domains of the scale. This supports the theoretical model of internalised stigma in that affect, cognitions and behaviours are three different processes. Moreover, as evident from the multifaceted consequences of stigma, a number of variables may affect the experience of affiliate stigma for the caregiver, some of which have been extensively studied.

10.3 Factors that may Affect Affiliate Stigma

In addition to extant research, the literature review in Part 1 highlighted a number of characteristics of the carer and the individual they care for, as well as other contextual variables, with demonstrated associations with affiliate stigma in relation to ID.

10.3.1 Carer characteristics

Research has indicated that carer characteristics, namely age, relationship to the person with ID, and ethnicity and culture, affect affiliate stigma in ID.

10.3.1.1 Age. Older caregivers tend to have had a longer period of contact with the individual with ID. They have been subjected to stigma for a longer duration and are therefore more likely to internalise stigma (Chiu, Yang, Wong, & Li, 2013). Moreover, carers who were aged 55 years and older have reported a strong effect of stigma on the quality of life (Chou et al., 2009; Sarkar, 2010).

10.3.1.2 Relationship to person with ID. In most cultures, mothers are regarded as the primary carer. They are more likely than fathers to be blamed for the child's disability and hence bear the shame of stigma (Chang, 2009). To avoid such shame, mothers sometimes isolate themselves from friends and families, and at times even their disabled child (Ali, Hassiotis, Strydom & King, 2012). In observations and in-depth interviews with family members of individuals with ID in Taiwan, Chang (2009) found that stigma was experienced by mothers but not fathers, as it was considered to be the moral duty of mothers to produce healthy children. The relationship to the family member with ID can therefore affect how stigma is perceived and internalised. However, most of these findings are derived from Chinese cultural contexts, where family shame and face are huge concerns. It is therefore unclear to what extent such findings translate to other cultural contexts.

10.3.1.3 Ethnicity and culture. In multi-ethnic societies such as the United Kingdom (UK), the role of culture may be particularly important in the experience of affiliate stigma. A collectivistic view of people as interdependent leads to an emphasis on group-oriented values and skills that contribute to effectively filling roles within the family (Leake & Black, 2005). As such, every member of the family is seen to play a part in upholding the reputation of the family and determining the family's social standing. This view is especially dominant in Asian contexts.

Parents of persons with ID from such communities, such as South-Asians, have reported feeling stigmatised by their extended families and communities due to having a child with ID (Sim & Bowes, 2005). Such carers experience similar or higher levels of stress as compared to White carers and are often unwilling to access specialist services due to mistrust and fear of services (Gangadharan, 2011). For example, in a study exploring the perceptions of UK-based Pakistani parents of an individual with ID, parents cited being conscious and affected by the stigmatising views held within the Pakistani community and their extended families. They were often excluded from social activities and eventually avoided these altogether. Moreover, they felt that things were easier for white families in a similar situation (Croot, Grant, Cooper & Mathers, 2008).

Another example is derived from Chinese cultures, where stigma moves quickly from the individual to their family, weakening or even severing vital links that connect families to social networks offering resources (Chiu et al., 2013). Similarly, studies from Africa, India, Pakistan and Bangladesh have shown that caregivers of individuals with ID are marginalised by the community in addition to their own families (Ali et al., 2012; Tilahun, 2016). Cultural factors evidently have implications on the way stigma is experienced by caregivers and therefore merit further study.

10.3.2 Characteristics of the Person with ID

10.3.2.1 Additional diagnoses. Individuals with ID often have a number of additional diagnoses. High prevalence rates of autism spectrum disorders and physical health conditions such as epilepsy, cerebral palsy and physical disabilities have been reported (Day & Jancer, 1994; Hand, 1994; Bhaumik, Tyrer, McGrother

& Ganghadaran, 2008). Carers of individuals with ASD have reported greater levels of stigma due to increased demands and stresses placed on them (Kwok, Leung & Wong, 2014; Werner & Shulman, 2015).

10.3.2.2 Visibility of disability and challenging behaviours. Individuals with ID often present with comorbid physical disabilities, some of which are highly visible. Moreover, some individuals with ID develop challenging behaviours. In interviews conducted with parents of 18 adult daughters and 15 adult sons with ID, Todd and Shearn (1996) found an increase in affiliate stigma as the offspring's disability became more apparent. Mak and Kwok (2010) tested an attribution model to explain the internalisation of stigma. They found that parents who perceived little control over the cause of their children's challenging behaviours and the extent of courtesy stigma encountered were more likely to experience affiliate stigma, i.e. internalise others' negative attitudes. Attributes that are less concealable and are easily identified permit society to discriminate and stigmatise on the basis of the person's visible attributes (Ahmedani, 2011). Research in the mental health field has found that more stigmatising stereotypes are attributed to conditions which generally have more visible symptoms, such as schizophrenia, compared to those that are more concealable, such as depression, even though both can lead to negative consequences for the stigmatised individual (Angermeyer & Matschinger, 1996; Lundberg, Hansson, Wentz & Björkman, 2007). With regard to ID, Levinson and Starling (1981) found greater stigma to be reported by families of lower socioeconomic status when their child's ID was visible. Studies also suggest that the public are more likely to be understanding when a child behaves "badly" if their disability is more visible, and less understanding if the disability is less visible, in which case they may attribute the behaviour to poor parenting (Siegel, 1996). Therefore, the experience of affiliate stigma may be predicated on the extent of challenging behaviours and the visibility of additional disabilities in ID.

10.3.3 Contextual and Psychosocial Variables

10.3.3.1 Subjective wellbeing. Subjective wellbeing is defined as a positive state of mind comprising one's entire life experience, including life satisfaction and happiness (Cummins & Weinberg, 2010). Recently, Werner & Shulman (2013) examined the relationship between affiliate stigma and subjective wellbeing in 176 caregivers of individual with developmental disabilities[1]. Subjective wellbeing was below the normative level, defined as the average set point of 75, and carers of individuals with autism spectrum disorders (ASD) reported particularly low subjective wellbeing compared to caregivers of those with ID and physical disabilities. Overall, greater levels of stigma were associated with lower ratings of subjective wellbeing. Moreover, an interaction was found between affiliate stigma and the child's diagnosis. Greater affiliate stigma was correlated to lower levels of subjective wellbeing among caregivers of individuals with ASD but not carers of individuals with ID or physical disabilities (Werner & Shulman, 2013).

10.3.3.2 Caregiver burden. A variable that is often examined together with stigma is subjective caregiver burden and positive associations have often been found. Mak & Cheung (2008) found affiliate stigma related to self-reported burden and distress in parents of children with ID and similar findings have been reported in parents of children with developmental disabilities (Werner & Shulman, 2015).

10.3.3.3 Positive meaning in caregiving. In general, research on affiliate stigma usually presents the caregiving experience as a negative one, wrought with

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[1] Developmental disabilities (DD) refer to severe and chronic disabilities which originate at birth or during childhood. DD are attributable to a mental or physical impairment or a combination of both, are manifested before age 22 and they continue indefinitely, substantially restricting the individual's functioning in several major life activities (Developmental Disabilities Assistance and Bill of Rights Act 2000). These include LD, ASD as well as physical disabilities.

stress and isolation. Recent studies however have shown that caregivers may view their circumstances in a positive light. For instance, Meyers, Mackintosh, and Goin-Kochel (2009) conducted interviews with parents of children who had ASD and found nine positive themes in addition to 15 negative ones. Some of these positive themes included seeing the child as a blessing, feeling that their lives had been enriched, and learning positive emotions such as compassion and patience through caregiving. Thus, in addition to affiliate stigma, it seems important to examine whether caregiving is viewed as a positive experience in order to better understand the caregiving experience.

10.3.3.4 Self-esteem. Self-esteem has been described as a sense of self-respect, worthiness, and adequacy (Rosenberg, 1965). It is concerned with psychological responses one makes toward the self and involves positive or negative cognitive, emotional, and behavioural reactions to the self (Mruk, 1999). Self-esteem influences other psychological processes. For example, in the area of developmental disabilities, self-esteem has emerged as the strongest predictor of subjective wellbeing (Werner & Shulman, 2013) and affiliate stigma has been found to be associated with feelings of decreased self-esteem in relation to others. Additionally, Werner & Shulman (2013) reported that positive psychosocial resources, including self-esteem, contributed to positive subjective wellbeing more than the negative influence of affiliate stigma and can serve as an internal coping resource (Werner & Shulman, 2013). Indeed, better psychological health among caregivers of individuals with disabilities has been related to self-esteem (Raina et al., 2005), although the direction of this relationship is unclear.

10.3.3.5 Social support. Social support has often been examined as part of the caregiving experience. It has been reported to have a distinct effect on parents' wellbeing (Mak & Kwok, 2010). In particular, support from significant others and friends reduced the effects of affiliate stigma (Mak & Kwok, 2010). In addition, positive psychosocial resources such as social support were more found to be more essential to positive subjective wellbeing than the negative influence of affiliate stigma. Social support therefore should be considered when examining affiliate stigma.

10.4 Rationale for Proposed Study

Research on the effects of stigma on families is very limited to date, and has mostly been conducted in Hong Kong and Israel. Moreover, there is no clear distinction between many of the terms used to describe the family's experience of stigma, thus it is still unclear if families perceive stigma in the first place, and what may determine whether this is internalised. Further, poor measurement of these constructs has limited research by reducing the interpretability of findings.

To date, the extant literature on the experience of stigma among carers of individuals with ID is limited at best. Research has focused on Chinese cultures (e.g. Mak & Cheung, 2008, 2010; Mak & Kwok, 2010; Chiu et al., 2013), where collectivistic views are dominant, and on Israel (e.g. Werner & Shulman 2013, 2015). In the UK, where an individualistic culture prevails, and there are higher levels of activism and more rights and legislation for people with disabilities, studies have been predominantly qualitative (Ali et al., 2012). Most have not explicitly focused on perceived or affiliate stigma (Sim & Bowes, 2005; Croot, 2012) and no studies have compared stigma across caregivers of different cultures.

Moreover, the Affiliate Stigma Scale (Mak & Cheung, 2008) was designed in Chinese for research in Hong Kong and China and subsequently translated into English. Critical examination of the scale's items by the author revealed that the scale was unsuitable for a Western context and has limited applicability with people of different ethnicities. Some items were too specific to Chinese cultures, such as the item “Having a family member with intellectual disability makes me lose face” under the cognitive aspect of the scale. The concept of “face”, defined as the desire to uphold one’s social image and worth based on performance and position within a relational context (Chiu et al., 2013), is a central tenet in Chinese society, but not one that resonates in a Western context. Many other items are worded in a way that presumes having a child with ID is of necessity a negative thing, e.g., “I feel emotionally disturbed because I have a family member with intellectual disability” and “I dare not to tell others that I have a family member with intellectual disability”. This would make the scale unacceptable to parents in a Western context, who would strongly object to the negative assumptions underpinning the questions.

10.5 Aims

Taking the aforementioned into account, this study set out to develop and evaluate a new scale measuring the different aspects of stigma among caregivers of those with ID, one that was suitable for assessing people of different ethnicities. To test the measure in an ethnically diverse sample, parents from White British and ethnic minority backgrounds were recruited. Further, assessing the previously mentioned variables suggested as being associated with affiliate stigma during the data collection stage helped in examining the psychometric properties of the newly developed scale.

The term “family stigma” was used throughout to collectively describe different aspects of stigma i.e. perceived family stigma and affiliate stigma. The following aims were pursued:

1. To develop a psychometrically sound measure suited to addressing family stigma among family members of people with ID from diverse backgrounds.
2. To determine which carer and ‘cared for person’ characteristics predict levels of family stigma.

3. To examine the association between different aspects of family stigma and the following variables: subjective wellbeing, caregiver burden, positive meaning in caregiving, self-esteem and social support.

11. Method

11.1 Participants

Participants were invited to complete an Internet survey hosted by Qualtrics, a web-based survey tool, through convenience and snowball sampling methods. Data were collected from 407 participants residing in the UK between 19th September 2016 and 16th February 2017, recruited mainly from Mencap's network and 3rd sector organisations supporting family carers of people with ID.

Demographic and other relevant information about family carers and individuals with ID were collected. This included relationship to the individual with ID, living arrangements, additional diagnoses, whether the family member was the primary caregiver, ethnicity and other relevant demographic variables, see Tables 1 and 2.

Of the 407 participants, 79.4% were mothers and 91.6% identified themselves as primary caregivers (Table 1). The sample of family members consisted primarily of White participants, making up 91.6% of the sample. While family carers generally had other children, 21.5% of the sample reported having another child with ID. Most carers had education levels up to 18 and were employed either full- or part-time. Looking at their family members with ID, there were almost twice as many males as females, and most had additional diagnoses, notably ASD, as well as additional disabilities (see Table 2).

11.2 Procedures

Ethical approval was granted by the UCL Research Ethics Committee (REC; Appendix B). This study was cross-sectional in nature and conducted in three parts.

The first part involved the assessment of validity and reliability, and factor analysis for the development of the new measure of family stigma. The second investigated the characteristics related to carers and individuals with ID that predicted family stigma using a web- based survey that included the new measure of affiliate stigma. The third examined the various psychological and contextual factors that may increase the likelihood of stigma being internalised or rejected using the same survey.

The total number of organisations that were sent an email requesting for them to disseminate information about the study (Appendix D) and a poster (Appendix E) to the caregivers was 461. Of these 103 agreed and subsequently confirmed that they had sent these out. No response was received from 334 organisations while 25 organisations declined, citing reasons that included ‘No time or resources to assess the value of different research studies’ (n=4), ‘not willing to contact family carers for research purposes’ (n=4) and ‘not interested’ (n=5). Other organisations that declined were unsuitable as their members did not care for someone with ID. The remaining eight organisations did not give a reason for declining.

The 103 organisations that agreed to participate in the study contacted between five and 100 caregivers in their networks via email, social media sites, such as Facebook groups and Twitter pages, and/or newsletters. Some also disseminated hard copies of a poster advertising the study. It was not possible to estimate the response rates as organisations did not track the number of carers they circulated information about the study to.

Table 1

Participant Demographics

	n	%
Relationship to Individual with ID		
Mother	323	79.4
Father	29	7.1
Other family member (please specify):	20	4.9
Sibling	35	8.6
Primary Caregiver	373	91.6
Yes	33	8.1
No		
Age of Caregiver		
Below 30	21	
31 – 44	117	28.7
45 – 54	157	38.6
55 – 65	74	18.2
Above 65	38	9.3
Ethnic Background		
White (any)	373	91.6
Asian	21	7.8
Black	8	1.9
Other	4	1.0
Living Arrangement		
Alone	55	13.5
With spouse/partner and/or children	310	76.1
With extended family	10	2.5
Other	29	7.1
Additional Children		
With ID	75	21.5
Without ID	280	68.8
Education Level		
Primary school (or equivalent if educated elsewhere)	9	2.2
To age 18 (including college)	216	53.1
University degree	180	44.2
Occupation		
Employed (full- or part-time)	176	41.8
Not employed (including homemaker or retired)	168	41.3
Student	11	2.7
Other, please specify:	57	14

Table 2

Information Related to Family Member with ID

	n	%
Gender of Individual with ID		
Male	278	68.3
Female	129	31.7
Additional Diagnoses		
Autism Spectrum Disorder	215	52.8
Down Syndrome	74	18.2
Other (includes fragile X, cerebral palsy and those with an unknown additional diagnosis	183	45.0
Additional Disabilities		
Mobility	74	18.2
Epilepsy/Seizure Disorder	80	19.7
Vision Impairment	59	14.5
Hearing Impairment	38	9.3
Other	119	29.9

Participants were informed that clicking on the electronic link to begin the survey would be taken as informed consent. Upon completion, participants were asked to provide their details if they wished to enter the prize draw or were happy to be contacted again for the test-retest survey. Personal information was immediately separated from participants' survey responses upon downloading the data into SPSS.

11.3 Measures

11.3.1 Perceived Family Stigma and Affiliate Stigma

Based on the original Affiliate Stigma Scale (Mak & Cheung, 2008), ten other scales relating to stigma and caregiving, and the existing literature on perceived family stigma and affiliate stigma, the Family Stigma Instrument (FAMSI) was initially constructed with a pool of 56 items. These items addressed perceived family stigma (e.g., "Other people might treat someone who has a family member with LD[2]

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[2] The term 'Learning Disabilities (LD)' was used throughout the measure as it is the most commonly used term in the UK to describe ID.

differently”), affiliate stigma (e.g., “I feel embarrassed about having a family member with LD”), and positive aspects of caregiving (e.g. “I have learned to speak out for my family member with LD”). Based on ethical and theoretical grounds, it was essential to include questions relating to positive aspects of caregiving (e.g. “Caring for my family member with LD has made me feel needed”) to highlight positive as well as negative dimensions in the experience of stigma.

A 5-point Likert scale ranging from *strongly disagree* (1) to *strongly agree* (5) was used. In order to assess face and content validity, six senior psychiatrists and clinical psychologists who worked in ID services were consulted. They were sent the initial 56 items and requested to provide ratings on the clarity, relevance and simplicity of each item. These criteria were based on an adapted version of the content validity index developed by Waltz and Bausell (1986). In addition, two focus groups facilitated by the author were conducted with eight to ten carers of diverse ethnicities in each. Professionals and carers provided feedback and rated the initial 56 items according to whether they felt the question was acceptable and appropriately worded. They also provided their views on the most useful format for the instrument; specifically whether a structured or unstructured format was more suitable. Feedback was analysed by sorting participants’ views into themes in order to determine a core set of items. The questionnaire was revised based on these findings. It was then piloted with ten carers of diverse ethnicities to identify any problems with the questionnaire. A final version of the measure consisting of 28 items (Appendix F) was generated based on feedback obtained during this process. A total stigma score was calculated by summing all the items of the scale. Scores range from 28 to 140, with a higher score indicating a higher level of stigma.

11.3.2 Subjective Wellbeing

Subjective wellbeing was assessed using the Personal Wellbeing Index (PWI; International Wellbeing Group, 2006; Appendix G). This has nine items measuring satisfaction in the following life domains: standard of living, personal health, achieving in life, personal relationships, personal safety, community-connectedness, future security and religion, as well as an overall question inquiring about satisfaction with life as a whole. However, the eighth item on ‘spirituality or religion’ and ninth item on ‘overall life satisfaction’ have been reported to be non-relevant in assessing Subjective Wellbeing (Cummins & Weinberg, 2010), and were therefore excluded in this study. An additional item on ‘leisure’, added by Werner and Shulman (2013), was used in this study instead. All items were rated on a scale ranging from 0 = completely dissatisfied to 10 = completely satisfied. Scores from the first seven questions were summed to yield an average score which represented subjective wellbeing.

The PWI has demonstrated good internal consistency in a sample of 2000 randomly selected participants (Cronbach’s alpha = 0.73; Lau, Cummins, & McPherson, 2005). Inter-item correlations (Cronbach’s alpha = 0.30 to 0.55) and item-total correlations (Cronbach’s alpha = 0.50; Cummins & Weinberg, 2010) have been reported as moderate. Test-retest reliability was good, yielding a correlation coefficient of 0.84 (Lau & Cummins, 2005).

11.3.3 Caregiving Burden

Caregiving burden was assessed with a single item taken from Werner and Shulman (2013): ‘‘To what degree do you feel burdened when you are with your child with a disability?’’ The item was rated from 1 = never to 5 = almost always.

11.3.4 Rosenberg’s Self-Esteem Scale

Self-esteem was measured using Rosenberg’s 10-item Self-Esteem Scale (1965; Appendix H). This is rated on a 4-point Likert scale, where 0 = strongly agree and 3 = strongly disagree.

An overall index was calculated by summing the score on the 10 item scores, with items three, five, eight, nine and ten reversed in valence. Possible scores ranged from zero to 30. Scores between 15 and 25 were within normal range while scores below 15 suggested low self-esteem (Rosenberg, 1965).

Internal consistency of the scale was 0.77 (Rosenberg, 1965) and test-retest reliability was 0.82 (Silber & Tippett, 1965). Criterion validity has been established at 0.55. The scale has shown correlations with measures of anxiety (r = -.64) and depression (r = -.54; Rosenberg, 1965).

11.3.5 Social Support

The Multidimensional Scale of Perceived Social Support (MSPSS; Zimet, Dahlem, Zimet & Farley, 1988; Appendix I) was used to assess perceptions of social support available from friends and a significant other. There were 12 items in total with three subscales addressing relationships with family, friends and a significant other in the following areas: social popularity, respect and items directly related to perceived social support. Ratings were made on a 7-point scale, from 1 = very strongly disagree to 7 = very strongly agree.

Scores were averaged to determine overall perception of social support. Mean total scale scores ranging from one to 2.9 would indicate a perception of low support, from 3 to 5 as moderate support, and a score from 5.1 to 7 as high support (Zimet et al., 1988).

The total scale had good internal consistency (α = 0.88), with internal reliability of .87, .85 and .91 reported for the family, friends and significant other subscales respectively (Zimet et al., 1988). The original authors also demonstrated negative associations between MSPSS and measures of depression (r = -.25).

11.3.6. Visibility of Disability and Challenging Behaviours

Parents were asked to indicate the presence of comorbid physical disabilities and provide a brief description of them in order to determine the visibility of disabilities. Challenging behaviour was assessed using Part I and some items from Part II of the Challenging Behaviour Interview (CBI; Oliver, McClintock, Hall, Smith, Dagnan & Stenfert-Kroese, 2003). Parents were asked whether their offspring had displayed one of the following five types of behaviour within the last month: self-injury, physical or verbal aggression, disruption of the environment and inappropriate vocalisations. An operationalised description taken from Oliver, McClintock, Hall, Smith, Dagnan and Stenfert-Kroese (2003) was provided for each behaviour type. Respondents were also be asked how concerned they are about each behaviour on a 7-point Likert scale, ranging from 0 = not at all concerned to 6 = extremely concerned.

In the original scale, only behaviours rated as three and above on the concern scale in Part I were considered in Part II of the interview, which consisted of 14 items. In this study, only the first three items assessing frequency, intensity and duration of each behaviour were used as the remaining were not relevant for the aims of this research. An overall mean score was obtained, with higher scores indicating increasingly challenging behaviour.

The mean Kappa coefficient across behaviours was 0.67 for inter-rater reliability and 0.86 for test-retest reliability (Oliver et al., 2003). The mean item reliability reported by Oliver et al. (2003) was .53 for inter-rater agreement and .74 for test-retest reliability. The reliability of the total overall score was very high for inter-rater and test–retest agreement (r = 0.90 and r = 0.96 respectively; Oliver et al., 2003). In terms of concurrent validity, the total score of the CBI was correlated to the total score of the Aberrant Behavior Checklist (r = 0.56; Oliver et al., 2003).

12. Results

12.1 Psychometric Properties of the FAMSI

SPSS Version 22 was used for statistical analysis. Descriptive statistics were computed to describe the sample and the main variables. The first aim of the study was to develop a psychometrically sound measure suitable for addressing family stigma among family members of people with ID from diverse backgrounds. To this purpose, an exploratory factor analysis (EFA) was conducted on responses to the 28 items using the scores obtained for each participant. Cronbach's alpha and interclass correlation coefficients (ICC) were calculated to assess internal consistency and test-retest reliability, respectively. Ali (2008) suggested a time period of two to six weeks to balance the likelihood that stigma might alter over time against the need to avoid recall bias. This approach was adopted for the current study and participants were asked to complete the new measure again after six weeks.

12.1.1 Factor analysis

The percentages of participants who selected each response option in the data set are shown in Appendix J. Notably, 65% of the carers perceived that the family would be excluded from social events (item 8) and 52.3% actually were excluded (item 22). Moreover, almost 79.1% of the carers indicated being aware of the way other people looked at them when they were in public with the individual with ID (item 23). Notably, there was unanimous disagreement with some of the items, for example items 26 and 28, suggesting that only a small minority of carers are heavily affected in terms of behavioural affiliate stigma. On the positive side, 60% felt that caring for an individual with ID had given them a more positive outlook on life (item 9). The Kaiser–Meyer–Olkin measure indicated excellent sampling adequacy for the analysis, KMO = .898, and Bartlett's test of sphericity χÇ (231) = 4645.603, $p <$ .0001, indicated that correlations between items were sufficiently large for EFA. Principal axis factoring and oblique rotation (direct oblimin) were used, with parallel analysis in combination with the scree plot supplementing factor retention. Oblique rotation allows for correlation, and was hence the preferred option as previous literature has shown the factors of stigma are correlated (e.g. Chang et al., 2015). The rest of the analyses are presented below.

The eigenvalues for each item were obtained. Five factors had eigenvalues above Kaiser's criterion of 1 and in combination explained 60.5% of the variance. Items with factor loadings greater than .40 and not cross loading on any other factor by more than .20 were considered to load significantly onto the respective factors (Table 3). Based on this, items 19, 'I feel worried that I/my family will be blamed for their LD' and 20, 'I am looked down upon when other people discover that I am related to my family member with LD' were removed. The scree plot (Figure 1) was slightly ambiguous and showed inflexions that would justify either a three or five factor solution.

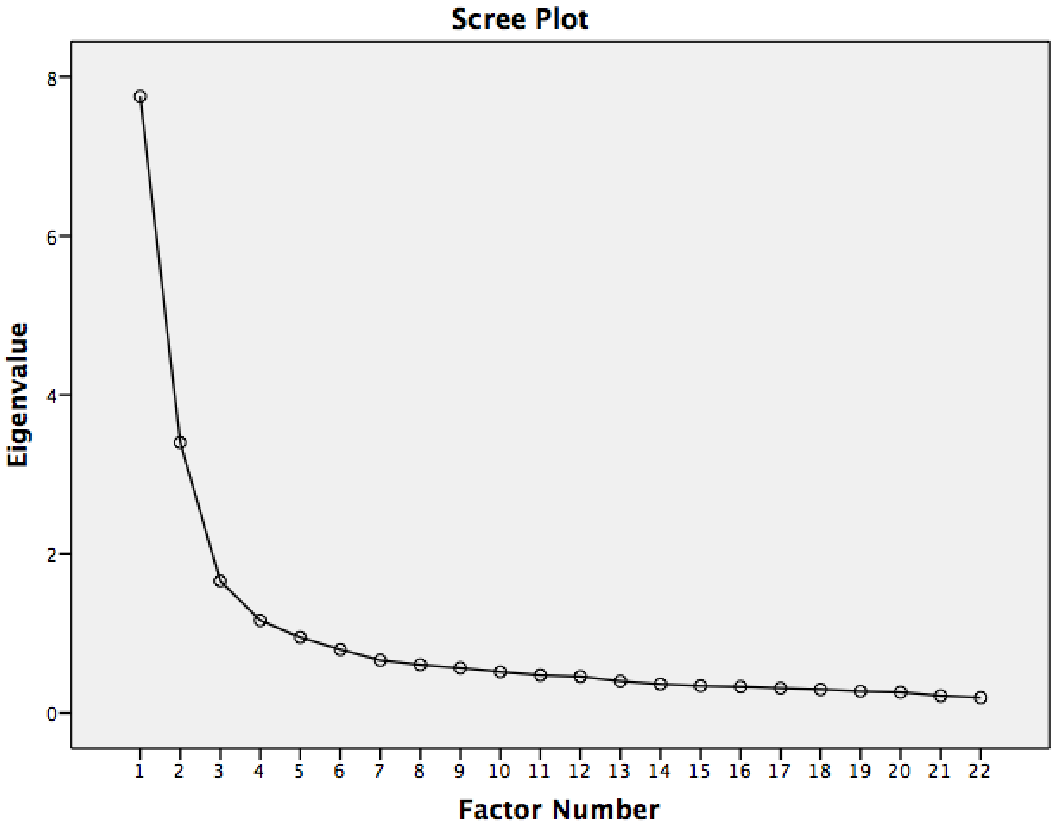

Figure 1. Scree Plot. This figure illustrates the scree plot from the exploratory factor analysis.

Given the large sample size, and the convergence of the scree plot and Kaiser's criterion on five factors, this is the number of factors that were retained. The items that cluster on the same components suggested that the first factor, which accounted for 27.7% of the total variance, represented perceived family stigma. The second factor explained 13.1% of the total variance and contained items about the affective aspects of affiliate stigma. The third factor related to the cognitive aspects of affiliate stigma and accounted for 6.1% of the total variance. The fourth factor accounted for only 4.4% of the variance and represented the behavioural aspects of affiliate stigma. As such, these three factors were labelled affective, cognitive and behavioural affiliate stigma. The final factor explained 9.3% of the variance and represented positive aspects of caregiving.

Perceived family stigma and each aspect of affiliate stigma were calculated using responses that were endorsed as either “agree” or “strongly agree”. Perceived family stigma was experienced by 59.3% of the respondents. The 34.5% who experienced affiliate stigma went on to endorse items related to affective affiliate stigma (11.4%), cognitive affiliate stigma (65.9%) and behavioural affiliate stigma (5.7%).

Table 3

Factor Loadings in the Exploratory Factor Analysis

Item	Factor Loadings
Factor 1: Perceived family stigma (α = 0.91)	
Some people might feel embarrassed about associating with the family of someone with LD.	.791
Some people might feel uncomfortable about going to the house of the family of someone with LD.	.775
Some people might treat the family of someone with LD more negatively.	.900
Some people might think that the family has done something wrong because of the person with LD.	.658
Some people might behave negatively towards the family of someone with LD when they are with the person with LD in public.	.821
Some people might avoid making friends with the family of someone with LD.	.805
Some people might not want to hear about any of the problems of the family of someone with LD.	.746
Some people might not invite the family of someone with LD to social events.	.621
Factor 2: Affective affiliate stigma	
I feel embarrassed about my family member with LD.	-.793
I feel distressed about being associated with my family member with LD.	-.766
I feel guilty about having my family member with LD in the family.	-.796
I feel uncomfortable when I have friends over because of my family member with LD.	-.631
I feel worried that I/my family will be blamed for the family member's LD.	-.469
Factor 3: Cognitive affiliate stigma	
I am looked down upon when other people discover that I am related to my family member with LD.	-.478
I am treated differently by some people when I am with my family member with LD.	-.788
I am excluded from activities when other people find out about their LD.	-.745
I am aware of how some people look at me when I am out with my family member with LD.	-.752
I am treated differently by some people because of my family member with LD.	-.808
Factor 4: Behavioural affiliate stigma	
I avoid introducing my friends to my family member with LD.	.704
I avoid telling people that I am related to my family member with LD.	.862
I avoid making new friends because of my family member with LD.	.653
I avoid being seen with my family member with LD.	.746
Factor 5: Positive aspects of caregiving	
Caring for my family member with LD has enabled me to develop a more positive attitude toward life.	.733
Caring for my family member with LD has made me feel needed.	.579
Caring for my family member with LD has strengthened my spirituality and faith.	.680
Caring for my family member with LD has allowed me to form friendships with others in a similar situation.	.603
Caring for my family member with LD has made me feel that I make a positive contribution to society.	.799
Caring for my family member with LD has strengthened some of my relationships with family/friends.	.705

Extraction Method: Principal Axis Factoring.
Rotation Method: Oblimin

Table 4

Test–retest reliability for subscales: Intraclass correlation coefficient and confidence interval

Subscale	ICC	95% Confidence interval	
		Lower	Upper
Perceived family stigma	0.45	0.09	0.67
Affective affiliate stigma	0.68	0.47	0.81
Cognitive affiliate stigma	0.77	.615	0.86
Behavioural affiliate stigma	0.68	0.47	0.81
Positive Aspects of Caregiving	0.78	0.63	0.87
Overall stigma	0.60	0.34	0.76

12.1.1.2 Internal consistency of subscales. Cronbach's alpha for the full 28-item scale was 0.84 (*M* = 73.32, *SD* =13.43), see Table 5. This indicates acceptable to excellent internal consistency for all five subscales (Tavakol & Dennick, 2011). Deletion of any of the items did not result in a substantial improvement in internal consistency.

Table 5

Reliability for subscales: Cronbach's alpha

Subscale	Cronbach's α	Mean	SD
Perceived family stigma	0.91	27.32	7.55
Affective affiliate stigma	0.80	8.15	4.09
Cognitive affiliate stigma	0.86	16.31	4.87
Behavioural affiliate stigma	0.77	5.81	2.73
Positive Aspects of Caregiving	0.78	19.83	5.20
Total Scale	0.84	73.32	13.43

12.1.1.3 Correlations between subscales. Correlations between the five factors were conducted using Bonferroni adjusted alpha levels of 0.01 per test (.05/4), see Table 6. The four family stigma factors were inter-correlated in the .19 to

.58 range, indicating that they tapped into related yet distinct aspects of stigma. The positive aspects of caregiving subscale was significantly negatively correlated with affective affiliate stigma and behavioural affiliate stigma subscales. The correlations between positive aspects of caregiving and two of the other subscales, perceived family stigma and cognitive affiliate stigma, were also negative but not significant.

Table 6

Correlations between FAMSI subscales.

Subscales	Perceived family stigma	Affective affiliate stigma	Cognitive affiliate stigma	Behavioural affiliate stigma	Positive aspects of caregiving
Perceived family stigma	-				
Affective affiliate stigma	.19**	-			
Cognitive affiliate stigma	.55**	.27**	-		
Behavioural affiliate stigma	.26**	.58**	.32**	-	
Positive aspects of caregiving	-.49	-.14**	-.04	-.20**	-

*** Pearson's correlation is significant at p<.01 level (2-tailed).*

12.2 Sociodemographic Characteristics

The second aim of the study was to determine which carer and 'cared for person' characteristics predicted levels of family stigma. In order to address this, total stigma scores were calculated for each participant, with higher scores indicating more stigma. Subscale scores of perceived family stigma and affiliate stigma were also obtained. Independent t-tests, one-way ANOVAs, Pearson's correlations and regression analyses were computed to determine the associations between family stigma and caregiver and 'cared for persons' characteristics, see Table 7. The risk of type 1 error as a result of multiple testing was managed by using the Bonferroni correction, with post hoc comparisons for t-tests and for one-way ANOVAs. Correlations were computed using adjusted alpha levels of 0.01 (.05/6).

For affective affiliate stigma, significant correlations were found with ethnicity, *t* (35.80) = -2.13, *p*=.002, and with challenging behaviours, *r* = .13, *p*=.007. Cognitive affiliate stigma was associated with the carer's relationship to the individual with ID, *t* (405) = 3.02, *p*=.003, caregiver age, *F* (4, 402) = 4.74, *p*=.001, number of additional diagnoses, *F* (3, 406) = 4.84, *p*=.003, and with challenging behaviours, *r* = .21, *p* <.001. Behavioural affiliate stigma showed significant effects only for challenging behaviours, *r* = .21, *p* < .001. The total affiliate stigma score i.e. the composite scores of affective, cognitive and behavioural affiliate stigma, was found to be associated with diagnoses, *F* (3, 406) = 7.41, *p* < .001, and challenging behaviours, *r* = .24, *p* < .001. Family members of those who had diagnoses of ID and ASD (*M* = 2.31, *SD* = 0.61), and those displaying one or more challenging behaviours (*M* = 2.18, *SD* = 0.65) were found to experience higher levels of affiliate stigma overall.

Based on these associations, a regression analysis was run with all the characteristics that were found to have significant effects to determine which of these independently predicted the different aspects of stigma (Table 8).

Table 7

Associations between stigma and predictor variables

	Affective		Cognitive		Behavioural		Total Affiliate stigma	
	Mean (*SD*)	*Statistic*	Mean (*SD*)	*Statistic*	Mean (*SD*)	*Statistic*	Mean (*SD*)	*Statistic*
Caregiver characteristics								
Relationship to Individual		*t* (405) = 0.13		*t* (405) = 3.02*		*t* (403) = 0.59		*t* (405) = 1.79
Mother (*n* = 323)	1.58 (0.82)		3.60 (1.01)		1.46 (0.67)		2.21 (0.63)	
All others (*n* = 84)	1.57 (0.89)		3.23 (1.03)		1.41 (0.72)		2.07 (0.69)	
Age		*F*(4,402) = 1.02		*F*(4,402) = 4.74**		*F*(4,400) = 0.47		*F*(4,402) = 1.77
< 30 (*n* = 21)	1.43 (0.63)		3.68 (1.02)		1.57 (0.59)		2.22 (0.55)	
31 – 44 (*n* = 117)	1.53 (0.78)		3.63 (0.93)		1.41 (0.60)		2.19 (0.56)	
45 – 54 (*n* = 157)	1.67 (0.89)		3.60 (1.01)		1.49 (0.79)		2.25 (0.71)	
55 – 65 (*n* = 74)	1.48 (0.82)		3.49(1.03)		1.40 (0.60)		2.12 (0.60)	
> 65 (*n* = 38)	1.57(0.84)		2.87 (1.10)		1.45 (0.68)		1.96 (0.74)	
Education Level		*F*(4,402) = 2.29		*F*(4,402) = 0.41		*F*(4,400) = 0.05		*F*(4,402) = 0.22
Primary to age 16 (*n* = 51)	1.41 (0.76)		3.65 (0.95)		1.48 (0.59)		2.18 (0.56)	
Ages 16 to 18 (*n* = 174)	1.54 (0.80)		3.50 (1.07)		1.44 (0.65)		2.16 (0.63)	
University degree (*n* =180)	1.67 (0.88)		3.52 (1.00)		1.44 (0.73)		2.20 (0.69)	
Ethnicity		*t* (405) = -2.13*		*t* (405) = 1.86		*t* (403) = -0.97		*t* (405) = -0.62
White (*n* = 373)	1.54 (0.79)		3.55 (1.01)		1.44 (0.66)		2.17 (0.62)	
Other (*n* = 34)	2.00 (1.17)		3.21 (1.12)		1.56 (0.93)		2.25 (0.87)	

Table 7 Continued

	Affective		Cognitive		Behavioural		Total Affiliate stigma	
	Mean (*SD*)	*Statistic*	Mean (*SD*)	*Statistic*	Mean (*SD*)	*Statistic*	Mean (*SD*)	*Statistic*
Individual characteristics								
Gender		*t* (405) = 1.00		*t* (405) = 0.60		*t* (403) = 1.12		*t* (405) = 1.32
Male (*n* = 278)	1.60 (0.88)		3.55 (1.02)		1.46 (0.72)		2.21 (0.67)	
Female (*n* = 129)	1.52 (0.73)		3.48 (1.04)		1.40 (0.60)		2.12 (0.59)	
Diagnoses		*F*(4,402) = 0.55		*F*(4,402) = 4.30**		*F*(4,400) = 1.12		*F*(4,402) = 7.41**
ID only (*n* = 52)	1.68 (0.90)		3.44 (0.01)		1.45 (0.80)		2.18 (0.73)	
ID + ASD (*n* = 140)	1.72 (0.90)		3.68 (0.96)		1.65 (0.76)		2.31 (0.61)	
ID + DS/Other (*n* = 133)	1.56 (0.74)		3.28 (1.07)		1.46 (0.72)		1.98 (0.60)	
ID + ≥2 diagnoses (*n* = 82)	1.74 (0.86)		3.71 (0.96)		1.60 (0.74)		2.28 (0.66)	
Physical disabilities		*F*(4,402) = 1.21		*F*(4,402) = 2.89		*F*(4,400) = 0.95		*F*(4,402) = 0.97
None (*n* = 208)	1.62 (0.88)		3.54 (1.04)		1.46 (0.73)		2.15 (0.69)	
< 2 (*n* = 119)	1.58 (0.81)		3.88 (0.84)		1.49 (0.68)		2.25 (0.60)	
≥ 3 (*n* = 80)	1.45 (0.77)		3.67 (0.87)		1.36 (0.53)		2.16 (0.58)	
Challenging behaviours		*r* = 0.19**		*r* = 0.55**		*r* = 0.26**		*r* = 0.24**

**p* < .05; ***p* < .01

Table 8

Results of Regression Analyses for all Characteristics Tested as Potential Predictors of Different Aspects of Affiliate Stigma (N *= 407)*

	Affective affiliate stigma			Cognitive affiliate stigma			Behavioural affiliate stigma			Total Affiliate stigma		
Characteristic	*B*	*SE B*	*β*	*B*	*SE B*	*β*	*B*	*SE B*	*β*	*B*	*SE B*	*β*
Relationship to individual[a]	-0.03	0.10	-.01	-0.35	0.12	-.14**	-0.04	0.08	-.02	-0.13	0.08	-.08
Caregiver age [b]	0.03	0.04	.05	-0.15	0.05	-.15**	0.01	0.03	.02	-0.03	0.03	-.05
Ethnicity [c]	0.46	0.15	.15**	-0.25	0.18	-.07	0.15	0.12	.06	0.12	0.11	.05
Diagnoses [d]	-0.04	0.04	-.05	-0.01	0.05	-.01	-0.02	0.04	-.03	-0.03	0.03	-.04
No. of physical disabilities [e]	-0.07	0.05	-.07	0.18	0.06	.14**	-0.04	0.04	-.05	0.02	0.04	.03
Challenging behaviour [f]	0.03	0.01	.15**	0.04	0.01	.17**	0.03	0.01	.21**	0.03	0.01	.23**
R^2			.05			.10			.05			.07
F			3.61**			7.72**			3.52**			5.01**

[a]Relationship to individual: 1 = *mothers*, 2 = *all others*. [b]Caregiver age: 1 = *<30*, 2 = *31-44*, 3 = *44-54*, 4 = *55-65*, 5 = *55-65*, 6 = *>65*. [c]Ethnicity: 1 = *White*, 2 = *other*. [d]Additional diagnoses: 1 = *ID only*, 2 = *ID+ASD*, 3 = *ID + DS/Other*, 4 = *ID + ≥2*. [e]Number of physical disabilities: 1 = *none*, 2 = *< 2*, 3 = *≥ 3*. [f]Intensity of challenging behaviours.
*p < .05. **p < .01.

Post hoc corrections were made using the Sidak-Bonferroni correction and regressions were interpreted using an adjusted alpha level of 0.01 (.05/5). The results indicated that two predictors, challenging behaviours and ethnicity, explained 5% of the variance in affective affiliate stigma (R^2=.05, F(6,400)=3.61, p=.002). The four predictors that explained 10% of the variance in cognitive affiliate stigma (R^2=.10, F(6,400)=7.72, p<.001) were challenging behaviours, the carer's relationship to the individual with ID, caregiver age and number of physical disabilities. The only predictor of behavioural affiliate stigma was challenging behaviour, explaining 50% of the variance (R^2=.05, F(6,398)=3.52, p=.002). Similarly, challenging behaviour was the only predictor for total affiliate stigma, accounting for 7% of the variance (R^2=.07, F(6,400)=5.01, p<.001).

12.3 Psychological and Contextual Variables

The third aim of study was to examine the association between different aspects of family stigma and the following variables: subjective wellbeing, caregiver burden, positive meaning in caregiving, self-esteem and social support. For this, along with the statistics used in the second aim, multivariate linear regression analyses were additionally computed to assess the role of these psychological and contextual variables in predicting the different aspects of family stigma.

The correlations between perceived family stigma, affiliate stigma, and the psychological and contextual variables with Sidak-Bonferroni adjusted alpha levels of 0.01 (.05/5), are presented in Table 9.

Table 9

*Correlation between Contextual/Psychological Variables and Different Aspects of Affiliate Stigma (*N *= 407)*

	Affiliate Stigma			
Variables	Affective	Cognitive	Behavioural	Total Affiliate
Burden	-.39**	-.26**	-.30**	-.40**
Positive Aspects of Caregiving	-.14**	-.42	-.20**	-.16**
Subjective wellbeing	-.15**	-.38**	-.31**	-.37**
Self-Esteem	-.10*	-.09	-.13**	-.14**
Social Support	.04	.25**	.20**	.22**

*$p < .05$. **$p < .01$.

Affective affiliate stigma was correlated with all variables except social support, most highly with carer burden. The correlation with self-esteem was not significant after correction. Cognitive affiliate stigma was correlated with all variables except positive aspects of caregiving and self-esteem. Behavioural affiliate stigma was correlated with all variables, as was overall affiliate stigma.

Multiple regression analysis was conducted using all psychological and contextual variables and the three subscales as well as total affiliate stigma as outcomes, with the Sidak-Bonferroni adjusted alpha levels of 0.01 (.05/5) (Table 10).

Table 10

Results of Regression Analysis for All Contextual/Psychological Variables Tested as Potential Predictors of Different Aspects of Affiliate Stigma (N = *407)*

	Affective affiliate stigma			Cognitive affiliate stigma			Behavioural affiliate stigma			Total Affiliate stigma		
Variable	*B*	*SE B*	*β*	*B*	*SE B*	*β*	*B*	*SE B*	*β*	*B*	*SE B*	*β*
Positive aspects of caregiving	0.04	0.07	.04	0.14	0.07	.15*	-0.06	0.06	-.07	0.05	0.05	.06
Burden	-0.31	0.05	-.37**	-0.13	0.06	-.16*	-0.14	0.05	-.19**	-0.19	0.04	-.31**
Subjective wellbeing	-0.02	0.07	-.02	-0.30	0.08	-.32**	-0.14	0.06	-.20*	-0.14	0.05	-.25**
Self-esteem	-0.05	0.05	-.06	-0.01	0.06	-.01	-0.06	0.05	-.10	-0.04	0.04	-.06
Social support	-0.17	0.16	-.08	0.16	0.18	.07	0.02	0.15	.01	<0.00	0.12	.00

*p < .05. **p < .01.

Burden was the only significant predictor of affective affiliate stigma, with 16% of the variance explained by the model (R^2=.16, F(6,400)=15.1, p<.001). Cognitive affiliate stigma was associated with three variables; positive aspects of caregiving, subjective wellbeing and burden, with the model accounting for 17.7% of the variance (R^2=.18, F(6,400)=15.0, p<.001). Three variables initially explained 14.9% of the variance in behavioural affiliate stigma (R^2=.15, F(6,398)=14.0, p<.001). These were burden, subjective wellbeing and self-esteem. However, self-esteem was not significant after correction. Total affiliate stigma was associated with burden and subjective wellbeing, which accounted for 22.2% of the variance (R^2=.22, F(6,400)=22.9, p<.001).

12.4 Importance of Sociodemographic, Psychological and Contextual Variables

In order to examine the relative importance of the caregiver/cared for characteristics and contextual variables, four separate multivariate linear regressions were performed with affective affiliate stigma, cognitive affiliate stigma, behavioural affiliate stigma, and overall affiliate stigma as outcome variables in each regression. For all four regressions, perceived family stigma was entered in Step 1, and the significant carer/cared for characteristics together with *all* contextual variables were entered in Step 2. Post hoc corrections were made using the Sidak-Bonferroni correction and regressions were conducted using adjusted alpha levels of 0.005 per test (.05/11). Results of these analyses are shown in Table 11.

Table 11

Summary of Final Hierarchical Regression Analysis for Different Aspects of Affiliate Stigma (N = 407)

		Affective affiliate stigma			Cognitive affiliate stigma			Behavioural affiliate stigma			Affiliate stigma		
	Variable	*B*	*SE B*	*β*	*B*	*SE B*	*β*	*B*	*SE B*	*β*	*B*	*SE B*	*β*
Step 1													
	Constant	1.02	0.16		1.45	0.12		0.80	0.13		1.09	0.11	
	Perceived family stigma	0.16	0.04	.18**$^{\Delta}$	0.61	0.05	.55**$^{\Delta}$	0.19	0.04	.26**$^{\Delta}$	0.32	0.03	0.46**$^{\Delta}$
Step 2													
	Constant	2.07	0.49		3.34	0.52		2.30	0.41		2.54	0.34	
	Perceived family stigma	0.15	0.04	.16**$^{\Delta}$	0.52	0.04	.47**$^{\Delta}$	0.14	0.04	.19**$^{\Delta}$	0.27	0.03	.39**$^{\Delta}$
	Challenging behaviours	0.01	0.01	.07	0.01	0.01	.04	0.02	0.01	.10*	0.01	0.01	.09*
	Relationship to individual	0.07	0.10	.03	-0.10	0.10	-.04	0.07	0.80	0.04	0.01	0.07	.01
	Caregiver age	0.02	0.04	.03	-0.19	0.04	-.19**$^{\Delta}$	<0.00	0.03	<.00	-0.05	0.03	-.08*
	No. of physical disabilities	-0.14	0.05	-.13**	0.06	0.05	.05	-0.10	0.04	-.18**	-0.06	0.03	-.07
	Ethnicity	0.41	0.14	.13**$^{\Delta}$	-0.24	0.15	-.06	0.15	0.12	.06	0.11	0.10	.05
	Burden	-0.25	0.04	-.35**$^{\Delta}$	-0.10	0.04	-.11**	-0.10	0.00	-.17**$^{\Delta}$	-0.15	0.03	-.27**$^{\Delta}$

Table 11 continued

Positive meaning in caregiving	-0.06	0.05	-.07	0.11	0.05	.09*	-0.64	0.40	-.08	-0.01	0.03	-.02
Subjective wellbeing	-0.01	0.03	-.03	-0.10	0.03	-.22**$^{\Delta}$	-0.06	0.02	-.17**	-0.05	0.02	-.18**$^{\Delta}$
Self-esteem	-0.22	0.14	-.07	-0.09	0.15	-0.02	-0.26	0.12	-.11*	-0.19	0.10	-.08
Social support	0.07	0.03	.12*	<0.00	0.04	<.00	0.02	0.03	.03	0.03	0.02	.06
R^2			.03			.31			.07			.21
ΔR^2			.19			.12			.14			.17
F for ΔR^2			9.31**$^{\Delta}$			8.46**$^{\Delta}$			6.86**$^{\Delta}$			10.5**$^{\Delta}$

*p < .05. **p < .01, $^{\Delta}p$ < .005

For affective affiliate stigma, perceived family stigma, number of physical disabilities, ethnicity, burden and perceived social support were predictive, explaining 3% of the variance. However, number of physical disabilities and perceived social support did not remain significant after correction. For cognitive affiliate stigma, perceived family stigma, caregivers' age, burden, positive aspects of caregiving and subjective wellbeing were predictive, explaining 31% of the variance. Burden and positive aspects were not significant following post hoc corrections. For behavioural affiliate stigma, perceived family stigma, challenging behaviours, number of physical disabilities, burden, subjective wellbeing and self-esteem were predictive, explaining 7% of the variance. The only two variables that survived correction were perceived family stigma and burden. For total affiliate stigma, perceived family stigma, caregiver's age, challenging behaviours, burden, and subjective wellbeing were predictive. After post hoc corrections, perceived family stigma, burden and subjective wellbeing remained significant.

13. Discussion

The main aim of this study was to develop and validate a new measure of family stigma as experienced by family caregivers of people with ID. This discussion first reviews the psychometric properties of the scale then considers the findings and their implications more broadly.

13.1 Psychometric Properties of the FAMSI

Factor analysis of the FAMSI identified a 26-item, self-report measure assessing five factors: (1) perceived family stigma; (2) affective affiliate stigma; (3) cognitive affiliate stigma; (4) behavioural affiliate stigma and (5) positive aspects of caregiving (Table 3).

The scale had an ICC of 0.60 and all of the identified factors had a Cronbach's alpha above 0.7, indicating good levels of test-retest and interval reliability. Correlations between the subscales demonstrated that they were measuring the same underlying construct of stigma. Taken together, this suggests that the new scale has a good level of content and construct validity.

Although the ICC for perceived family stigma was low, that for the other subscales was relatively higher, between 0.68 to 0.78. This suggests that awareness of stigma may fluctuate over time as it is more dependent on the reactions of others and a whole host of other contextual variables, while actual internalisation is more stable. Research on self-stigma in schizophrenia has found that stigmatising beliefs may be stronger at the point of recall of discriminatory experiences, yet the internalisation of stigma persists even long after exposure to stigmatising experiences (Lysaker, Tunze, Yanos, Roe, Ringer & Rand 2012). While stigma has been found to be enduring (Ali et al., 2012), it is possible that conscious awareness of it fluctuates markedly depending on other intervening factors and events. Therefore, when researching perceived family stigma, the measure may only be useful to assess this at specific points in time. This was confirmed by some of the qualitative feedback provided by participants in this study. For example, one carer stated that it was difficult to answer some questions "as my child is changing and behaviours evolving... So this is a point in time answer".

The perceived family stigma factor contains eight items pertaining to how carers perceive that family members of people with ID may be affected by stigma (associated with ID). The remaining factors represented three aspects of affiliate stigma and positive aspects of caregiving. The affective subscale represents feelings associated with affiliate stigma. The item relating to blame was removed from this scale as it failed to load sufficiently. A possible reason for this could be that the experience of blame involves assignment of responsibility by others, while the other emotions mentioned in the factor, such as guilt and embarrassment, are self-initiated.

The cognitive factor of the scale consists of items relating to self-evaluation, while the behavioural factor consists of items assessing the behavioural (or enacted) consequences of affiliate stigma. One item from the cognitive factor was removed as it loaded on all factors. The final factor comprised items addressing positive aspects of caregiving. It was an addition deemed important not only for the aforementioned ethical and theoretical reasons, but also because currently there are no validated scales relating to stigma that also assess positive dimensions of the caregiving experience in ID. As noted above, more than half of the family carers felt that caring for an individual with ID had given them a more positive outlook on life. This was further highlighted by some of the qualitative feedback provided by participants in this study, where they mentioned feeling “blessed” and that their lives were “fulfilled” and “enriched” by having the individual with ID in it. Caregiver burden intensifies negative affect to a greater extent than it would lessen positive affect, hence it might be especially important to regularly emphasise the positive aspects of caregiving (Lawton et al., 1991). Moreover, the lack of such positive aspects may be a risk factor in identifying family carers who are finding it difficult to cope and are therefore more vulnerable to stigma (Lloyd, Patterson & Muers, 2014).

Unlike Mak and Cheung (2008), who identified a single factor in their Affiliate Stigma Scale, the findings from this study were more in line with those of Chang et al. (2015), who found three separate domains using Mak and Cheung’s (2008) scale with 453 family caregivers of relatives with mental health problems. These domains were similar to the three factors relating to affiliate stigma in this study.

The development of the FAMSI in this study has attempted to clarify the conceptualisation of stigma among family members of individuals with ID. In line with the literature review in Part 1, the results have shown that family carers indeed do initially experience perceived family stigma, whereby they become aware of others' negative perceptions of the individual with ID and their family members before potentially internalising negative evaluations and developing affiliate stigma with its consequent behavioural responses. Indeed, a large proportion of the carers in the study indicated in their responses that they were aware of the way others looked at them when they were in public with the individual with ID. Over half also indicated that they were in fact excluded from social events. Interestingly, 65% of the respondents perceived that, in general, the family of someone with ID would be excluded from social events, despite not necessarily encountering social exclusion themselves. A possible explanation for this could be anticipated stigma, described as the actual or expected fear of societal response due to the stigmatised condition (Weiss, 2006). It may be that family carers in this study came to anticipate social exclusion due to the awareness of stereotypes related to caring for an individual with ID. They may then respond to such anticipated exclusion by concealing the individual's condition and withdrawing from social activities (Ngo et al., 2012). As highlighted in the literature review, with increasing affiliate stigma, family carers may fear future discrimination and stigma, eventually creating a vicious circle of increasing levels of perceived family stigma and affiliate stigma. Interventions targeting such anticipated stigma should therefore involve raising public awareness of family stigma in ID and challenging existing socio-cultural norms in order to break the circle of stigma.

Further, the findings on the proportion of participants experiencing each aspect of stigma indicate that some family carers may be well aware of the impact of ID stigma on the families of people with ID, yet show no signs of internalising negative attitudes. This could be due to a number of reasons. For example, carers who have had positive experiences of caregiving and value their relationship with the individual with ID may recognise ID stigma but actively resist it or be less vulnerable to it due to perceiving their caring role and their loved one with ID in positive terms. Research in the area of self-stigma in the mental illness field suggests that the internalisation of stigma involves a sense of threat to one's sense of worth, regard and confidence (Vogel & Wade, 2009). Indeed, the proportion of carers who experienced cognitive affiliate stigma in this sample was higher than for the other two aspects of affiliate stigma. Therefore, a family carer who sees the individual with ID or their relationship with them as undermining their fundamental worth may be more likely to internalise stigma. This implies that the pathway to affiliate stigma may occur via different affective, cognitive or behavioural means. As suggested in the literature review in Part 1, perceived family stigma may prompt different processes that involve negative self-evaluations and negative social comparisons (Ali et al. 2012), and thus initiate the internalisation of stigma by caregivers.

Based on the factors derived in the scale, there appear to be two processes involved in the experience of stigma among family members of individuals with ID. These processes can best be described in terms of the four-stage model of the internalisation of mental illness stigma proposed by Corrigan, Michaels, Vega, Gause, Watson and Rüsch (2012). In the first stage, there is an awareness of the negative stereotypes associated with intellectual disability, as captured by the perceived family stigma factor. In subsequent stages, there is an agreement with these stereotypes, an application of these to oneself and finally the risk of harm to self through various emotional and behavioural responses. This reinforces the findings of the literature review in Part 1, which suggested that awareness was a prerequisite of the experience of stigma before it is internalised in the three different dimensions of affective, cognitive and behavioural affiliate stigma. Moreover, in this second process, the three aspects of affiliate stigma identified in this study appear to map onto the three components of attitudes, which have similarly been described in terms of affect, cognition and behaviour (Eagly & Chaiken, 1993). According to Jain (2014), the affective component encompasses the emotional reactions towards an attitude object. This component works in tandem with the cognitive one, which concerns thoughts and beliefs about the object and stores information about the object; and the behavioural component consists of actions or overt responses that are an outcome relating to the attitude object. These can be either negative or positive and in combination establish one's overall attitude to an object (Jain, 2014).

These processes have important implications for family members. It could be that caregivers who are aware of ID stigma (i.e. perceived family stigma in process 1) may then apply the three domains of attitudes to themselves and construct an overall attitude towards their role as family members of someone with ID (i.e. aspects of affiliate stigma in subsequent processes). By assessing the three aspects of attitudes, a clinician may be able to gain a better understanding of not only family members' attitudes but also their relationship with the individual with ID in order to identify whether these may be the source of conflict and potentially meriting sensitive intervention.

13.2 The Role of Carer/Cared for Characteristics

Three regression analyses involving carer/cared for characteristics, contextual and psychological variables, and a combination of these were carried out with the different aspects of affiliate stigma as outcomes.

The proportion of variance explained by the sociodemographic and contextual variables notably fluctuated for each aspect of affiliate stigma in the final regression. While the variables explained up to 31% of the variance for cognitive affiliate stigma, they only accounted for 3% of the variance in affective affiliate stigma, 7% in behavioural affiliate stigma, and 21% in total affiliate stigma.

In terms of ethnicity, the results indicated that the White majority experienced more affective affiliate stigma and burden even. This association held in the final regression for affective affiliate stigma, when perceived family stigma was controlled for, though the regression coefficient decreased slightly. In addition to ethnicity, burden emerged as a significant predictor of affective affiliate stigma in the same regression. In Western cultures that are strongly individualistic, attitudes towards those with ID have been shown to be more accepting and less stigmatising (Bhugra 1989; Westbrook, Legge & Pennay, 1993). Additionally, higher levels of public stigma towards disorders such as schizophrenia have been found in Nigeria, Ethiopia and India (Cohen, Thara & Gureje, 2008). Extending these findings to the present study, it could be the case that, when compared to the ethnic minority in this sample, the White majority were more willing to report shame and embarrassment arising from affiliate stigma and related levels of burden. For the family carers from BME communities, where collectivistic views tend to dominate, every member of the family is seen to play a part in upholding the reputation of the family and determine the family's social standing (Leake & Black, 2005). Hence, family members from

BME communities may have been less forthcoming about the stigma and burden they experience in giving care to an individual with ID.

In general, taking into account the adjusted *p*-value, the contribution of challenging behaviours disappeared for all aspects of affiliate stigma when perceived family stigma was controlled for in the final regression analyses. There was also a strong correlation between perceived family stigma and the intensity of challenging behaviours, suggesting that challenging behaviour might affect perceived family stigma more than the other components of affiliate stigma. Indeed, as highlighted in the literature review in Part 1, initial awareness of stigma may be largely dependent on the nature and extent of challenging behaviours displayed by the individual with ID when the family carer is in public with them. Others may show negative responses to an individual exhibiting such behaviours and an awareness of these may induce negative emotions in the family member. Such repeated encounters may gradually be associated with increased feelings of perceived family stigma and threat to one's confidence and sense of worth (Cantwell et al., 2015).

Caregiver age, relationship to the individual with ID, number of additional physical disabilities and the severity and intensity of challenging behaviours were found to be significant predictors of cognitive affiliate stigma in the initial regression. Caregiver age remained a significant predictor of cognitive affiliate stigma, together with subjective wellbeing, even after perceived family stigma was controlled for in the final regression, while burden and positive aspects of caregiving were no longer predictive. This is an interesting finding as caregiver burden was a significant predictor of all other aspects of affiliate stigma. Contrary to findings on older caregivers as more likely to internalise stigma that affects their quality of life (Chiu et al., 2013; Chou et al., 2009), the results suggest that younger family members of individuals with ID experience a decrease in subjective wellbeing and higher levels of cognitive affiliate stigma. This effect disappears in affiliate stigma as a whole though when burden is taken into account. Indeed, older caregivers have shown higher levels of burden in conditions such as schizophrenia, dementia and even chronic illnesses (Caqueo-Urízar & Gutiérrez-Maldonado, 2006; Almberg, 1997; Limpawattana, 2012).

In a literature review seeking to understand the stigma experiences of parents of adults with ID, Sarkar (2010) reported that among a sample of 88 parents, 48% of parents below the age of 55 felt stigma affected their ability to interact with other relatives. The findings of the present study suggest that a cohort effect may influence the internalisation of stigma by younger family members. As compared to older parents, younger parents have been found to be more likely to report that others thought less of and were afraid of their offspring with ID (Sarkar, 2010).

Furthermore, due to a changing societal context and the increasing emphasis on the rights of the individual, including those with ID (e.g. 2010 Equality Act in the UK, and the 2006 UN Convention on the Rights of Persons with Disabilities at an international level), younger relatives may be more alert to stigma and also less willing to tolerate it. In this vein, younger family members in this study may perceive greater stigma in response to others' negative responses to challenging behaviours exhibited by the individual with ID. In addition, younger family members have had relatively less time to accustom themselves to the individual's diagnosis and disruptive behaviours, and as such may experience poorer quality of life. On the other hand, older parents may not only have had more time to adjust to the diagnosis and to find ways to manage challenging behaviours displayed by the individual, but additionally may have developed and adopted strategies to help them cope with others' negative attitudes and discrimination (Sarkar 2010).

The family lifecycle model (Carter & McGoldrick, 1989) suggests that the primary task for families in the fifth stage of the family lifecycle is to modify existing familial roles in order to incorporate young children into their lives. Problems in doing so may result in difficulties. Families of a child with a diagnosis of ID potentially have to deal with more changes than other families, and thus their physical and psychological wellbeing may suffer. Furthermore, individuals in their twenties have yet to emerge from adolescence as mature adults and often do not attain such maturity until their thirties (Fleming, 2004). Such identity exploration occurs against the background of attempts to negotiate one's social roles (Erikson 1980; Robinson 2015). This is a complex process that can lead to a "quarter-life" crisis, making younger adults more vulnerable to mental health issues (Robinson, 2015). Therefore, younger parents may be more susceptible to cognitive affiliate stigma as they are also more likely to be vulnerable to feelings of incompetence related to their ability to parent the individual with ID in the face of perceived family stigma. It should be noted however that in this study, family members below the age of 54 (*n* = 295) made up a larger proportion of the total sample compared to those over 55 (*n* = 112). Therefore, conclusions need to be drawn with caution.

13.3 The Relationship between Perceived and Affiliate Stigma

The contribution of perceived family stigma was particularly high for cognitive affiliate stigma compared to the other two aspects of affiliate stigma, suggesting that it may be most affected by the awareness that others perceive the individual with ID and their family members negatively. Cognitive affiliate stigma involves the endorsement of negative beliefs that one is incompetent as a carer

(Chang et al., 2015). Thus awareness of the stigmatising glances of others may lead some carers to gradually perceive themselves as less worthy or somehow flawed. Moreover, when perceived family stigma was controlled for in the final regression, only caregiver age and subjective wellbeing remained significant predictors of cognitive affiliate stigma. The awareness that leads to self-evaluation may therefore be affected by the age of the family member and their quality of life.

Caregiver burden emerged as a contextual factor in most components of affiliate stigma except for cognitive affiliate stigma, even with the inclusion of caregiver and cared for characteristics in the final regression. Similar findings were reported by Mak and Cheung (2008), who found that higher levels of affiliate stigma were associated with more subjective burden and fewer positive perceptions of care-giving, even after controlling for caregiving stress and demographic factors. Werner and Shulman (2015) also reported caregiving burden to be the chief contributor to affiliate stigma. Interestingly, burden was not associated to perceived family stigma in this study and its effects only diminished for cognitive affiliate stigma when controlling for perceived family stigma. Instead, it seemed a more prominent variable in the actual internalisation of stigma. Moreover, burden was most strongly associated with the affective dimension of affiliate stigma as compared to the behavioural dimension, suggesting that it plays an important part in feelings of shame, despair and embarrassment that may result in an internalisation of a stigmatised status by the carer. Indeed, stigma and discrimination faced by family members of those with ID have been shown to create substantial emotional burden, which is defined as the challenges faced by carers due to the emotions associated with caregiving (Chang et al., 2017; Kwok et al., 2014). Previous research has also indicated that emotional burden was linked to anxiety and depression in mothers, which affected their overall quality of life (Zhang & Yi, 2011). Facing the embarrassment caused by one’s son or daughter displaying behaviours that attract attention and challenge social and cultural norms in addition to the consequent stigma may result in emotional and psychological distress in carers (Green, 2003). These findings suggest that the pathway to affective affiliate stigma and emotional burden may involve related mechanisms. Accordingly, interventions aimed at tackling affiliate stigma may need to involve assessing and reducing the impact of caregiving burden experienced by family members of individuals with ID.

The lack of findings related to social support was surprising, given that previous studies have shown it to be a consistent protective factor against affiliate stigma (e.g. Ali et al., 2012; Shin et al., 2006). With increasing awareness of ID stigma, it could be that family carers in this study are increasingly drawing from their own internal psychological resources to build resilience against the adverse effects of family stigma. Cantwell et al. (2015) found that perceived emotional support had a protective effect against depression from caregiver stigma at low to moderate levels of self-esteem, high levels of self-esteem alone protected psychological health whether perceived emotional support was low or high. Additionally, Yang (2015) reported that family carers may manage the effects of affiliate stigma through defining themselves in relation to multiple affirmative social identities, instead of defining themselves solely in relation to the individual with ID. Another internal coping strategy, self-compassion, defined as ‘a caring and compassionate attitude toward oneself in the face of hardship or perceived inadequacy’ (Wong et al., 2016, p.2), has also been cited as being beneficial in emotional regulation and alleviating cognitive anxiety arising as a result of affiliate stigma (Yang, 2015; Wong et al., 2016).

14. Limitations and Future Directions

This study possessed several strengths, such as a good sample size, but potential limitations require mention. First, it is possible that face-to-face administration of the survey may have been preferable to online administration. The sensitive nature of the experience of caregiving and stigma may have warranted a detailed explanation of the survey and an opportunity for family members completing the survey to allay their concerns regarding participant. Relatedly, the choice of the Internet may introduce self-selection and dropout biases associated with Internet research, suggesting that generalisation of results may be unclear (Eysenbach & Wyatt, 2002). Also carers from non-white ethnic backgrounds are perhaps less likely to use the Internet and English may not be their first language. However, it is also possible that an online survey permitted anonymity, which could facilitate disclosure (Eysenbach & Wyatt, 2002). Indeed, some researchers believe that online surveys might be a more appropriate method compared to personal administrations when it comes to sensitive issues (Kays, Keith & Broughal, 2013). Future research should attempt to employ heterogeneous methods to ascertain whether distinct methodologies result in different findings about the experiences of family stigma of individuals with ID.

A second limitation to be noted is that the significance value was adjusted for individual tests but not across all tests, which means that some of the findings regarded as significant may actually be relatively spurious. Third, the original CBI was adapted for the study in two ways. While the original version was administered as a face-to-face interview, it was presented in the form of an online survey, and it was shortened. Hence, it cannot be assumed that the adapted version of the CBI has similar psychometric properties to the original one (Streiner, Norman & Cairney, 2015). For the purposes of studies adopting this simplified version, researchers may need to consider the reliability and validity of the measure in specific populations.

Fourthly, carers from White ethnic backgrounds made up 91.6% of the sample compared to approximately 10% from BME backgrounds. According to the Office for National Statistics in the 2011 Census, 80% of the population in England and Wales were white, while other groups comprised 12.8%. The study sample was therefore not particularly representative of the UK population.

Lastly, some individuals with ID had more than one primary diagnosis, were awaiting or were unsure of confirmed additional diagnoses of the individual with ID. Moreover, these were based on carers' subjective reports rather than an objective diagnosis from a clinician. Future studies should aim to use multiple sources of information in order to arrive at a more accurate diagnostic profile of the individual.

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Part 3: Critical Appraisal

16. Introduction

This critical appraisal aims to further explore the process and challenges of undertaking research on the experience of stigma among family members of individuals with intellectual disabilities (ID). First, it will begin with a discussion of what attracted me to this area of research, and offer reflections on the development of the FAMSI. Second, the process and challenges of pursuing a nationwide recruitment strategy will be considered, followed by a brief discussion of qualitative feedback from family members. Third, future research directions and implications of the present findings for research and clinical practice will be explored. Finally, concluding remarks and personal reflections will be provided.

17. Research Rationale

I became interested in stigma faced by family members during my work experiences with parents of children with ID. These children were frequently subjected to discrimination, which often extended to their parents. In a collectivistic country like Singapore, ‘face value’, which signified one’s social self-esteem and aspiration to be valued in social settings, is tightly connected to one’s identity (Ting-Toomey & Kurogi, 1998). Awareness of ID is limited, therefore parents were often judged and blamed for simply having a child who had a condition that deviated from social norms.

In such a stigmatising context, the needs of family carers often went unheard. In my interactions with them during my pre-training work, family members talked about experiences of stigma and a sense that their problems were overshadowed by the needs of the individual with ID. Many stayed at home, giving up their careers and avoiding social events for fear of others mentioning their child in conversation. Contact with members of the public during ID awareness campaigns confirmed these views, giving me first-hand experience of negative sentiments not merely towards individuals with ID, but also their family carers. My desire to make the voices of family carers heard and my increasing awareness of the repercussions of the lack of appropriate carer support for those giving care to individuals with ID motivated me to carry out a research project with the potential to positively affect the provision of support to such family members.

18. Measure Development

18.1 A New Scale

Initially, I aimed to better understand the experience of affiliate stigma in family carers in the UK through quantitative research in order to make recommendations for interventions. My preliminary literature search on this topic led to the discovery that existing measures of courtesy and affiliate stigma, such as the Affiliate Stigma Scale (Mak & Cheung, 2008) and the Stigma by Association Scale (Pryor, Reeder & Monroe, 2012), would be unsuitable for use with family carers in the UK due to the numerous negative connotations of the items. In particular, the only existing Affiliate Stigma Scale (Mak & Cheung, 2008) specific to ID, which was developed in Hong Kong, presents caring for someone with ID in a rather negative light and paints a bleak picture of the lives of family carers.

Based on these findings, I initially set out to validate the Affiliate Stigma Scale in the UK population. Following numerous discussions with my supervisors however, it became evident that the scale would not be well received even with substantial modifications. Therefore, I decided instead to devise a new measure, one that would acknowledge positive aspects of caregiving and be suitable for use with an ethnically diverse population.

18.2 The Concept of Affiliate Stigma

The first stage of measure development entails developing a theoretical definition of the key variable, which involves referencing other studies that have measured the same or similar variables (Korb, 2012). This was the primary obstacle I faced in the development of the new scale as it proved challenging to define affiliate stigma. Only by defining a construct can we operationalise it for consistent measurement. A preliminary search of the literature led to the discovery that there was no coherent definition of the term. Notably, various terms were used interchangeably, most of which did not quite do justice to the experience of affiliate stigma. Moreover, there was merely an assumption that most family members were aware that such stigma exists, which is then experienced and internalised by them. This made me question the validity of current measures and the efficacy of current interventions for family carers of those with ID in addressing stigma experiences of the family. For example, the NICE guidelines for support and interventions for family members do not specifically include support to manage stigma (NICE, 2015).

In the first stage of my thesis, even I grew increasingly confused about the experience of stigma in family members of individuals with ID, but my initial misgivings were confirmed by the systematic literature review presented in Part 1 of this thesis. I realised that an awareness of stigma, labelled ‘perceived family stigma’ in the empirical paper presented in Part 2, is in fact a different process from affiliate stigma, which is the actual process of internalisation of stigma.

19. Recruitment and Data Collection

19.1 Nationwide Recruitment Strategy

The process of recruitment in research commences with knowing the target population. Testing the psychometric properties of a new measure that is suitable for the UK context meant that I needed a large, ethnically diverse sample of UK residents. The key to conducting such a study successfully was to engage the community in a way that built positive and lasting community networks (National Institute of Mental Health, 2005).

Recovering from the shortfalls of recruitment can be challenging as the study proceeds (Institute of Translational Health Sciences, 2017). Therefore, I devised a detailed recruitment strategy identifying the organisations that I wanted to engage, mainly organisations and support groups for individuals with ID as well as their families across the UK. Developing a collaborative partnership with Mencap was the pivotal first step in this strategy. Contact was established over email and study information was provided. Given that the study had a close fit with one of the their strategic priorities for the period 2015-2020, namely a focus on attitudes towards people with ID (and their family carers), Mencap agreed to disseminate the study via their social media channels.

The next step involved the creation of a database collating the contact details of all organisations in the Mencap database, as well as other services discovered through Google searches and word-of-mouth. Given that I physically resided in London, contact was established through email and/or telephone. To ensure that each organisation was contacted and there was no replication, I set up a recruitment database to track and monitor the number of emails (including reminders, follow-ups and updates) sent, respective responses and agreed outcomes for each site. Organisations that did not respond were sent two reminder emails before they were considered as having declined to participate. When organisations did agree to participate in the project, it was occasionally necessary to make further contact them over the telephone. It was also vital for me to identify a key liaison person from each organisation with whom regular communication was established and maintained.

The third step occurred in tandem, whereby I prepared information materials for the intended audience, as both print and electronic versions. I started out with the basic study materials, including the participant information sheet, the survey itself, email templates, advertisements and fliers. Eventually however, the mechanisms adopted to encourage recruitment grew to include newsletter articles and online posts as many recruitment sites provided opportunities to publicise the study on their websites, blogs, online community forums, newsletters and even social media sites. I designed posters from scratch and wrote articles to feature in such newsletters and social media sites, at times customising these even further to meet the needs of individual organisations. Of note, all these materials were closely aligned in contents and wording with study materials approved by the UCL Research Ethics Committee (REC).

As I was recruiting from multiple sites to ensure a large and diverse sample, it was essential for me to provide the coordinators and administrators of all sites with adequate information about the study and to allow them opportunities to ask questions. Moreover, once they had been provided with the relevant study materials, the decision to disseminate information about the study to their network of family carers was at the discretion of the respective organisation’s management.

Fourth, several barriers to participation warranted consideration. The first possible barrier concerns access to the online survey used for data collection. Internet-based research is cost-effective and time-efficient while allowing the researcher to access large groups of people that have similar attitudes and beliefs regarding an issue. It also offers greater anonymity, which may encourage participants to give honest answers about sensitive topics, and enables participants to forward survey links to others (Wright, 2006). However, such methods are limited to the extent that they can reach respondents who do have Internet access, which may lead to difficulties with sampling (Andrews, Nonnecke & Preece, 2003). Furthermore, it was impossible to calculate a response rate as it was uncertain how many family carers received the survey link. These disadvantages are relevant to the current research (Wright, 2006).

Indeed, some of the organisations in the further parts of the UK, such as Gloucestershire and Northern Ireland, reported that not all carers' households in more remote areas had Internet access. Moreover, some of the older family carers were not familiar with using the Internet. At least 11% of the households in the UK have no Internet access and only 38.7% of those aged 75 and over have used the Internet at least once in the last three months (Office of National Statistics, 2016). Even with access, speeds can be as poor as 1.30Mbps in rural areas, compared to the average speed of 25Mbps in London (Payton, 2016). This problem was address by posting physical copies of the survey to organisations that requested them. To minimise postage costs on their end, they agreed to scan the completed copies and email them to me. However, despite numerous follow-up emails, these organisations never returned the completed copies.

Another barrier was language. This could have been the reason the survey was not completed by more family carers from BME communities, especially those of South Asian ethnicity. Unfortunately, given the time and funding constraints, this barrier could not be adequately addressed, as translation into other languages was not possible. In future studies study, information in other languages should be included, as should data collection methods. Moreover, more attempts should be made to engage BME communities directly.

In the final stage of the recruitment strategy, a review of all sites was undertaken towards the end of the recruitment phase to ascertain that actions had been completed and desired outcomes achieved. Each site involved in the study was then closed and sent a thank you email.

Although tedious and time-consuming, it was an accomplishment to have devised a recruitment strategy at a national level in a short time. It gave me the opportunity to meet people who were advocates and passionate about having carers' voices heard, justifying the importance of seeking to advance our understanding of family stigma. Moreover, in order to validate the crucial role of the participants in research and reduce the prospect of participants feeling exploited (Fernandez, Kodish & Weijer, 2003), I agreed to disseminate the results of the study to participating organisations in the form of a handout.

19.2 Ethical Considerations

One of the biggest setbacks I faced during the course of this project was a complaint by one of the organisations that had agreed to take part in the study. As per the recruitment procedure, this organisation had been given the study material to vet before sending them out to their network of family caregivers, which the organisation's manager subsequently agreed to do. However, a carer in their network became distressed while completing the survey as they had found the survey content too negative –of note, their concerns appeared to centre particularly on Rosenberg's Self-Esteem scale (1965), one of the most widely used measures in psychological research. The carer concerned relayed this to the organisation's manager. It was only then the manager looked at the actual survey before proceeding to contact my supervisor in parallel with making a complaint directly to UCL REC about the survey. The organisation held the view that, given the sensitive nature of the topic, asking carers the questions in the survey was only suited to exploring via face-to-face qualitative research.

Although this matter was swiftly resolved by my supervisor and the UCL REC, it highlighted key issues faced by families of those with ID. The director of the organisation had felt that the survey trivialised the experiences of the family caregivers. Her reaction (and that of the carer who had originally made the complaint) ironically served to emphasise the relevance of conducting this study, leading me to realise the validity of my research. The family carer who had objected to the survey had done so as it had clearly evoked strong emotional reactions that were related to the experience of stigma in giving care to an individual with ID. The manager of the organisation had herself mentioned in a subsequent correspondence that it was erroneous for researchers to perceive the caregiving experience as a negative one, thereby portraying carers in a negative light. This in itself reinforced the need for research in the first place in order to better understand the caregiving process for family members of individuals with ID.

The families of individuals with ID have been monolithically conceived to endure inevitable suffering, distress and pathology (Glidden Valliere & Herbert, 1988). Recent research has suggested that this view does not hold (Kauffman, Hallahan & Pullen, 2017). Many families in fact consider the effects of their relatives with ID as positive and often even respond with resilience (Hastings, 2005; Helf & Glidden, 1998). Moreover, the current study approach was criticised for being callous and insensitive. Indeed, compared to their qualitative counterparts, quantitative research approaches have been regarded as superficial, often reducing meaningful lived experiences to numbers without regard of context (Choy, 2014). I was made aware of the value of qualitative research in examining topics that are delicately intertwined with the lives of individuals such as stigma. Such qualitative and phenomenological methods enable researchers to engage with participants in the construction of their stories while affording their experiences acknowledgement and validation (Dickson-Swift, 2006; Dickson-Swift, James, Kippen,& Liamputtong, 2007).

20. Feedback from Family Carers

To enable family carers to freely express their views, they were invited to provide comments at the end of the survey. A hundred and ten carers gave feedback. Although conducting a detailed thematic analysis was beyond the scope of this project, the feedback was collated into themes. The most pertinent themes with quotes are presented below.

20.1 The Carer as an Individual

On a personal level, carers raised important points about their identity as a caregiver for someone with ID. While some acknowledged the positive new identity they had adopted (“I don't think I ever laughed so hard, cried so many 'happy tears' or loved so freely as I do now. She's given me perspective and empathy as well as grey hair!”), others reported the emergence of a negative one, with some even mourning the loss of their old identity. For example, one carer said others saw them as “mythically being 'special' parents, pity and difficulties around exhaustion and the world of work”, while another described the process of being told that their child had a disability as “The impact of bringing a handicapped human being into the world is like grieving. There is real pain there; real loss. Only no one to turn to. It passes but like any shock it leaves you changed. Many people never get over it.”

Carers also revealed the impact caregiving had on their mental health. On one hand, many stated the adverse effects it had on their mental and emotional wellbeing: “You live life on a knife edge and the effects on family life are dramatic. Constant stress can lead to mental health issues”. On the other hand, many felt it was their very resilience that enabled them to cope with stresses: “The feeling of isolation and inability to cope that affects me from time to time has nothing to do with my own resilience”.

20.2 The Social Effects of Caregiving

It has been often noted in the literature that caregivers face negative reactions and stigma not just from members of the community, but also from their families and friends (Ali, Hassiotis, Strydom & King, 2012; Yang, 2015). Indeed, this was the case for a large proportion of the carers in this study. One carer said she was “accused of abuse [by my family] in having my son assessed and my father no longer acknowledges my wonderful son as his grandchild”. Another carer described unhelpful behavioural responses of others: “…so called friends and the public appear aloof and wary. And either stare, ignore her or move away.”

Such stigmatising responses frequently came from professionals involved in the care of the individuals as well. Family carers mentioned their experiences with stigma from professionals in different contexts, including health, social and educational settings, which evoked feelings of disappointment (“feel very let down by social services”) and injustice (“The locus of injustice is not in the individual, the family, or even the community; it's in the official manifestations of society as a whole”). Some even described being labelled as a troublesome parent and blamed for their queries about the provision of care for the individual with ID: “Health. Education. Social care. Housing. Being constantly framed as 'the troublemaker' for asking the questions that you know are 100% necessary to secure your relative's wellbeing, even their life…. we get blamed, over and over, by statutory services”. Such stigma was succinctly summed up by one of the carers: “[the] attitude of professionals to parents and how professionals feel and think and act around LD… Most professionals I (and the friends I have with LD relatives) meet are the major challenge”.

Often such stigmatising societal behaviour leads family members to withdraw from society (Ali et al., 2012; Ngo, Shin, Nhan & Yang, 2012). Indeed, social isolation and exclusion were noted among carers, sometimes even when they were surrounded by loved ones: “yes have friends and family but still feel isolated. Difficult to find other families to do social events with/holidays with”. Drawing on sources of support appeared to be protective from some of these ill effects of stigma in some instances but at other times the available social resources were inadequate: “…been to carer support groups but find them difficult… find other carers off load their problems and I am not able to share mine”.

20.3 Economical/Political Challenges Faced

Government funding, or rather the perceived inadequacy of it, was a key challenge raised by many carers. Only 7.7% of the UK Budget is dedicated to health services in the NHS and of that, a mere 13% goes to mental health (Department of Health, 2017). Further cuts are expected, leading one carer to remark: “No government is willing to invest the money for the care of our most vulnerable in society”. The economical strain of such cuts as well as the inability to work in order to provide full-time care consequently led to an increased caregiving burden for most as there were “financial restrictions on earnings because of providing support [to the individual].”

20.4 Research Implications

The option to provide open ended comments as part of the survey allowed carers to express their feelings towards the survey and also suggest some directions future research might take. Many voiced their gratitude and appreciation for researchers showing interest in their caregiving experiences, some even finding it “very thought provoking”. Moreover, they were hopeful that the findings would be shared, one even explicitly stating, “I hope you can make your findings public to raise awareness of the carers who look after people with LD, they are a very hidden part of the community”. However, the survey induced negative feelings in some. Carers spoke about the survey being too general and too “basic for such a complex subject”, giving “a superficial impression of caring for a dependent”. Even though I had aimed to capture the positive aspects of caregiving, a number of carers still felt that it “in no way collects this positive experience” and found that it took “a slightly condescending professional approach”. Moreover, there was disappointment about there being no translation of research in general into tangible benefits for carers: “surveys do not change anything - at all. We are not personally seeing improvements”.

In terms of future directions, many of the carers highlighted the variable nature of the stigma they experienced, depending on contextual factors. For example, some carers found that their feelings toward the individual with ID “varied around my age and all of our circumstances” or that the passage of time had allowed them to be more accepting of the individual’s disability and therefore cope with stigma better. Others cited changes in medication or life transitions as affecting the views they had towards the individual over time. Clearly, using longitudinal mixed methods approaches could help address this, which was another point highlighted by carers. Some requested more space to explain certain sections, while others even suggested “a companion to a survey could be a short 'day in the life of...' narrative which would allow people to be more expansive in their responses… difficult to analyse, but perhaps more revealing”.

20.5 Clinical Implications

The endorsement of a negative identity could have developed due to role engulfment and losing a sense of self, which occurs when the activities and behaviours that used to define a person have been overshadowed by the responsibilities of caring (Eifert, Adams, Dudley & Perko, 2015). The new caregiver role may replace other important identities and become the dominant one (Eifert et al., 2015). Family carers who perceived caregiving as a normal and natural expansion of their former role might have adopted a more positive identity. Yang (2015) showed that some stigmatised family caregivers of individuals with ID develop multiple identities. They switch between these to guard their self-concept and mental health against the stigmatised identity and to develop self-efficacy at their achievements (Yang, 2015). Effective interventions to support caregivers therefore need to take into consideration the multi-dimensional nature of their needs. The focus is often on the individual with ID but it is clear that caregivers themselves often require individualized plans to maintain their own wellbeing. Clinicians may first need to understand the extent the carer identifies with the caregiver role using an instrument such as the Family Caregiver Identity Scale (Eifert, 2014). This could be used as an accompaniment to the FAMSI in order to match existing resources most relevant to them and foster resilience. Psychotherapy and psychoeducation have been found to be particularly effective interventions for caregivers (Sörensen, Pinquart, & Duberstein, 2002). Psychoeducational awareness programmes may even need to be extended to relevant members of the caregivers' social setting, including other family members, friends, the community and even other professionals, to raise awareness of their roles as caregivers.

20.6 Broader Research Approaches

The feedback provided by family carers certainly revealed a lot more about their caregiving experiences beyond the survey, emphasising the importance of capturing their stories through the richer and more in-depth means offered by qualitative methods and often missed by quantitative approaches. When investigating a phenomenon as complex as stigma experienced by family carers, using a combination of the two would enable researchers to obtain a broader perspective of caregiver stigma. Such mixed-methods designs can create a stronger theory of family caregiver stigma by strengthening the shortfalls of either research approach (Creswell, 2013). In addition, given the variability of stigma, longitudinal studies using a mixed-methods approach can be effective in examining patterns and long-term changes in family carer stigma over time (Teti, 2008).

20.7 Structural Stigma

Structural stigma signifies the inequities and injustices characteristic of social structures which constrain the resources and freedoms of a certain population (Link & Phelan, 2001). Caregivers revealed stigmatising experiences not merely at an interpersonal level, but also at a structural one. While the FAMSI may be suitable to use in evaluating caregiver stigma experiences with the general public, their experiences specifically with professionals indicate that the scale may need to be modified to assess structural in addition to interpersonal stigma. Measuring such structural stigma may indeed be the starting point to develop legal and policy interventions to protect stigmatised caregivers and ensure recognition of the extent of their caregiving roles. This may be particularly pertinent in addressing funding issues and financial burden faced by family carers.

20.8 Translation of Research

A significant challenge to improving the experiences of caregivers is the inadequate translation of research findings into sustainable outcomes. One way to facilitate this process would be through the use of a community-engaged research framework. Such research involves collaboration between the researcher and community partners with the aim of contributing to existing research and strengthening the welfare of the community (Ahmed & Palermo, 2010). For example, Bazzano et al. (2013) demonstrated that a community-based mindfulness-based stress reduction programme was an effective intervention to decrease stress and enhance mental wellbeing for caregivers of children with developmental disabilities. In the study, partnerships between parents/caregivers and researchers informed programme development, recruitment, execution and evaluation (Bazzano et al., 2013). An extension of this to family stigma may acknowledge and validate the experiences of caregivers while enabling the development of research that is reactive to their needs. Moreover, community-engaged research can facilitate the sharing of findings with community partners, a point that was emphasised by the family carers in this study.

21. Conclusions and Personal Reflections

In summary, this study has significantly contributed to the existing research. The analyses conducted confirmed the psychometric reliability and validity of the FAMSI, a measure of the experiences of stigma faced by families of individuals with ID. The present findings, together with the valuable qualitative feedback provided by carers, also indicated that interventions for families of people with ID might need to first target underlying family stigma in order to adequately address the various practical and psychological difficulties faced by them. It is clear that the focus of their caregiving experiences requires a paradigm shift from viewing their lives as being wrought with obstacles to recognising their value as advocates of individuals with ID. The aim of researchers and clinicians should be to empower them and celebrate their contributions in the lives of those with ID. As succinctly summed up by one carer, “Listen to us. Respect us. Value us. Only then can you actually help us”.

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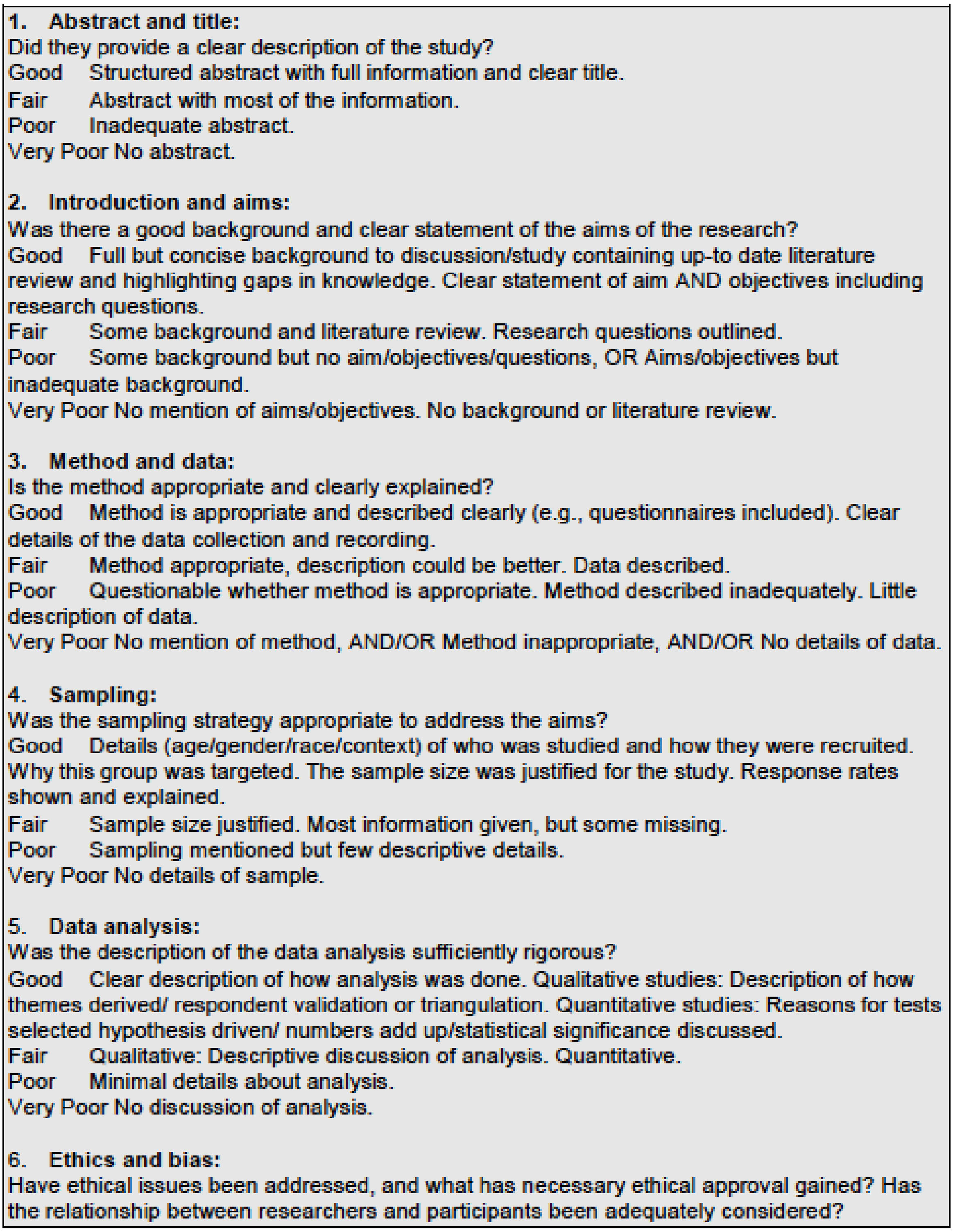
Appendices

Appendix A

Quality Appraisal Tool from Hawker et al. (2002).



1. Abstract and title:
Did they provide a clear description of the study?
Good Structured abstract with full information and clear title.
Fair Abstract with most of the information.
Poor Inadequate abstract.
Very Poor No abstract.

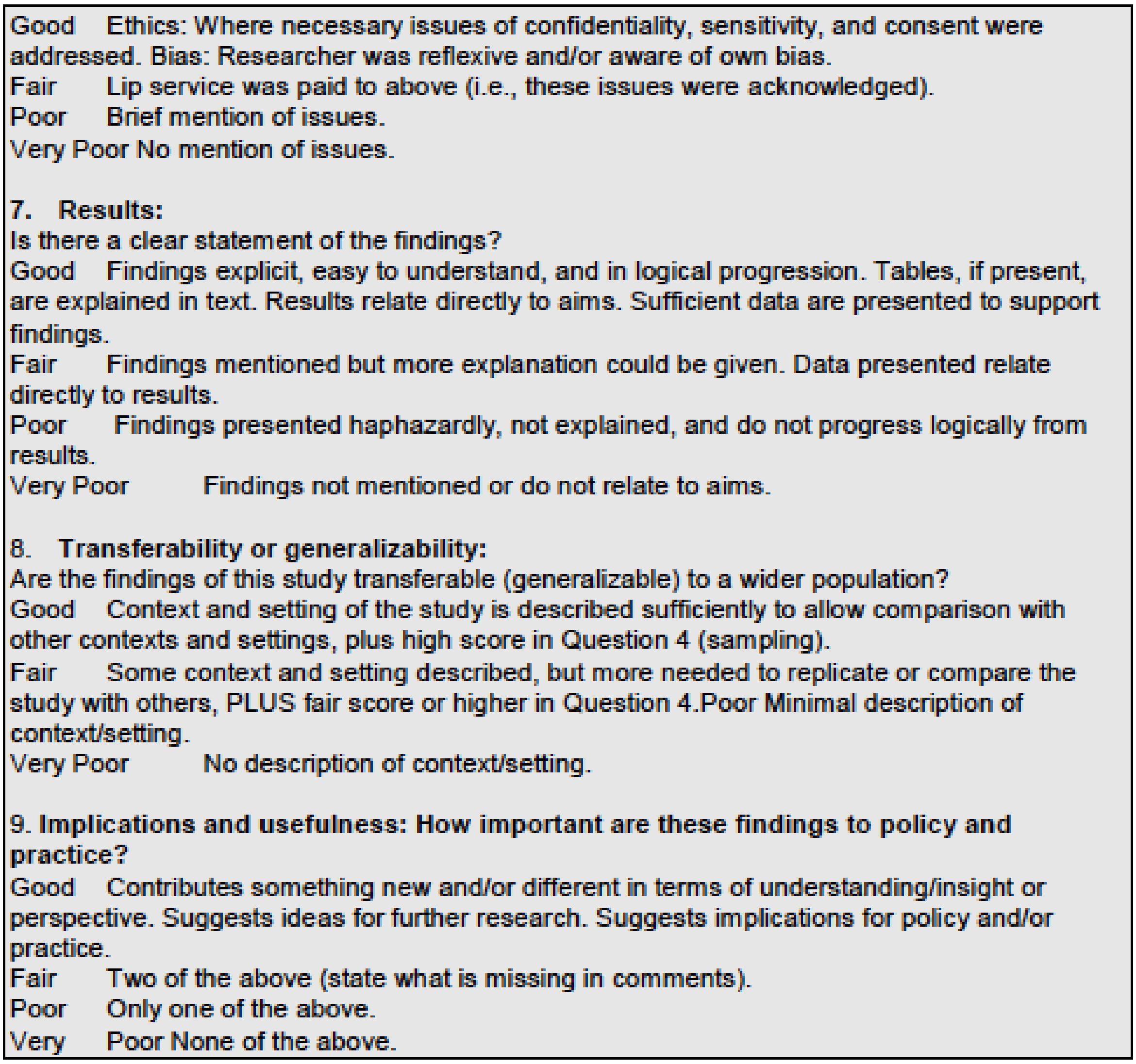
2. Introduction and aims:
Was there a good background and clear statement of the aims of the research?
Good Full but concise background to discussion/study containing up-to date literature review and highlighting gaps in knowledge. Clear statement of aim AND objectives including research questions.
Fair Some background and literature review. Research questions outlined.
Poor Some background but no aim/objectives/questions, OR Aims/objectives but inadequate background.
Very Poor No mention of aims/objectives. No background or literature review.

3. Method and data:
Is the method appropriate and clearly explained?
Good Method is appropriate and described clearly (e.g., questionnaires included). Clear details of the data collection and recording.
Fair Method appropriate, description could be better. Data described.
Poor Questionable whether method is appropriate. Method described inadequately. Little description of data.
Very Poor No mention of method, AND/OR Method inappropriate, AND/OR No details of data.

4. Sampling:
Was the sampling strategy appropriate to address the aims?
Good Details (age/gender/race/context) of who was studied and how they were recruited. Why this group was targeted. The sample size was justified for the study. Response rates shown and explained.
Fair Sample size justified. Most information given, but some missing.
Poor Sampling mentioned but few descriptive details.
Very Poor No details of sample.

5. Data analysis:
Was the description of the data analysis sufficiently rigorous?
Good Clear description of how analysis was done. Qualitative studies: Description of how themes derived/ respondent validation or triangulation. Quantitative studies: Reasons for tests selected hypothesis driven/ numbers add up/statistical significance discussed.
Fair Qualitative: Descriptive discussion of analysis. Quantitative.
Poor Minimal details about analysis.
Very Poor No discussion of analysis.

6. Ethics and bias:
Have ethical issues been addressed, and what has necessary ethical approval gained? Has the relationship between researchers and participants been adequately considered?



Good Ethics: Where necessary issues of confidentiality, sensitivity, and consent were addressed. Bias: Researcher was reflexive and/or aware of own bias.
Fair Lip service was paid to above (i.e., these issues were acknowledged).
Poor Brief mention of issues.
Very Poor No mention of issues.

7. Results:
Is there a clear statement of the findings?
Good Findings explicit, easy to understand, and in logical progression. Tables, if present, are explained in text. Results relate directly to aims. Sufficient data are presented to support findings.
Fair Findings mentioned but more explanation could be given. Data presented relate directly to results.
Poor Findings presented haphazardly, not explained, and do not progress logically from results.
Very Poor Findings not mentioned or do not relate to aims.

8. **Transferability or generalizability:**
Are the findings of this study transferable (generalizable) to a wider population?
Good Context and setting of the study is described sufficiently to allow comparison with other contexts and settings, plus high score in Question 4 (sampling).
Fair Some context and setting described, but more needed to replicate or compare the study with others, PLUS fair score or higher in Question 4.Poor Minimal description of context/setting.
Very Poor No description of context/setting.

9. **Implications and usefulness: How important are these findings to policy and practice?**
Good Contributes something new and/or different in terms of understanding/insight or perspective. Suggests ideas for further research. Suggests implications for policy and/or practice.
Fair Two of the above (state what is missing in comments).
Poor Only one of the above.
Very Poor None of the above.

Appendix B
UCL Ethical Approval

DIVISION OF PSYCHOLOGY AND LANGUAGE SCIENCES

Ethics Application Form for Non-Invasive Research on Healthy Adults

SECTION A APPLICATION DETAILS

A1 Project details

Project title: **Intellectual Disability Stigma: Understanding its Impact and Developing Innovative and Effective Interventions**

Date of submission: 10.2.2016

Proposed start date: 1.3.2016

Proposed end date: 30.7.2021

A2 Principal researcher

(Note: A student – undergraduate, postgraduate or research postgraduate – cannot be the principal researcher for ethics purposes).

Full name: Dr Katrina Scior

Position held: Senior Lecturer

Research Department: Clinical, Educational and Health Psychology

The principal researcher must read and sign (electronic signature or scanned pdf with signature are acceptable) the following declaration. Please tick the box next to each of the statements below to acknowledge you have read them and provided all required information.

I will ensure that changes in approved research protocols are reported promptly and are not initiated without approval by the Departmental Ethics Committee, except when necessary to eliminate apparent immediate hazards to the participant.	√
I have completed a risk assessment for this programme of research and hereby confirm that the risk assessment document will be discussed with any researcher/student involved in this programme of research (currently or in the future). I will ensure that all researchers/students sign the risk assessment form following this discussion. **Risk assessment forms for projects can be downloaded from the Ethics section of the PaLS Intranet.**	√
I have obtained approval from the UCL Data Protection Officer stating that this research project is compliant with the Data Protection Act 1998. My Data Protection	√

Registration Number is: Z6364106/2016/01/46 You can find a data protection registration form here: http://www.ucl.ac.uk/efd/recordsoffice/data-protection/	
I have included examples of the Information Sheet and Consent Form for the proposed research. It will be made clear to the participants that they can withdraw from the study at any time, without giving a reason.	√
I will ensure that all adverse or unforeseen problems arising from the research project are reported in a timely fashion to the UCL Research Ethics Committee.	√
I will undertake to provide notification when the study is complete and if it fails to start or is abandoned.	√
I have met with and advised students on the ethical aspects of this project/programme of research.	√
I am satisfied that the proposed research complies with current professional, departmental and university guidelines.	√

[redacted]

Signature: **Date: 10/02/2016**

A3 Contact details

Principal Researcher

Full name: Dr Katrina Scior

Position held: Senior Lecturer

Research Department: Clinical, Educational and Health Psychology

Email: k.scior@ucl.ac.uk Telephone: 0207 679 1845

Additional applicant

Full name: Natasha Mitter

Position held: Doctoral Student (DClinPsy)

Research Department: Clinical, Educational and Health Psychology

Email: natasha.mitter.14@ucl.ac.uk

A4 Approval from the Departmental Ethics Committee

(Approval cannot be given by the principal researcher of this project – if necessary the application must be sent to an Ethics Officer from a different Research Department, or to the College Ethics Committee, for approval)

Declaration by the Research Department Ethics Chair:

I have reviewed this project and I approve it. ☐ YES

The project is registered with the UCL Data Protection Officer and a formal signed risk assessment form has been completed.

Allocated Departmental Project ID Number for the approved application:

CEHP/2016/551

Name of the Research Department Ethics Chair (type in): John King

Date: 18/03/2016

Appendix C

Email to sent to experts and ID organisations

Dear Sir/Madam,

We are undertaking a study to understand family members' experiences of and perceptions of other people's attitudes to learning disability, and more specifically towards family members' themselves as the parent/sibling or close relative of someone with a learning disability. As I'm sure you'll be fully aware, many things are assumed about caregivers' perceptions but research that actually asks them directly is thin on the ground. For this reason, we are looking to hear directly from family members of individuals with learning disabilities (children as well as adults) – they will be asked to complete a survey that will take approximately 20 minutes. The study has formal ethical approval from University College London's Research Ethics Committee.

As this study has a close fit with Mencap's Changing Attitudes priority and we have their endorsement for this, we very much hope you would be able to disseminate information about it.

We'll be happy to provide more details, which will include a draft email to family carers and an ad. Alternatively, please feel free to contact us:

Email - natasha.mitter.14@ucl.ac.uk
Contact No. - 07784 297380

Yours Sincerely,

Dr Afia Ali | Dr Katrina Scior | Natasha Mitter
Senior Clinical Lecturer | Senior Lecturer | Trainee Clinical Psychologist

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Research Dept of Clinical, Educational and Health Psychology
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e-mail: natasha.mitter.14@ucl.ac.uk
Tel: 0207-6791845
Social Media Tag: #LDhaveyoursay

Also: Centre for Research in Intellectual & Developmental Disabilities: www.ucl.ac.uk/ciddr

Appendix D

Study information for ID organisations for family carers

Suggested Subject: Family Carers of Individuals with LD

Suggested Email/Message:

Dear parents/siblings and other relatives of individuals with learning disabilities,

Our research group at University College London (UCL) is looking to gain a better understanding of family members' experiences of caregiving, with a particular focus on perceptions and experiences relating to others' attitudes to learning disability. There has been quite a lot of research on attitudes to people with learning disabilities among professionals and the general public. There has been very little research though on family members' perceptions of others' attitudes and behaviours, both to their family member with a learning disability and to them as parent/sibling/aunt or uncle etc. In order to gain a better understanding of this we would be grateful if you would consider completing a survey. Your responses will be anonymous and completion of the survey on average will take 15 to 20 minutes. You will be asked about positive and negative experience you've had as well as about broader aspects of caring for/supporting your family member. There will be some additional questions about your family members' needs to help us put your responses into context. There is also a chance to enter a prize draw upon completion of the survey.

We are looking to hear from family members of children as well as adults with learning disabilities. The study has formal ethical approval from University College London's Research Ethics Committee.

Have your say by accessing the survey at this link: **https://uclpsych.eu.qualtrics.com/SE/?SID=SV_37CMEb2ZdwYusXH**

We've also attached an ad for the survey and would be grateful if you could share this with anyone else who may be interested in taking part.

We thank you in advance for your help with this research.

Yours Sincerely

Afia, Katrina & Natasha

Dr Afia Ali Senior Clinical Lecturer	Dr Katrina Scior Senior Lecturer

Natasha Mitter
Trainee Clinical Psychology

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Twitter & FB Tag: #LDhaveyoursay

Also: Centre for Research in Intellectual & Developmental Disabilities: www.ucl.ac.uk/ciddr

Appendix E

Study poster

Do you have a family member with learning disabilities? Have your say!

Many things are assumed about caregivers' perceptions & attitudes regarding caregiving for someone with LD.

Now it's time to hear your perceptions of caregiving & other attitudes. #LDhaveyoursay

If you care for such a family member, our research team would like to hear from you. We'r conducting a survey online and looking specifically for *family members who are 18 years and older.*

Your views will help greatly in understanding caregivers' experiences. Simply copy and paste the following link:

https://uclpsych.eu.qualtrics.com/SE/?SID=SV_37CMEb2ZdwYusXH

Call at 07784297380 or email natasha.mitter.14@ucl.ac.uk for more information. #LDhaveyoursay

Appendix F

The 26-item Family Stigma Instrument (FAMSI)

(A) To what extent do you agree that some people might respond in the following ways towards a family member of someone with LD? (Note: here we are not necessarily asking about your personal experiences but rather what you may have seen or heard regarding how some people respond to the family members of people with LD). The questions are framed as such: Some people might . . . , where "them" or "their" refers to the family of someone with LD.

Some people might…

	Strongly disagree	Somewhat disagree	Neither agree nor disagree	Somewhat agree	Strongly agree
1) ... feel embarrassed about associating with them.	○	○	○	○	○
2) … feel uncomfortable about going to their house.	○	○	○	○	○
3) … treat them more negatively.	○	○	○	○	○
4) … think that the family has done something wrong because of them.	○	○	○	○	○
5) … behave negatively towards them when they are with the person with LD in public.	○	○	○	○	○
6) … avoid making friends with them.	○	○	○	○	○
7) … not want to hear about any of their problems.	○	○	○	○	○
8) … not invite the family to social events.	○	○	○	○	○

Please Turn Over

(B) To what extent do you agree that caring for your family member with LD has changed you in the following aspects?

Caring for my family member with LD has…

	Strongly disagree	Somewhat disagree	Neither disagree nor agree	Somewhat agree	Strongly agree
9) … enabled me to develop a more positive attitude toward life.	○	○	○	○	○
10) … made me feel needed.	○	○	○	○	○
11) … strengthened my spirituality and faith.	○	○	○	○	○
12) … allowed me to form friendships with others in a similar situation.	○	○	○	○	○
13) … made me feel that I make a positive contribution to society.	○	○	○	○	○
14) … strengthened some of my relationships with family/friends.	○	○	○	○	○

Please Turn Over

(C) To what extent do you experience the following responses towards your family member with LD? The questions are framed as such: I feel/am/avold . . . , where “them” or “their” refers to your family member with LD.

I feel…

	Strongly disagree	Somewhat disagree	Neither agree nor disagree	Somewhat agree	Strongly agree
15) … embarrassed about them (my family member with LD).	❍	❍	❍	❍	❍
16) … distressed about being associated with them.	❍	❍	❍	❍	❍
17) … guilty about having them in the family.	❍	❍	❍	❍	❍
18) … uncomfortable when I have friends over because of them.	❍	❍	❍	❍	❍

Please Turn Over

I am…

	Strongly disagree	Somewhat disagree	Neither agree nor disagree	Somewhat agree	Strongly agree
19) … treated differently by some people when I am with them.	❍	❍	❍	❍	❍
20) … excluded from activities when other people find out about their ID.	❍	❍	❍	❍	❍
21) … aware of how some people look at me when I am out them.	❍	❍	❍	❍	❍
22) … treated differently by some people because of them.	❍	❍	❍	❍	❍

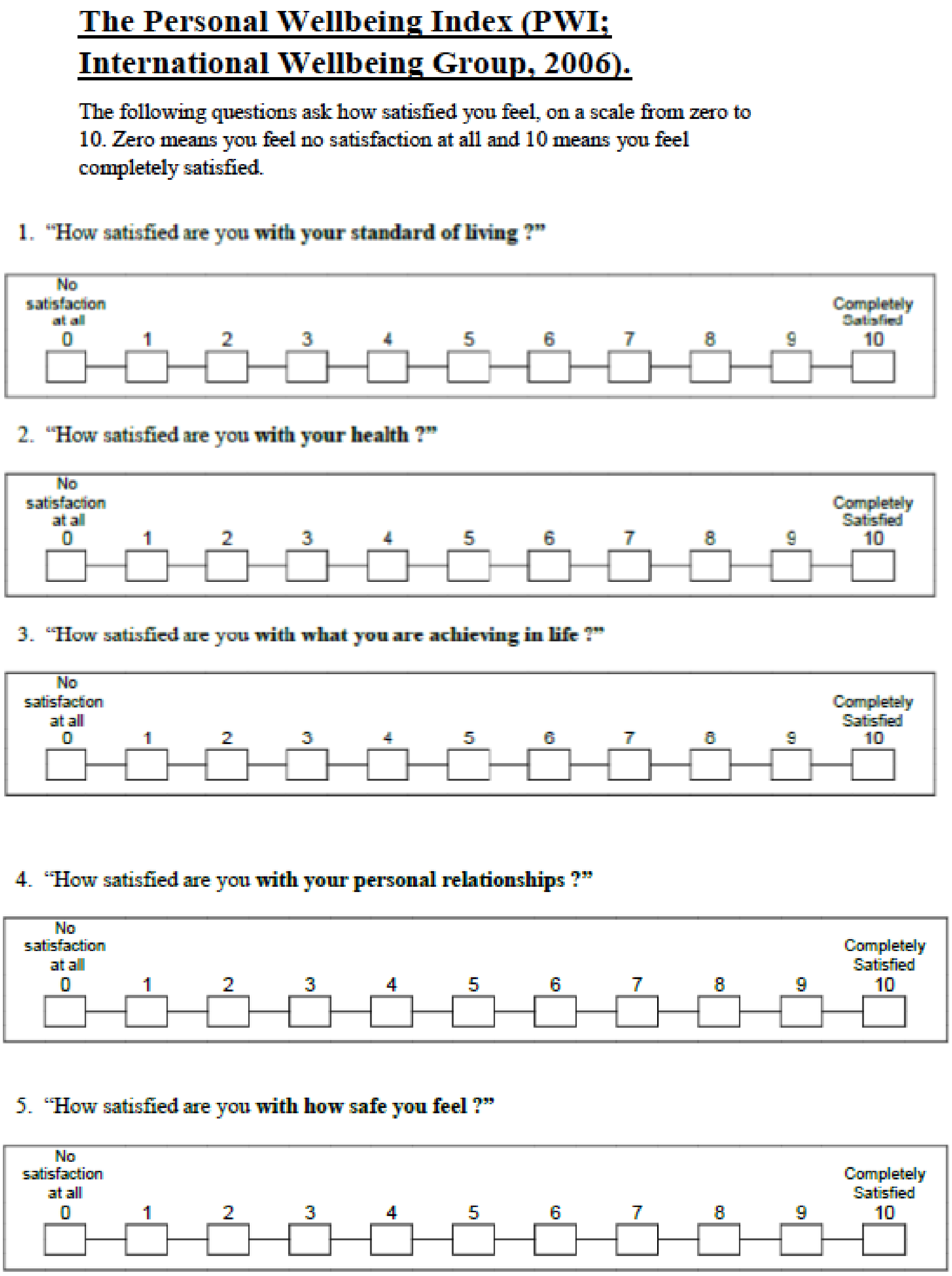
Please Turn Over

I avoid…

	Strongly disagree	Somewhat disagree	Neither agree nor disagree	Somewhat agree	Strongly agree
23) … introducing my friends to them (my family member with LD).	○	○	○	○	○
24) … telling people that I am related to them.	○	○	○	○	○
25) … making new friends because of them.	○	○	○	○	○
25) … being seen with them.	○	○	○	○	○

Appendix G

The Personal Wellbeing Index (PWI; International Wellbeing Group, 2006)



The Personal Wellbeing Index (PWI; International Wellbeing Group, 2006).

The following questions ask how satisfied you feel, on a scale from zero to 10. Zero means you feel no satisfaction at all and 10 means you feel completely satisfied.

1. "How satisfied are you **with your standard of living** ?"

No satisfaction at all										Completely Satisfied
0	1	2	3	4	5	6	7	8	9	10
☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

2. "How satisfied are you **with your health** ?"

No satisfaction at all										Completely Satisfied
0	1	2	3	4	5	6	7	8	9	10
☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

3. "How satisfied are you **with what you are achieving in life** ?"

No satisfaction at all										Completely Satisfied
0	1	2	3	4	5	6	7	8	9	10
☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

4. "How satisfied are you **with your personal relationships** ?"

No satisfaction at all										Completely Satisfied
0	1	2	3	4	5	6	7	8	9	10
☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

5. "How satisfied are you **with how safe you feel** ?"

No satisfaction at all										Completely Satisfied
0	1	2	3	4	5	6	7	8	9	10
☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

6. "How satisfied are you **with feeling part of your community ?"**

No satisfaction at all										Completely Satisfied
0	1	2	3	4	5	6	7	8	9	10
☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

7. "How satisfied are you **with your future security** ?"

No satisfaction at all										Completely Satisfied
0	1	2	3	4	5	6	7	8	9	10
☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

[Optional item]

8. "How satisfied are you **with your spirituality or religion** ?"

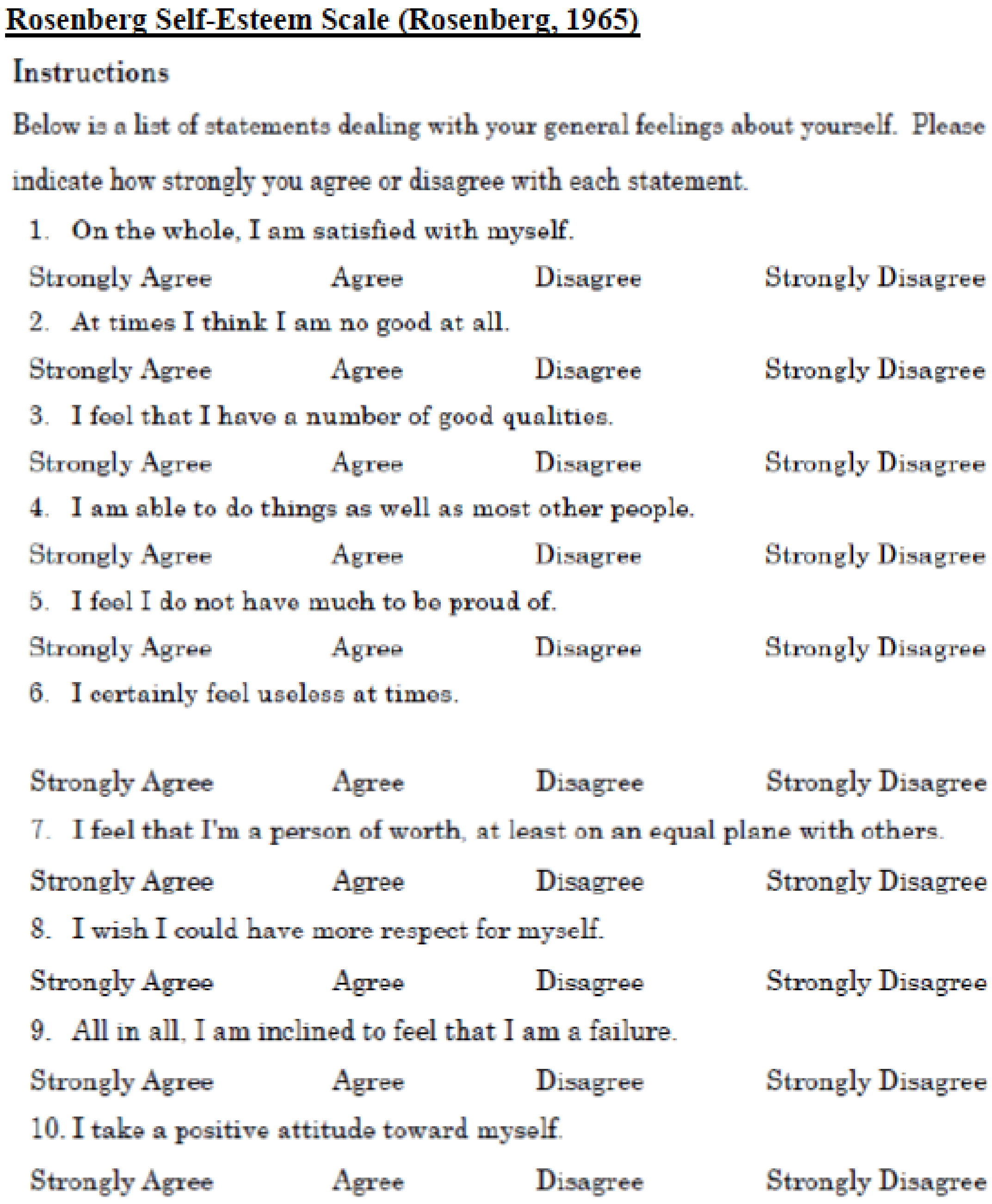
No satisfaction at all										Completely Satisfied
0	1	2	3	4	5	6	7	8	9	10
☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

1. "Thinking about your own life and personal circumstances, how satisfied are you **with your life as a whole ?"**

No satisfaction at all										Completely Satisfied
0	1	2	3	4	5	6	7	8	9	10
☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

Appendix H

Rosenberg Self-Esteem Scale (Rosenberg, 1965)



Rosenberg Self-Esteem Scale (Rosenberg, 1965)

Instructions

Below is a list of statements dealing with your general feelings about yourself. Please indicate how strongly you agree or disagree with each statement.

1. On the whole, I am satisfied with myself.

Strongly Agree Agree Disagree Strongly Disagree

2. At times I think I am no good at all.

Strongly Agree Agree Disagree Strongly Disagree

3. I feel that I have a number of good qualities.

Strongly Agree Agree Disagree Strongly Disagree

4. I am able to do things as well as most other people.

Strongly Agree Agree Disagree Strongly Disagree

5. I feel I do not have much to be proud of.

Strongly Agree Agree Disagree Strongly Disagree

6. I certainly feel useless at times.

Strongly Agree Agree Disagree Strongly Disagree

7. I feel that I'm a person of worth, at least on an equal plane with others.

Strongly Agree Agree Disagree Strongly Disagree

8. I wish I could have more respect for myself.

Strongly Agree Agree Disagree Strongly Disagree

9. All in all, I am inclined to feel that I am a failure.

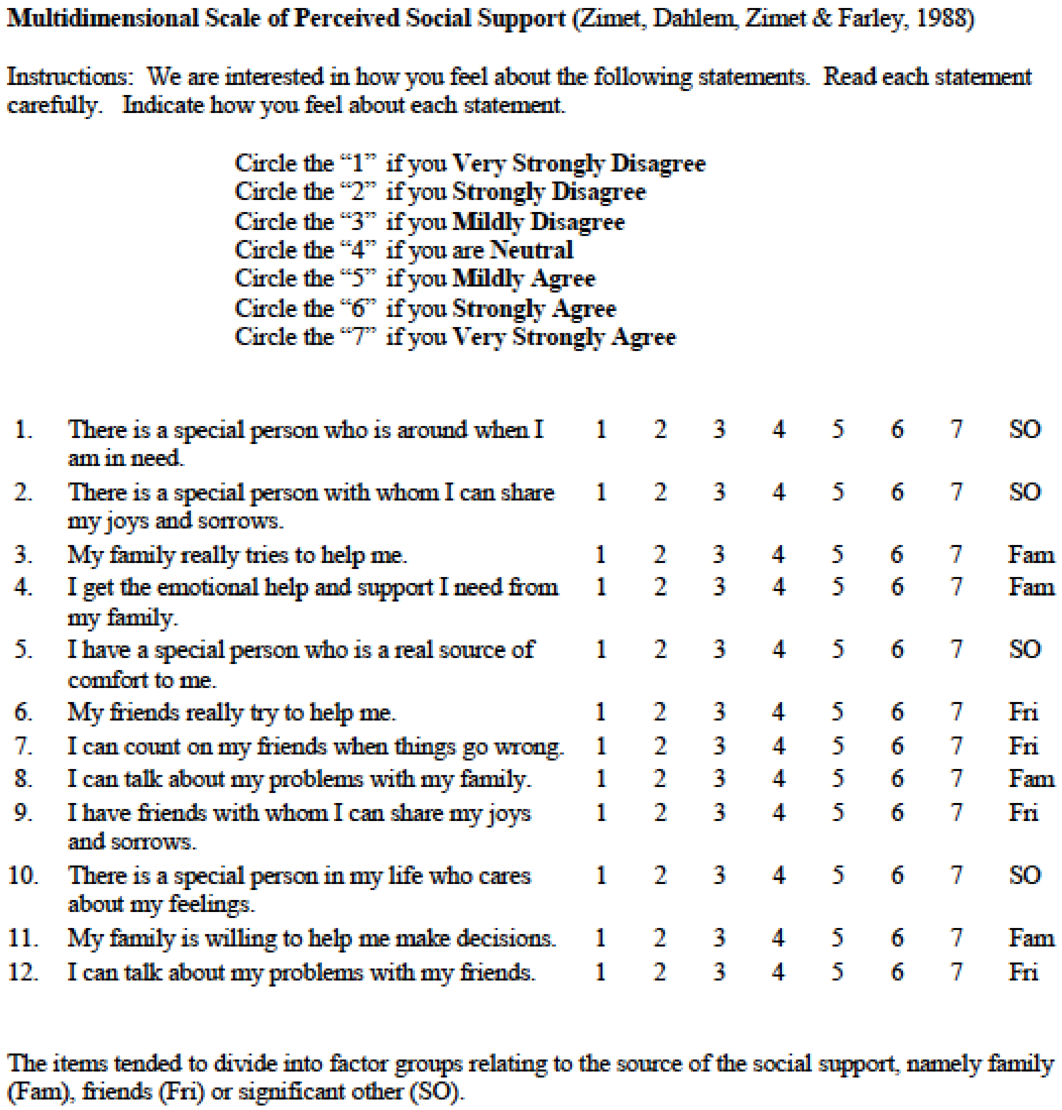
Strongly Agree Agree Disagree Strongly Disagree

10. I take a positive attitude toward myself.

Strongly Agree Agree Disagree Strongly Disagree

Appendix I

The Multidimensional Scale of Perceived Social Support (Zimet, Dahlem, Zimet &Farley, 1988).



Multidimensional Scale of Perceived Social Support (Zimet, Dahlem, Zimet & Farley, 1988)

Instructions: We are interested in how you feel about the following statements. Read each statement carefully. Indicate how you feel about each statement.

Circle the "1" if you **Very Strongly Disagree**
Circle the "2" if you **Strongly Disagree**
Circle the "3" if you **Mildly Disagree**
Circle the "4" if you are **Neutral**
Circle the "5" if you **Mildly Agree**
Circle the "6" if you **Strongly Agree**
Circle the "7" if you **Very Strongly Agree**

1.	There is a special person who is around when I am in need.	1	2	3	4	5	6	7	SO
2.	There is a special person with whom I can share my joys and sorrows.	1	2	3	4	5	6	7	SO
3.	My family really tries to help me.	1	2	3	4	5	6	7	Fam
4.	I get the emotional help and support I need from my family.	1	2	3	4	5	6	7	Fam
5.	I have a special person who is a real source of comfort to me.	1	2	3	4	5	6	7	SO
6.	My friends really try to help me.	1	2	3	4	5	6	7	Fri
7.	I can count on my friends when things go wrong.	1	2	3	4	5	6	7	Fri
8.	I can talk about my problems with my family.	1	2	3	4	5	6	7	Fam
9.	I have friends with whom I can share my joys and sorrows.	1	2	3	4	5	6	7	Fri
10.	There is a special person in my life who cares about my feelings.	1	2	3	4	5	6	7	SO
11.	My family is willing to help me make decisions.	1	2	3	4	5	6	7	Fam
12.	I can talk about my problems with my friends.	1	2	3	4	5	6	7	Fri

The items tended to divide into factor groups relating to the source of the social support, namely family (Fam), friends (Fri) or significant other (SO).

Appendix J

Number and Percentages of Participants who selected Each Response Option

Item	Strongly Disagree	Somewhat Disagree	Neither Disagree nor Agree	Somewhat Agree	Strongly Agree
Factor 1: Perceived family stigma					
Some people might feel embarrassed about associating with the family of someone with LD.	39 (9.6)	56 (13.8)	50 (12.3)	204 (50.1)	58 (14.3)
Some people might feel uncomfortable about going to the house of the family of someone with LD.	34 (8.4)	42 (10.3)	66 (16.2)	210 (51.6)	55 (13.5)
Some people might treat the family of someone with LD more negatively.	33 (8.1)	42 (10.3)	64 (15.7)	190 (46.7)	77 (18.9)
Some people might think that the family has done something wrong because of the person with LD.	87 (21.4)	85 (20.9)	99 (24.3)	101 (24.8)	33 (8.1)
Some people might behave negatively towards the family of someone with LD when they are with the person with LD in public.	52 (12.8)	62 (15.2)	79 (19.4)	162 (39.8)	51 (12.5)
Some people might avoid making friends with the family of someone with LD.	37 (9.10)	35 (8.6)	60 (14.7)	184 (45.2)	89 (21.9)
Some people might not want to hear about any of the problems of the family of someone with LD.	32 (7.9)	56 (13.8)	70 (17.2)	164 (40.3)	85 (20.9)
Some people might not invite the family of someone with LD to social events.	42 (10.3)	49 (12.0)	47 (11.5)	158 (38.8)	111 (27.3)

Table 3 continued

Item	Strongly Disagree	Somewhat Disagree	Neither Disagree nor Agree	Somewhat Agree	Strongly Agree
Factor 2: Affective affiliate stigma					
I feel embarrassed about my family member with LD.	281 (69.0)	45 (11.1)	28 (6.9)	48 (11.8)	5 (1.2)
I feel distressed about being associated with my family member with LD.	329 (80.8)	35 (8.6)	23 (5.7)	12 (2.9)	4 (1.0)
I feel guilty about having a family member with ID in the family.	327 (80.3)	33 (8.1)	16 (3.9)	26 (6.4)	4 (1.0)
I feel uncomfortable when I have friends over because of my family member with LD.	243 (59.7)	48 (11.8)	27 (6.6)	76 (18.7)	11 (2.7)
I feel worried that I/my family will be blamed for the family member's LD.	261 (64.1)	37 (9.1)	34 (8.4)	52 (12.8)	22 (5.4)
Factor 3: Cognitive affiliate stigma					
I am looked down upon when other people discover that I am related to my family member with LD.	163 (40.0)	74 (18.2)	101 (24.8)	52 (12.8)	15 (3.7)
I am treated differently by some people when I am with my family member with LD.	65 (16.0)	44 (10.8)	50 (12.3)	194 (47.7)	52 (12.8)
I am excluded from activities when other people find out about my family member's LD.	78 (19.2)	53 (13.0)	62 (15.2)	162 (39.8)	51 (12.5)
I am aware of how some people look at me when I am out with my family member with LD.	30 (7.4)	20 (4.9)	32 (7.9)	184 (45.2)	138 (33.9)
I am treated differently by some people because of my family member with LD.	32 (7.9)	29 (7.1)	54 (13.3)	200 (49.1)	91 (22.4)

Table 3 continued

Item	Strongly Disagree	Somewhat Disagree	Neither Disagree nor Agree	Somewhat Agree	Strongly Agree
Factor 4: Behavioural affiliate stigma					
I avoid introducing my friends to my family member with LD.	274 (67.3)	53 (13.0)	49 (12.0)	18 (4.4)	11 (2.7)
I avoid telling people that I am related to my family member with LD.	341 (83.8)	36 (8.8)	21 (5.2)	3 (0.7)	2 (0.5)
I avoid making new friends because of my family member with LD.	254 (62.4)	54 (13.3)	46 (11.3)	39 (9.6)	9 (2.2)
I avoid being seen with my family member with LD.	354 (87.0)	27 (6.6)	11 (2.7)	9 (2.2)	2 (0.5)
Factor 5: Positive aspects of caregiver					
Caring for my family member with LD has enabled me to develop a more positive attitude toward life.	38 (9.3)	63 (15.5)	62 (15.2)	120 (29.5)	124 (30.5)
Caring for my family member with LD has made me feel needed.	27 (6.6)	25 (6.1)	116 (28.5)	118 (29.0)	120 (29.5)
Caring for my family member with LD has strengthened my spirituality and faith.	119 (29.2)	50 (12.3)	133 (32.7)	56 (13.8)	49 (12.0)
Caring for my family member with LD has allowed me to form friendships with others in a similar situation.	29 (7.1)	43 (10.6)	54 (13.3)	145 (35.6)	136 (33.4)
Caring for my family member with LD has made me feel that I make a positive contribution to society.	46 (11.3)	65 (16.0)	120 (29.5)	97 (23.8)	75 (18.4)
Caring for my family member with LD has strengthened some of my relationships with family/friends.	68 (16.7)	99 (24.3)	100 (24.6)	92 (22.6)	47 (11.5)