Review article

Stigma experienced by families of individuals with intellectual disabilities and autism: A systematic review

Natasha Mitter[a],[*], Afia Ali[a], Katrina Scior[b]

[a] Research Department of Clinical, Educational and Health Psychology, University College London, Gower Street, London WC1E 6BT, United Kingdom
[b] Division of Psychiatry, University College London, Gower Street, London WC1E 6BT, United Kingdom

ABSTRACT

Studies have investigated the experiences of courtesy stigma and affiliate stigma in family members of individuals with intellectual and developmental disabilities (IDD) 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 and/or autism. A systematic search of PsycINFO, ERIC and Scopus identified relevant articles published between 2012 and 2016, to expand on an earlier review on this topic published in 2012.

Ten articles pertaining to eight studies were identified. They revealed that family carers do experience stigma and various consequences related to these, with family culture influencing these experiences. This review identifies a number of psychosocial variables that are associated with the development of courtesy stigma, affiliate stigma and their consequences. It highlights protective factors and strategies family carers use to cope with stigma, and a lack of clarity in distinguishing the concepts of courtesy stigma and affiliate stigma in family members.

What this paper adds

Studies on stigma in the intellectual and developmental disabilities (IDD) field have mainly focused on the experience of stigma for the individual with IDD but stigma may extend to and affect their family members as well. The stigma of being undervalued by others and challenges in accessing support add to caregiver burden, adversely affecting their quality of life. To mitigate these effects, caregivers may react by withdrawing socially and isolating themselves.

However, research on stigma in family members of people with IDD remains limited. To date, only one systematic review has examined stigma in family members. It is unclear whether carers initially perceive stigma before its potential internalisation, and the variables that may affect this process. The present systematic review aimed to fill these gaps in the evidence base while also paying close attention to positive aspects of caregiving and the role of culture. It integrates different conceptualisations of stigma and arrives at a more holistic understanding of the process of stigmatisation for caregivers. Carers do in fact perceive courtesy stigma, which precedes the internalisation of negative evaluations and subsequent development of affiliate stigma. Consequences of this internalisation include social withdrawal or concealment which may in turn negatively influence access to coping resources. Culture appears to underlie this entire process.

1. Introduction

Individuals with intellectual disabilities (ID) and autism spectrum disorders (ASD) face stigma, prejudice and significant obstacles

that restrict their human rights and access to opportunities others take for granted (Scior & Werner, 2016). 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 (Goffman, 1963). The construct of stigma refers to attitudes, stereotypes, prejudice, and discrimination (Corrigan, Roe, & Tsang, 2011), which result from misconceptions about an individual's attributes by the dominant cultural group, and are perpetuated through biased social structures (Corrigan, 2000).

Goffman (1963) noted that stigma not only affects individuals carrying a stigmatising attribute, but also extends to those affiliated with them. In this paper, we summarise and critically appraise studies concerned with stigma experienced by families of those with ID and ASD, and discuss the implications of their findings.

1.1. Forms of stigma

Four 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, Pryor, Reeder, & Stutterheim, 2013). The second type, self-stigma, occurs when the stigmatised person becomes aware of *and* internalises public stigma. Typically, studies in the IDD field have focused on self-stigma as experienced by the individual with IDD. Often however, persons associated with the stigmatised person, particularly family members, are also subjected to stigmatisation, which may lead to the family member to be mocked or blamed for the person's disabilities (Ali, Hassiotis, Strydom, & King, 2012). This third type of stigma has been referred to as family stigma (Phelan, Bromet, & Link, 1998), courtesy stigma (Goffman, 1963), or associative stigma (Mehta & Farina, 1988). Consequently, family members may develop negative emotions and negative cognitions about themselves, resulting in social withdrawal or concealment. This process of internalisation of courtesy or associative stigma by the stigmatised individual's affiliates, most likely family members, gives rise to the fourth type of stigma, affiliate stigma (Ali et al., 2012). As previously conceptualised, it comprises three interlinking psychological responses: stigmatised cognitions, affect and behaviour.

Although caregiving can often be a rewarding experience, the family caregivers of individuals with IDD 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, Lin, & Kröger, 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). Mothers of children with IDD 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 IDD. 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 IDD to avoid association (Mak & Cheung, 2008).

To date, there has only been one published systematic review on this topic - Ali et al. (2012) reviewed the literature on affiliate stigma in family members of individuals with ID published between 1990 and February 2012. Of the 20 studies included in their review as pertaining to courtesy and affiliate stigma, ten had used qualitative methodologies, six were cross-sectional, three used mixed methods and one was longitudinal. The findings across 1415 family members and 11 countries across Africa, Asia, Australia, Europe, and North America indicated that family members experienced negative attitudes or responses from the public, and that many were subjected to negative treatment by their own extended families and communities, especially in non-Western cultures. Moreover, stigma negatively affected psychological well-being in parents, leading to reduced quality of life. The authors concluded that adverse effects of stigma may be alleviated by having a supportive network and through the use of acceptance and education.

1.2. Aims and objectives

The present systematic review set out to summarise the findings of research into courtesy and affiliate stigma in family members of people with IDD carried out since Ali et al.'s review. The review aimed to answer the following questions:

1 What is known about the experience of stigma associated with IDD in family members of individuals with IDD?
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?

2. Method

2.1. Search strategy

Study reports published over the period January 2011 to July 2016 were identified by searching the electronic databases PsycINFO, ERIC ProQuest, and Scopus. The terms 'intellectual disability', 'mental retardation', 'learning disability'', 'autism', 'autism spectrum disorder', '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', 'caregiver*', 'family' and 'relative'. The search was filtered by abstract field. Identified studies were first screened and duplicates and irrelevant studies were removed. The remaining studies were read in full to identify whether they met the inclusion criteria. Their references were also searched to identify further relevant articles not picked up in the database searches.

2.2. Inclusion and exclusion criteria

Included articles had to report in English on a qualitative or quantitative study published in a peer reviewed journal. The reported studies' aims had to include to examine the experience of affiliate stigma in family members of people with ID or ASD. In addition, studies had to formally measure affiliate stigma. Family members included mothers, fathers, siblings and other relatives of individuals with ID or ASD. Studies that did not specifically refer to individuals with ID, focused on ASD in the absence of ID, or on other developmental disorders (e.g. epilepsy), were excluded as were articles that made brief reference to the experience of affiliate stigma but did not make this a central focus of the reported study and findings.

2.3. Quality assessment and analysis

A structured questionnaire developed by Hawker, Payne, Kerr, Hardey, and Powell, (2002) was used to assess the quality of articles included in the review (see Appendix A). Using this tool, the articles were rated on a scale of 1 (very poor) to 4 (good) on nine different facets related to methodology. The tool provided well-defined instructions on how to score these different facets and was suitable to address the aims of the studies as it allows systematic evaluation of studies from different paradigms (Hawker et al., 2002). Using this tool, information was extracted about the design, sample size, selection of participants, type of measures or interviews utilized, and reliability and validity (if appropriate), the quality of reporting of the findings, generalisability, and any methodological limitations for each study. 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. The tool has shown to have good inter-rater reliability (Hawker et al., 2002).

3. Results

A total of ten articles met the inclusion criteria and were included in this review (Fig. 1). They were overall of a fair to good quality (M = 28.8, SD = 3.49). Table 1 provides an overview of these studies. The ten articles reported on eight different studies: two articles by Chiu et al. (2013, 2015) reported on the same Chinese study, and two articles by Werner and Shulman (2013, 2015) reported on the same Israeli study. Eight were quantitative, while two adopted a mixed methods design. Studies were conducted mainly in Asia and Israel, with only one article from Europe and one from the African subcontinent.

3.1. Study design

In half of the studies, scales measuring courtesy/affiliate stigma 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. 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.
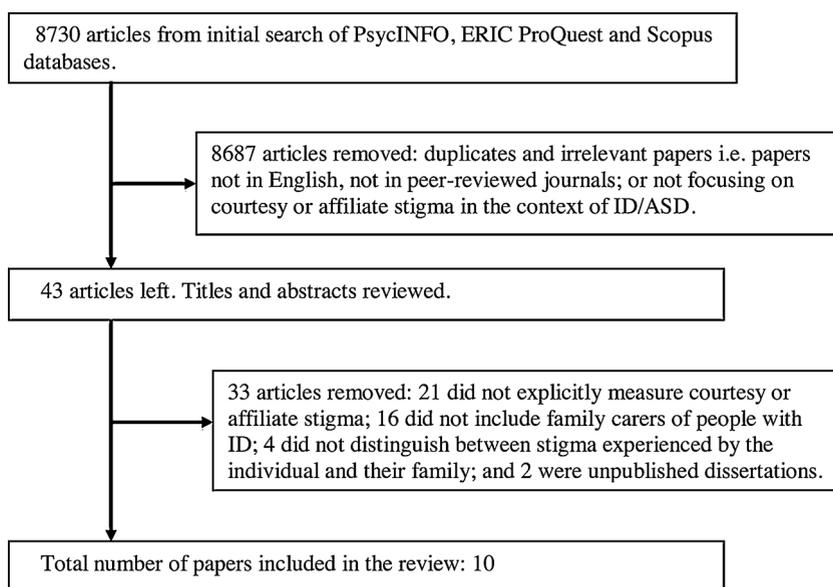
Fig. 1. Flow diagram of search process.

Table 1
Overview of the studies examining courtesy or affiliate stigma in family members.

Study & Location	Design	Sample	Method	Key Findings	Limitations
Cantwell et al. (2015); Rep. 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. (1998). 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. (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.
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, 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.
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 measured caregiving burden and marital satisfaction.	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.
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.
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.
Werner and 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.
Werner and Shulman (2015); Israel	Cross-sectional	170 parents (129 mothers, 13 fathers and 11 others) of individuals with ID, ASD and physical disabilities (PD).	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 stigma according to diagnosis. Affiliate stigma not related to caregiver variables, but to child-related variables in ASD.	Coping mechanisms not examined. Small convenience sample. More than one primary diagnosis for most and based on caregiver reports.
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.

(continued on next page)

Table 1 (*continued*)

Study & Location	Design	Sample	Method	Key Findings	Limitations
Yang (2015); China	Mixed methods	120 family members (102 parents, 18 others) of children with ID. Of these, 10 (5 mothers, 1 grandmother, 1 sister and 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.

3.2. Sample characteristics and recruitment

Participants were recruited from schools for pupils with special needs (n = 5), non-governmental organisations (NGOs) focused on IDD (n = 3), official registries (n = 2), and outpatient child mental health clinics (n = 1). Some recruited from multiple populations. Out of the eight studies, only five included siblings and other relatives in their samples (Chiu, Yang, Wong, & Li, 2015; Chiu, Yang, Wong, & Li, 2013; Werner & Shulman, 2013, 2015; Yang, 2015); the remainder focused on parents, primarily mothers. Eight of the articles reported on experiences of middle-aged family members (between the ages of 30 and 50) caring for children (aged two to 12.7 years). The experiences of younger carers, i.e. those below the ages of 30, or those of adults with IDD may well differ.

3.3. Courtesy and affiliate stigma

The articles reviewed provide evidence on both the experience and internalisation of stigma. While all reported caregivers experiencing stigma based on various cognitive, emotional and behavioural responses, most studies did not differentiate between courtesy and affiliate stigma. Five articles explicitly stated that they examined affiliate stigma (Chiu et al., 2013,2015; Werner & Shulman, 2013, 2015; Wong, Mak, & Liao, 2016). The other five used terms such as 'familial/family stigma' (Ngo, Shin, Nhan, & Yang, 2012; Tilahun et al., 2016), 'perceived stigma' (Kwok, Leung, & Wong, 2014; Cantwell, Muldoon, & Gallagher, 2015), 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, the term 'courtesy stigma' is used to describe findings related to the perception of stigmatisation due to the association with individuals with IDD, while 'affiliate stigma' is used when referring to 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.

Relatively lower levels of stigma were reported in Israel (Werner & Shulman, 2015) compared to Hong Kong, Taiwan and China; 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 may vary in different cultural settings.

3.4. The experience of courtesy stigma

Based on the studies reviewed, family caregivers experienced courtesy stigma both within their families and communities. Most of the studies appeared to describe courtesy stigma in relation to caregivers' experiences of marginalisation by their communities and families. 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. (2012) study reported that their child with ID would never be able to get married 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 IDD led to concerns about 'saving face' in the community (2015, Chiu et al., 2013; Yang, 2015), the Chinese concept of 'face' referring to one's sense of dignity in a social context. People from Asian cultures may have a tendency to highlight the genetic origins of IDD (Kung, 2001) and may be more inclined to consider children with disabilities as "bad seeds" who bring shame to their families (Sue & Zane, 1987). Similarly, Tilahun et al. (2016) found that Ethiopian 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.

Only one study (Cantwell et al., 2015) directly examined the association between challenging behaviour in the individual with IDD and courtesy stigma, while two others referred to it in the context of disability type (Kwok et al., 2014; Werner & Shulman, 2013). Cantwell et al. (2015) found no association between child problem behaviours and courtesy stigma. This lack of association could be explained by the choice of measures of challenging behaviours used, which may not have identified the full range of behavioural problems. Kwok et al. (2014) found that mothers of children with ASD reported higher levels of courtesy stigma than mothers of children with ID. Similarly, Werner and Shulman (2013) reported stigma to be higher in caregivers of individuals with ASD as compared to ID or physical disabilities. In comparing individuals with ID alone to those with ID and ASD, McCarthy et al. (2010) 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 (Kwok et al., 2014). As such, the carer may face discrimination by others and come to internalise feelings of shame.

Kinnear, Link, Ballan, and Fischbach, (2015) developed 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 may carry assumptions about poor parenting and about the capabilities of the individual with challenging behaviours, which in turn increase the likelihood of social exclusion. The studies reviewed support this conceptual model, adding a dimension of culture that underlies this experience.

3.5. 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.

3.5.1. Sociodemographic variables and affiliate stigma

Seven articles examined the role of sociodemographic variables in the process of internalisation (2015, Cantwell et al., 2015; Chiu et al., 2013; Ngo et al., 2012; Tilahun et al., 2016; Werner & Shulman, 2015; Yang, 2015), with two of these reporting no association (Cantwell et al., 2015; Werner & Shulman, 2015). Four of these examined affiliate stigma in relation to socioeconomic status (2015, Chiu et al., 2013; Ngo et al., 2012; Yang, 2015). Ngo et al. (2012) concluded that carers of higher socioeconomic status perceived and internalised more stigma than those of lower socioeconomic status, consequently placing restrictions on their social lives. Those with higher education perceived less restriction on their social life when they experienced affiliate stigma. The authors suggested that more educated carers may not endorse traditional Vietnamese beliefs of karma and past sins which implicate carers in the aetiology of the child's disability, thus reducing effects of stigma on their social lives. Chiu et al. (2013) described contrary findings, with family carers of higher socioeconomic status generally reporting lower levels of face concern and affiliate stigma than carers of low and middle socioeconomic status. The authors explained this with reference to these carers having more resources to cope with the effects of stigma, and thus reporting reduced psychological distress, less anxiety and increased personal empowerment.

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, showed higher levels of affiliate stigma on the behavioural component of the Affiliate Stigma Scale and lower empowerment. They concluded that due to increasing emotional and behavioural problems of individuals with IDD as they age, older caregivers have been subjected to affiliate stigma for a longer duration and are therefore more likely to internalise stigma (Chiu et al., 2013).

3.5.2. 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 reported self-esteem, social support, positive meaning in caregiving and affiliate stigma to be 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 therefore appears to be 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 symptomatology in parents caring for children with IDD, Cantwell et al. (2015) found an association between caregiver stigma and social support, self-esteem, depressive symptomatology. 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 - 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.

3.6. The consequences of affiliate stigma

The articles 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 their 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.

3.6.1. Caregiver mental health

Five studies involving a total of 806 participants investigated the relationship between caregiver mental health and affiliate stigma, and found carers who experienced higher levels of stigma to be more likely to experience symptoms of anxiety and depression (2015, Cantwell et al., 2015; Chiu et al., 2013; Wong et al., 2016; Yang, 2015). Two other studies considered negative emotional reactions experienced by caregivers as a result of stigma (Ngo et al., 2012; Tilahun et al., 2016), including sadness, embarrassment, shame, guilt and fear. Chiu et al. (2013) reported 61% of their sample of family caregivers experienced mental health problems 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 problems were associated with the affective component of affiliate stigma and in one study were 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 may unconsciously affect the carer by negatively affecting internal coping resources and making them feel helpless (Chiu et al., 2015). 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 interactions between the affective component and cultural factors that lead to mental health problems in caregivers, particularly in collectivist cultures. 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 their 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.

Cantwell et al. (2015) reported affiliate stigma as one of the psychosocial pathways influencing high depressive symptomatology in

carers, with self-esteem mediating this association. The path through self-esteem varied as a function of emotional support: for parents with higher self-esteem, the mediation of stigma and depressive symptoms was not dependent on emotional support. In the same vein, Wong et al. (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.

3.6.2. 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.

3.7. Managing the impact of affiliate stigma

Four studies evaluated coping strategies and reported the use of both positive and negative strategies by family members to cope with the impact of affiliate stigma. 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). Yang (2015) found that carers used compensation/overprotection, where the anticipation of stigma and associated feelings of self-blame led to overprotection of the individual with ID from potential harm.

Tilahun et al. (2016) reported more than half of their sample of carers found support in their religious beliefs and rituals by using prayer as a means of coping with the negative effects of affiliate stigma. Yang (2015) reported that family carers may manage the effects of affiliate stigma 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. While talking to health professionals, family and friends was the most common strategy employed among carers in Tilahun et al.'s (2016) study, increased stigma was associated with seeking help from traditional sources, including religious centres and traditional healers. The sample of caregivers in the study sought help from biomedical sources as a 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), was examined as a unique coping strategy in two of the more recent Chinese studies (Wong et al., 2016; Yang, 2015). 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. Self-compassion can therefore serve as an internal coping resource without having the family carer rely on external sources of help. 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 help to develop a repertoire of internal coping resources in order to address the different dimensions (i.e. cognitive, affective and behavioural) of affiliate stigma.

4. Discussion

4.1. 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 IDD, 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 IDD. The conclusions drawn concur with the findings of Ali et al. (2012) to some extent. As in Ali et al. (2012) review, most family caregivers experienced courtesy and affiliate stigma, but the extent and form these took varied across cultures. Specifically, this review confirmed that family members are marginalised by not only the community and their own families, but also by professionals. This review expanded on the psychological factors family members experienced in courtesy and affiliate stigma, with a focus on the positive and negative coping strategies employed by them. It went beyond Ali et al.'s conclusions by highlighting that experiences of courtesy stigma depend on carers' settings and the behaviours the individual with IDD exhibits in others' presence. Repeated exposure to others' stigmatising attitudes and behaviours increased the likelihood of internalisation of stigma. The extent of internalisation in turn appeared contingent on factors associated with carers' backgrounds and internal coping resources. Consequently, as reported by Ali et al. (2012), affiliate stigma has an effect on carers' experienced burden of care and their mental health, and can lead to the use of positive as well as negative coping strategies.

The key difference to Ali et al. (2012) review is that findings from the current review indicated a shift in research from a rather negative view of parenting of individuals with IDD 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. 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 Fig. 2 (Appendix A2).
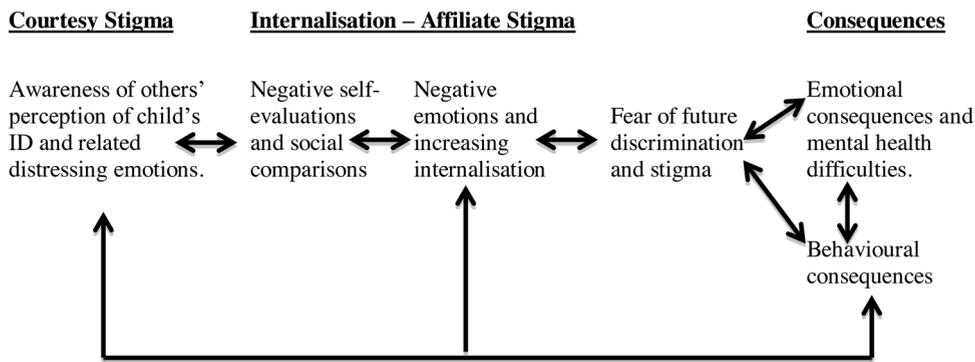
Fig. 2. Synthesis of findings – The circle of stigma.

4.2. The circle of stigma

Courtesy stigma and related experiences precede the internalisation of negative evaluations and eventually lead to the development of affiliate stigma (Fig. 2). 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 IDD who exhibits 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. Repeated occurrences 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. Emotions have been theorised to be a response to cognitive activity, specifically that they involve 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 such as shame, guilt and embarrassment and therefore unconscious.

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, family carers may resort to such maladaptive coping strategies as concealment and social exclusion, which then reinforce affiliate stigma. 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.

The findings of this review lend some support to the notion that different aspects or phases of courtesy stigma, affiliate stigma and their 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.

4.3. The role of culture

Findings from this review highlight the importance of the cultural context for courtesy and affiliate stigma experienced by caregivers. The Chinese concept of face concern is useful in illustrating how culture plays a role in the stigma circle presented in Fig. 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 IDD is in itself stigmatising (Chiu et al., 2013). In Western cultures, where individualistic beliefs dominate, other variables, such as child behaviours that run counter to social norms or the type of IDD may have a larger influence on the process. Culture also plays a role in the use of coping strategies for carers. Cinnirella and Loewenthal (1999) found that prayers offered ethnic minority 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 and Loewenthal, 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.

4.4. Implications

The findings have a number of implications for the provision of interventions and support for family members caring for an

individual with ID and ASD, in consideration of their cultural context. In particular, family members should be supported in developing internal coping resources such as self-compassion and establishing or emphasising/valuing other social identities to bolster their self-esteem (Wong et al., 2016; Yang, 2015). This is especially important in highly stigmatising communities where families may not be able to depend on others for support. 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 (Perkins, Holburn, Deaux, Flory, & Vietze, 2002; Thoits, 1983). Establishing multiple identities and switching between these seems to protect carers' self-esteem and psychological wellbeing from stigma. Additionally, regardless of their self-perceptions, experiences that impinge carers' cultural beliefs promptly evoke emotional responses (Yang, 2015). Culturally sensitive interventions that enhance coping skills to help with the management of self-stigmatised feelings may thus be more effective than attempting to change stigmatising cognitions (Mittal, Sullivan, & Chekuri, 2012).

Affiliate stigma is less likely to occur if courtesy stigma is minimised (Papadopoulos, 2016). Collaborations with traditional sources of support may be necessary to improve public awareness and could foster more positive community attitudes which may in turn reduce affiliate stigma. On a more practical side, given the comorbidity of challenging behaviours among individuals with ID and ASD in particular, parents should have access to behavioural management strategies. Given that affiliate stigma can affect one without conscious awareness (Chiu et al., 2015), the identification of parents susceptible through recognition of risk factors for affiliate stigma is critical. Offering culturally sensitive counselling and psychotherapy services to families deemed at risk of affiliate stigma and establishing peer support groups for them to provide emotional support through the exchange of shared experiences and reflections could serve as important early intervention strategies.

4.5. Limitations

Limitations of this review have to be acknowledged. Relatively narrow search parameters were applied. Using other potential sources of information such as dissertations or unpublished and non-English publications would possibly uncover other relevant literature. It can also be argued that the sample size is too small for the kinds of generalisations made in this paper. This study included ID and ASD, which can complicate findings, especially since differences have been reported between these groups. In addition, the experiences of family members caring for individuals with other types of developmental disabilities were not included and might differ.

4.6. Potential areas for future research

The review identified shortcomings in the existing literature. Firstly, 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 the field, more accurate measures need to be developed for this, especially if we want to test the outcomes of interventions for family caregivers.

Second, a majority of the studies focused on the internalised emotional basis of stigma experienced by caregivers, as well as factors that may 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. 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, particularly siblings, 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.

5. Conclusions

This review showed that family carers experienced stigma and various consequences related to these, with family culture influencing these experiences and various psychosocial variables associated with its development. Phases of courtesy stigma, affiliate stigma and its consequences are interlinked, which may create a vicious circle of increasing levels of courtesy stigma and affiliate stigma. Family carers used number of coping strategies and protective factors to buffer against the effects of these. Further research would help gain a better understanding of stigma in family members of individuals with ID and ASD.

Conflict of interest statement

None declared.

Appendix A

Table A1

Table A1
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)	Sampl-ing (Q4)	Data Analysis (Q5)	Ethics & bias (Q6)	Findings & results (Q7)	Transferability/ generalis-ability(Q8)	Implications & usefulness (Q9)	
Ngo et al. (2012)	3	3	3	2	2	1	3	2	4	23
Chiu et al. (2013)	4	3	4	3	4	2	4	3	3	30
Werner and 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 and 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

References

Ali, A., Hassiotis, A., Strydom, A., & King, A. (2012). Self stigma in people with intellectual disabilities and courtesy stigma in family carers: A systematic review. *Research in Developmental Disabilities, 33*, 2122–2140.

Andersson, A. (1993). Depression and anxiety in families with a mentally handicapped child. *International Journal of Rehabilitation Research, 16*, 165–169.

Baxter, C., Cummins, R. A., & Yiolitis, L. (2000). Parental stress attributed to family members with and without disability: A longitudinal study. *Journal of Intellectual & Developmental Disability, 3*, 105–118.

Blacher, J., & Mink, I. T. (2004). Interviewing family members and care providers: Concepts, methodologies and cultures. In E. Emerson, C. Hatton, T. Thompson, & T. R. Parmenter (Eds.). *In: The International handbook of applied research in intellectual disabilities* (pp. 133–160). Chichester: Wiley.

Bos, A. E. R., Pryor, J. B., Reeder, G. D., & Stutterheim, S. E. (2013). Stigma: Advances in theory and research. *Basic and Applied Social Psychology, 35*, 1–9.

Cantwell, J., Muldoon, O., & Gallagher, S. (2015). The influence of self-esteem and social support on the relationship between stigma and depressive symptomology in parents caring for children with intellectual disabilities. *Journal of Intellectual Disability Research, 59*, 948–957.

Chiu, M. Y. L., Yang, X., Wong, F. H. T., & Li, J. H. (2013). Caregiving of children with intellectual disabilities in china - an examination of affiliate stigma and the cultural thesis. *Journal of Intellectual Disability Research, 57*, 1117–1129.

Chiu, M. Y. L., Yang, X., Wong, H. T., & Li, J. H. (2015). The mediating effect of affective stigma between face concern and general mental health – The case of Chinese caregivers of children with intellectual disability. *Research in Developmental Disabilities, 36*, 437–446.

Chou, Y. C., & Palley, H. A. (1998). The impact of having a child with developmental disabilities on the family in Taiwan: Cultural context review. *Social Development Issues, 20*, 35–52.

Chou, Y. C., Pu, C. Y., Lee, Y. C., Lin, L. C., & Kröger, T. (2009). Effect of perceived stigmatisation on the quality of life among ageing female family carers: A comparison of carers of adults with intellectual disability and carers of adults with mental illness. *Journal of Intellectual Disability Research, 53*, 654–664.

Cinnirella, M., & Loewenthal, K. M. (1999). Religious and ethnic group influences on beliefs about mental illness: A qualitative interview study. *The British Journal of Medical Psychology, 72*, 505–524.

Corrigan, P. W. (2000). Mental health stigma as social attribution: Implications for research methods and attitude change. *Clinical Psychology Science and Practice, 7*, 48–67.

Corrigan, P., Roe, D., & Tsang, H. (2011). *Challenging the stigma of mental illness.* Chichester, West Sussex, UK: Wiley-Blackwell.

Goffman, E. (1963). *Stigma: Notes on the management of spoiled identity.* New York: Simon & Schuster.

Hawker, S., Payne, S., Kerr, C., Hardey, M., & Powell, J. (2002). Appraising the evidence: Reviewing disparate data systematically. *Qualitative Health Research, 12*, 1284–1299.

Kinnear, S. H., Link, B. G., Ballan, M. S., & Fischbach, R. L. (2015). Understanding the experience of stigma for parents of children with autism spectrum disorder and the role stigma plays in families' lives. *Journal of Autism and Developmental Disorders, 46*, 942–953.

Kung, W. W. (2001). Consideration of cultural factors in working with Chinese American families with a mentally ill patient. *Families in Society, 82*, 97–107.

Kwok, S. Y. C. L., Leung, C. L. K., & Wong, D. F. K. (2014). Marital satisfaction of Chinese mothers of children with autism and intellectual disabilities in Hong Kong. *Journal of Intellectual Disability Research, 58*, 1156–1171.

Lazarus, R. S. (1991). Cognition and motivation in emotion. *The American Psychologist, 46*, 352–367.

Mak, W. W. S., & Cheung, R. Y. M. (2008). Affiliate stigma among caregivers of people with intellectual disability or mental illness. *Journal of Applied Research in Intellectual Disabilities, 21*, 532–545.

McCarthy, J., Hemmings, C., Kravariti, E., Dworzynski, K., Holt, G., Bouras, N., et al. (2010). Challenging behavior and co-morbid psychopathology in adults with intellectual disability and autism spectrum disorders. *Research in Developmental Disabilities, 31*, 362–366.

Mehta, S., & Farina, A. (1988). Associative stigma: Perceptions of the difficulties of college-aged children of stigmatized fathers. *Journal of Social and Clinical Psychology, 7*, 192–202.

Mittal, D., Sullivan, G., Chekuri, L., et al. (2012). Empirical studies of self-stigma reduction strategies: A critical review of the literature. *Psychiatric Services, 63*, 974–981.

Ngo, H., Shin, J., Nhan, N., & Yang, L. (2012). Stigma and restriction on the social life of families of children with intellectual disabilities in Vietnam. *Singapore Medical Journal, 53*, 451–457.

Papadopoulos, C. (2016). Self-stigma among carers of autistic people. *Network Autism.* Retrieved December 23, 2016, from http://network.autism.org.uk/good-practice/evidence-base/self-stigma-among-carers-autistic-people.

Papadopoulos, C., Foster, J., & Caldwell, K. (2013). 'Individualism-Collectivism' as an explanatory device for mental illness stigma. *Community Mental Health Journal, 49*, 270–280.

Perkins, T. S., Holburn, S., Deaux, K., Flory, M. J., & Vietze, P. M. (2002). Children of mothers with intellectual disability: Stigma, mother-child relationship and self-esteem. *Journal of Applied Research in Intellectual Disabilities, 15*, 297–313.

Phelan, J. C., Bromet, E. J., & Link, B. G. (1998). Psychiatric illness and family stigma. *Schizophrenia Bulletin, 24*, 115–126.

Sanders, K. Y. (2006). Overprotection and lowered expectations of persons with disabilities: The unforeseen consequences. *Work, 27*, 181–188.

Scior, K., & Werner, S. (Eds.). (2016). *Intellectual disability and stigma: Stepping out from the margins.* Basingstoke: Palgrave Macmillan.

Sue, S., & Zane, N. (1987). The role of culture and cultural techniques in psychotherapy: A critique and reformulation. *The American Psychologist, 42*, 37–45.

Sylwester, R. (2001). Unconscious emotions, conscious feelings, and curricular challenges. January *Educational Leadership, 58*(3), 20–24.

Thoits, P. A. (1983). Multiple identities and psychological well-being: A reformulation and test of the social isolation hypothesis. *American Sociological Review, 48*(2), 174–187.

Tilahun, D., Hanlon, C., Fekadu, A., Tekola, B., Baheretibeb, Y., & Hoekstra, R. A. (2016). Stigma, explanatory models and unmet needs of caregivers of children with developmental disorders in a low-income African country: A cross-sectional facility-based survey. *BMC Health Services Research, 16*, 1383–1389.

Wahl, O. F., & Harman, C. R. (1989). Family views of stigma. *Schizophrenia Bulletin, 15*, 131–139.

Werner, S., & Shulman, C. (2013). Subjective well-being among family caregivers of individuals with developmental disabilities: The role of affiliate stigma and psychosocial moderating variables. *Research in Developmental Disabilities, 34*, 4103–4114.

Werner, S., & Shulman, C. (2015). Does type of disability make a difference in affiliate stigma among family caregivers of individuals with autism, intellectual disability or physical disability? *Journal of Intellectual Disability Research, 59*, 272–283.

Wong, C. C. Y., Mak, W. W. S., & Liao, K. Y.-H. (2016). Self-compassion: A potential buffer against affiliate stigma experienced by parents of children with autism spectrum disorders. *Mindfulness, 7*, 1385–1395.

Yang, X. (2015). No matter how I think, it already hurts. *Journal of Intellectual Disabilities, 19*, 367–380.