Relationships between Stigma, Social Support, and Distress in Caregivers of Chinese Children with Imperforate Anus: A Multicenter Cross-Sectional Study

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ABSTRACT

Purpose: This study was conducted to explore the relationships between stigma, social support, and distress in caregivers of children with imperforate anus (IA) and determine whether social support mediates the relationship between stigma and distress.

Design and methods: This cross-sectional study was conducted in three tertiary children's hospitals in Eastern China. Primary caregivers completed the Social Support Scale and the Chinese versions of the Parent Stigma Scale and Kessler Psychological Distress Scale, and provided their demographic information. The children's demographic and clinical data were also collected. The hypothesized relations were explored using structural equation modeling via the bootstrap method.

Results: A total of 229 caregivers were enrolled. Distress was positively associated with stigma ($r = 0.396$, $P < 0.01$) and negatively associated with social support ($r = -0.413$, $P < 0.01$) in all dimensions ($r = 0.314$–0.346, $P < 0.01$). Stigma was also negatively correlated with social support ($r = 0.280$, $P < 0.01$). Furthermore, social support could partially mediate the relationship between stigma and distress ($b = 0.135$; 95% confidence interval: 0.072, 0.233).

Conclusions: Stigma can increase caregivers' distress, while social support can reduce it. Stigma can also negatively influence caregivers' social support. Therefore, stigma should be mitigated to enhance caregivers' social support and decrease their distress.

Practice implications: The study findings may aid in the identification of the psychological status of caregivers of children with IA, and also inform targeted intervention programs.

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Imperforate anus (IA) is the major phenotype of anorectal malformation and a relatively common anomaly in pediatrics (Kim et al., 2018). IA has an incidence of 1:2000 to 1:5000 in newborns worldwide (Van der Steeg et al., 2016), and the diagnosis is usually made postnatally, which is an unexpected and distressing circumstance for families (Zhu et al., 2017). On the basis of anatomical location, IA is classified as low, intermediate, or high (Cairo, Gasior, Rollins, & Rothstein, 2018). To correct the anomaly, the neonate is required to undergo at least one anoplasty. The treatment of intermediate and high IA is normally a three-stage process, including a colostomy, anoplasty, and colostomy closure (Tannuri, Ferreira, Mathias, & Tannuri, 2016). The anoplasty is followed by a long period of anal dilation (Jenetzky et al., 2015), necessitating frequent visits during and after treatment.

The treatment of IA is difficult not only for patients but also for caregivers. Despite surgical innovations, the treatment process does not necessarily conclude with the operation or completion of dilation as abnormalities such as incontinence and constipation may occur (Springford, Connor, Jones, Kapetanakis, & Giuliani, 2016). These can have huge impacts on the physical and mental health of both patients and caregivers (Grano, Bucci, Aminoff, Lucidi, & Violani, 2014; Marieke et al., 2013).

IA is rarely fatal in the absence of a life-threatening abnormality such as severe intestinal perforation (Beudeker, Broadis, Borgstein, & Heij, 2013). However, the fact that it is a chronic disease raises concerns about quality of life of the affected children (QoL). It has been reported that even though their physical function improves with age, their psychological condition worsens (Grano, Bucci, Aminoff, Lucidi, & Violani, 2015). In a previous study, up to 73% of patients with IA lived with reduced QoL, with particularly low scores on emotional items (Schmidt

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et al., 2010). Moreover, the QoL of children with IA as reported by their parents can be lower than their self-reported QoL, indicating higher anxiety on the part of parents (Grano et al., 2015). Additionally, one study suggested that caregivers experience more stress than the affected children; this is especially true of primary caregivers, who are at an increased risk for psychological distress (Nisell, Ojmyr-Joelsson, Frenckner, Rydelius, & Christensson, 2009).

Given the particular and sensitive position of the malformation, along with the invasive nature of treatment, IA is usually a cause of embarrassment for school-aged patients, and they are reluctant to speak about their condition (Marieke et al., 2013). Stigma is a prejudicial attitude endorsed by a majority about a special group of people, including the process of stereotyping, labeling, isolation, and discrimination (Nyblade et al., 2019). School-aged patients with IA generally experience stigma because of their impaired self-esteem, while caregivers experience it because of their association with the affected children (Hamlington et al., 2015). Especially in Chinese culture, such congenital malformation is often deemed a curse that falls upon people who have done something bad. People with stigma may experience a high degree of negative feelings (Zhou, Wang, & Yi, 2018), leading to increased distress. Stigma may also negatively influence social support as the fear of disclosure may cause caregivers to isolate themselves. This would indirectly impact their psychological well-being and increase distress via dampened social support (Li, Mo, Wu, & Lau, 2017).

However, examinations of stigma among caregivers of children with IA, especially quantitative studies, are limited. Most studies concerning stigma concentrate on caregivers of children with intellectual disability (Chiu, Yang, Wong, & Li, 2015; Nyblade et al., 2019). Only few qualitative studies use words such as "shame" or "fear of diagnosis disclosure" to summarize the feelings of caregivers of children with IA. (Xiaoran, Lijun, Li, & Benjuan, 2016). In addition, correlational studies focusing on caregivers' psychological status and exploring the relationships between stigma, social support, and distress in this population are lacking. Therefore, we investigated stigma, social support, and distress among primary caregivers of children with IA and the associations among these variables. First, we hypothesized that Chinese primary caregivers of children with IA experience a certain degree of distress. Second, we hypothesized that higher stigma is associated with increased distress. Finally, we hypothesized that the relationship between stigma and distress is mediated by social support.

Methods

Design, setting, and participants

A cross-sectional study was conducted in three tertiary children's hospitals in Eastern China between November 2018 and February 2019. A total of 254 caregivers were screened. To be eligible for participation, children had to (1) be diagnosed with IA, which means the anus area is completely covered by skin after birth; there is no normal opening, perhaps only a fistula and (2) have received surgical treatment. The exclusion criteria included (1) children with life-threatening diseases and (2) children whose parents have refused treatment. Caregivers had to (1) be aged ≥18 years and (2) have cared for the child for at least four weeks without receiving payment for it. In the case of more than one primary caregiver, the one who had spent the most time with the child was enrolled. The exclusion criteria for primary caregivers included (1) having experienced major life events such as divorce or severe medical problems in the past three months (a short screening questionnaire was completed before participating) and (2) refusal to participate in the survey. Of the 254 caregivers, 18 declined to participate because of a lack of interest or the desire to keep information about the child's disease private. Two hundred and thirty-six primary caregivers completed the survey (participation rate = 92.91%). Seven samples were excluded from final analysis because of missing essential information or showing obvious self-contradiction. After deleting the

seven sets of ineffective data, 229 sets of data were finally used for analysis (effective response rate = 90.16%).

Procedures

In order to test the feasibility and suitability of the questionnaires, a pilot test including 30 caregivers of children with IA was conducted at an inpatient pediatric surgery department. First, 15 caregivers with educational levels ranging from primary school to college completed the pen-and-paper surveys to identify areas in need of improvement to ensure each question could be understood correctly. Then, an electronic questionnaire was created using wjx (www.wjx.cn), which another 15 caregivers completed online (the link of the questionnaire was sent through a social media site). They were also asked for advice on questionnaire modifications. Pilot test data were not used for the final analysis.

During the pilot study, three WeChat groups were set up. The WeChat group was a social media platform that everyone could see others' identity. In order to protect privacy, the caregivers could use nick names instead of their true names in the group. Medical staff would change their name to their medical identity, such as surgeon Qin, head nurse of pediatric surgery or wound ostomy continence nurse. The role of the group was to provide the caregivers with a platform to communicate with each other and consult medical staff and also to facilitate the survey process. A certified wound ostomy continence nurse in pediatric surgery and a pediatric surgeon were invited as consultants for each WeChat group.

The final version of the survey was completed online, the primary caregivers were enrolled during their child's follow-up at pediatric surgery outpatient clinics. During the follow-up, the primary caregivers were informed about the WeChat groups and survey and asked about their willingness to participate. They could join a WeChat group irrespective of whether or not they were willing to participate in the survey. If they were willing to participate in the survey, verbal informed consent was obtained, after which the principal researcher contacted them, sent the link to the questionnaire, and provided completion instructions. The link had two parts—the electronic informed consent and a set of questions. Only after the informed consent statement, "I have read all the content and agree to participate in the study," was agreed to could the caregivers continue with the questionnaire. The caregivers could either complete the survey on the spot in a separate room of the hospital or at their convenience.

Ethical considerations

Approval was obtained from the ethical review committee. All participants were given an introduction to the study and notified about their right to withdraw at any time. Informed consent was obtained from each participant, which indicated that they had understood the study in its entirety.

Measures

Independent variable: stigma

The Parent Stigma Scale was used to measure stigma perceived by primary caregivers. The scale comprises five items, each of which is rated on a five-point Likert scale from 1 (strongly disagree) to 5 (strongly agree), with total scores ranging from 5 to 25 and higher scores indicating higher perceptions of stigma (Austin, MacLeod, Dunn, Shen, & Perkins, 2004). The scale's reliability has been demonstrated in previous research, with Cronbach's alpha ranging from 0.76 to 0.81 (Austin et al., 2015). Its revised version has also demonstrated adequate reliability with a Cronbach's alpha of 0.65 (Rolston, Gardner, Vilain, & Sandberg, 2015). The Chinese Version of Parent Stigma Scale was first translated in this study, and minor modifications were made to make it more suitable to the context (e.g., "seizure condition" was

changed to "imperforate anus"). In this study, Cronbach's alpha was 0.883, and factor analysis showed that one factor alone accounted for 68.25% of the variance (Kaiser-Meyer-Olkin coefficient = 0.852, $P <$ 0.01), which indicated its reliability for stigma measurement.

Mediator: social support

The Social Support Scale (Xiao, 1994) was used to measure caregivers' social support. The scale, developed in the Chinese context, comprises three dimensions including subjective social support, objective social support, and social support utilization. This is a 10-item self-reported scale, and scored by comprehensive methods. Item one to four and eight to ten were rated by four-point response scales. Item five have five options and each option was rated on a four-point scale. For item six and seven, the scores were calculated on the number of support sources the caregivers chose. The total score of the full scale ranged from 12 to 66. Higher scores indicated more social support. The scale has proven reliability, with a Cronbach's alpha of 0.89, and is widely used in Chinese studies (Yang, Zhang, Meng, Liu, & Sun, 2019).

Dependent variable: distress

The Chinese version of the Kessler Psychological Distress (K10) Scale (https://www.hcp.med.harvard.edu/ncs/k6_scales.php) was used to measure caregivers' distress. On this 10-item self-report instrument, each item is assessed on a five-point scale from 1 (none of the time) to 5 (all of the time). Total scores range from 10 to 50, with higher scores indicative of greater distress (Kessler et al., 2003). The scores can be divided into four levels: scores of 10–15 are defined as low, 16–21 as moderate, 22–29 as high, and 30–50 as very high (Department of Health, Melbourne (2008)). In a study of Chinese parents of children with cancer, Cronbach's alpha for the scale was 0.93 (Bu et al., 2017). In this study, we used the K10 total score as a measure of distress. The categories were also calculated and discussed.

Covariates: sociodemographic and clinical information

Primary caregivers' sociodemographic information gathered included age, gender, marital status, educational level, occupation, relationship with the children, residence, household structure (extended/ nuclear), and religion. The children's sociodemographic and clinical information collected included age, gender, IA type, time since diagnosis (years), and medical insurance. Moreover, a question of concealment behavior–"whether you had intentionally concealed their children's disease in the social context" was added in the questionnaire.

Data Analysis

Categorical data were summarized using frequencies and proportions. Continuous data were expressed with mean and standard deviation or median (quartile). The differences between groups were analyzed using the Student's *t*-test, one-way analysis of variance, or Mann-Whitney *U* test. Bonferroni correction was used to counteract the problem of multiple comparisons. Analyses were conducted using SPSS version 21.0 (IBM Corporation, Armonk, NY, USA). The mediating effect of social support was estimated by the bootstrap method (Nevitt & Hancock, 2001) (random sampling with replacement to enlarge the sample, thus making the estimation more accurate) with 5000 samples, performed in IBM SPSS Amos 24.0 (IBM Corporation, Armonk, NY, USA). $P < 0.05$ was considered statistically significant.

Results

Descriptive statistics

The descriptive statistics are shown in Table 1. The median age of primary caregivers was 30 years (quartiles: 28 and 36), with a range of 18–67 years. The median age of the children with IA was 0.87 years (quartiles: 0.37 and 1.78), with a range of 0.08–6.37 years. The time

Table 1
Descriptive statistics for sociodemographic characteristics and univariate analysis (N = 229).

Item	N (%)	Distress	χ^2/t/z	P
Caregiver				
Age (years)			0.402	0.670
< 30	100 (43.7)	22.80 ± 7.33		
30–40	106 (46.3)	23.47 ± 7.30		
> 40	23 (10.0)	22.13 ± 8.56		
Gender			0.677	0.499
Male	41 (17.9)	23.76 ± 6.70		
Female	188 (82.1)	22.89 ± 7.58		
Marital status			−0.087	0.930
Married	222 (96.9)	23.04 ± 7.49		
Other	7 (3.1)	23.29 ± 5.44		
Educational level			0.543	0.653
Primary school or below	17 (7.4)	24.06 ± 6.83		
Junior high school	68 (29.7)	23.77 ± 6.81		
High school	45 (19.7)	22.91 ± 6.81		
University/college or above	99 (43.2)	23.04 ± 7.42		
Occupation			1.725	0.181
Part-time job	32 (14.0)	25.22 ± 6.50		
Full-time job	84 (36.7)	22.39 ± 7.72		
Unemployed	113 (49.3)	22.91 ± 7.40		
Relationship with patient			1.636	0.197
Mother	178 (77.7)	22.76 ± 7.44		
Father	44 (19.2)	24.64 ± 7.00		
Other	7 (3.1)	20.29 ± 8.90		
Residence			1.692	0.187
City	61 (26.6)	21.72 ± 7.61		
Suburban	76 (33.2)	22.99 ± 7.16		
Countryside	92 (40.2)	23.97 ± 7.46		
Household structure			−1.097	0.274
Extended family	150 (65.5)	22.65 ± 6.72		
Nuclear family	79 (34.5)	23.79 ± 8.60		
Religion			1.250	0.212
Yes	71 (31.0)	23.96 ± 8.62		
No	158 (69.0)	22.63 ± 6.81		
Children				
Age (years)			−0.849	0.397
< 2	189 (82.5)	22.85 ± 7.39		
≥ 2	40 (17.5)	23.95 ± 7.62		
Gender			0.062	0.951
Male	154 (67.2)	23.07 ± 7.18		
Female	75 (32.8)	23.00 ± 7.96		
IA type			5.136	0.007
Low	124 (54.1)	21.64 ± 6.63		
Intermediate	41 (17.9)	24.29 ± 9.60		
High	64 (27.9)	24.97 ± 6.82		
Time since diagnosis (years)			−1.995	0.047
< 1	128 (55.9)	22.18 ± 7.37		
≥ 1	101 (44.1)	24.14 ± 7.39		
Medical insurance			−1.633	0.104
Yes	173 (75.5)	22.59 ± 7.29		
No	56 (24.5)	24.45 ± 7.71		
Concealment		Stigma	4.455	< 0.01
Yes	161 (70.3)	15.86 ± 4.74		
No	68 (29.7)	12.88 ± 4.36		

since diagnosis ranged from 0.08 to 6.37 years with a median of 0.86 (0.34, 1.76). The scores of stigma, social support, and distress are depicted in Table 2. The numbers and proportions of caregivers at

Table 2
Descriptive statistics for measurement scales (N = 229).

	Total score (range)	Item average value (range)
Stigma	14.98 ± 4.81 (5.00–25.00)	3.00 ± 0.96 (1.00–5.00)
Social support	40.33 ± 8.36 (16.00–62.00)	2.88 ± 0.60 (1.14–4.43)
SSS	23.87 ± 5.24 (9.00–32.00)	2.98 ± 0.65 (1.13–4.00)
OSU	9.51 ± 3.17 (1.00–18.00)	3.17 ± 1.06 (0.33–6.00)
SSU	6.95 ± 2.02 (3.00–12.00)	2.32 ± 0.67 (1.00–4.00)
Distress	23.04 ± 7.42 (10.00–50.00)	2.30 ± 0.74 (1.00–5.00)

Note: SSS = subjective social support; OSU = objective social support; SSU = social support utilization.

different distress levels were as follows—low: 34 (14.8%), moderate: 74 (32.3%), high: 75 (32.8%), and very high: 46 (20.1%).

There were significant differences in caregivers' distress with regard to IA type ($t = 5.136, p < 0.01$) and time since diagnosis ($t = -1.995, p < 0.05$). Distress was highest among caregivers of children diagnosed with intermediate or high IA and those who had been diagnosed more than a year ago. About 70% caregivers had concealment behavior, and conceal behavior was associated with higher level of stigma ($t = 4.455, p < 0.01$).

Correlations between stigma, social support, and distress

Pearson's correlation analysis (Table 3) showed that stigma was positively correlated with distress ($r = 0.396, P < 0.01$) and social support was negatively associated with distress ($r = -0.413, P < 0.01$) in all three dimensions ($r = 0.314–0.346, P < 0.01$), which indicated a moderate effect. Stigma was also negatively associated with social support ($r = -0.280, P < 0.01$), mainly in subjective social support ($r = -0.278, P < 0.01$) and social support utilization ($r = -0.265, P < 0.01$). Cronbach's alphas for the Parent Stigma Scale, Social Support Scale, and K10 Scale were 0.883, 0.819, and 0.933, respectively, which showed good reliability.

Model test

The relationships between stigma, social support, and distress were tested using structural equation modeling while controlling the covariates (IA types and time since diagnosis) (Fig. 1). The results suggested that stigma positively predicted distress ($\beta = 0.240, P < 0.01$) and social support negatively predicted distress ($\beta = -0.40, P < 0.01$). Stigma and social support could explain 31% of the variance in caregivers' distress. Stigma negatively predicted social support ($\beta = -0.34, P < 0.01$) and explained 12% of the variance in social support. Moreover, social support partially mediated the relationship between stigma and distress [indirect effect = 0.135, 95% confidence interval (CI): 0.072–0.233]. The analysis of the structural equation model yielded a good model fit (Iacobucci, 2010; Wang, Tsai, & Tsai, 2014) (Table 4).
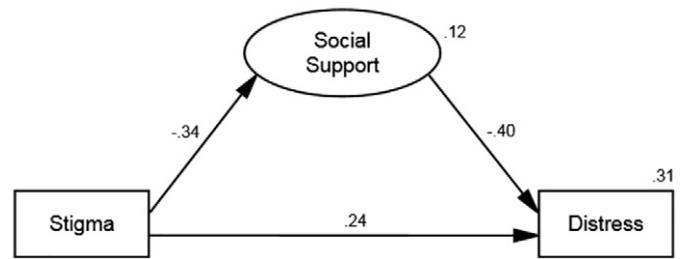
Discussion

The purpose of this study was to explore whether the effect of stigma on the distress experienced by primary caregivers of children with IA was mediated by social support. As hypothesized, the caregivers of children with IA were likely to have a high level of distress on average, and a considerable proportion (52.9%) experienced high or very high levels of distress. Stigma was found to have a moderate positive correlation with distress, and this relationship was partially mediated by social support.

Fig. 1. Model depicting the mediating role of social support in the association between stigma and distress. Note: Direct effect = −0.240, indirect effect = 0.135, total effect = 0.375. Values on the path are standardized coefficients. The path coefficient tests were all statistically significant ($P < 0.001$).

Sociodemographic/clinical variables and caregivers' distress

The results of this study suggested that caregivers of children with intermediate and high IA reported higher levels of distress than caregivers of children with low IA. As intermediate and high IA requires multiple operations, treatment is longer and more expensive than is the case with low IA, which could greatly increase caregivers' burden, leading to high distress levels (Hegde, Chakrabarti, & Grover, 2019). Additionally, the higher the malformation position, the worse the prognosis (Nam, Kim, & Kim, 2016). Children with intermediate and high IA might experience worse outcomes than children with low IA owing to poorly developed interior sphincters (Wang, Hu, Jin, Li, & Xu, 2016), causing their caregivers to worry about their future, leading to higher distress levels. Furthermore, caregivers of children who had been diagnosed more than a year ago had higher levels of distress than caregivers of children who had been diagnosed more recently, contradictory to a previous study (Witvliet, Bakx, Zwaveling, van Dijk, & van der Steeg, 2016). One possible explanation might be the differences in the populations studied. In China, many family members are usually involved in children's care during the first year of an illness, but later, the task of caregiving is usually taken over by one person. Additionally, as children grow and begin feeling self-conscious, it may become difficult to get them to cooperate with the treatment because of the invasion of their privacy (Noiseux, Rich, Bouchard, Noronha, & Carnevale, 2019), contributing to greater distress among caregivers.

Stigma, social support, and distress

Recently, stigma has gained attention in special populations such as patients with psychological or congenital diseases and their caregivers. An increasing number of studies now focus on caregivers' stigma, also called affiliate stigma, which refers to the fact that caregivers can also experience self-stigma because of their close relationships with stigmatized individuals (Chang et al., 2015). It is very important for medical

Table 3
Correlations (r) between stigma, social support, and distress.

	Stigma	Social support	SSS	OSU	SSU	Distress
Stigma	1					
Social support	−0.280***	1				
SSS	−0.278***	0.882***	1			
OSU	−0.110	0.727***	0.378***	1		
SSU	−0.265***	0.705***	0.462***	0.457***	1	
Distress	0.396***	−0.413***	0.346***	0.314***	0.320***	1

Note: SSS = subjective social support; OSU = objective social support; SSU = social support utilization.
*** *P* value <0.001.

Table 4
Fit indices.

Fit indices	Model value[1]	Model value[2]	Reference value	Overall model fit
χ^2/df	2.021	2.010	1–3	Good
GFI	0.986	0.970	> 0.90	Good
AGFI	0.948	0.936	> 0.90	Good
CFI	0.980	0.940	> 0.90	Good
IFI	0.980	0.942	> 0.90	Good
TLI	0.949	0.902	> 0.90	Good
SRMR	0.0324	0.0695	< 0.05	Acceptable
RMSEA	0.067	0.067	< 0.08	Good

Note: Model value 1: original model that did not control for the effect of IA type and time since diagnosis; Model value 2: the model that controlled for covariates. GFI = goodness-of-fit index; AGFI = adjusted goodness-of-fit index; CFI = comparative fit index; IFI = Bollen's incremental fit index; TLI = Tucker-Lewis index; SRMR = standardized root mean square residual; RMSEA = root mean square error of approximation.

staff to understand the stigma of caregivers because they are the link between health care professionals and patients. High stigma among caregivers may result in low cooperation with patients' care, thus leading to reduced care quality for patients (Chang et al., 2015; Nyblade et al., 2019).

The results of this study demonstrated that stigma had both direct and indirect effects on the distress experienced by caregivers of children with IA. Caregivers' stigma is usually associated with embarrassment about their situation (Chai, Mahadevan, Ng, Chan, & Dai, 2018) and fear of diagnosis disclosure (Turner, Biesecker, Leib, Biesecker, & Peters, 2007). Even in this study, caregivers who had concealment behavior were found to have a higher level of stigma than those who did not reveal concealment behavior. Furthermore, caregivers with stigma might perceive greater strain and burden in their daily caregiving because of a distortion in their personal view of the patients (Mark & Cheung, 2008). Feeling burdened by the unfriendly and discriminatory environment, they may experience difficulties in caring for the affected children, ultimately feeling a high level of distress. Therefore, medical professionals should pay more attention to stigma among caregivers of children with IA, and targeted stigma reduction interventions, especially for those who have concealment behavior, should be developed to replace the passive tactic of concealment. Presentations, videos, and discussions could be used to help caregivers gain more knowledge about stigma and help change their attitudes (Bamgbade, Barner, & Ford, 2017; Bamgbade, Ford, & Barner, 2016). Effective strategies are required to be developed instead of concealment, to help reduce the stigma of caregivers of children with IA.

Moreover, caregivers with high levels of reported stigma isolate themselves because of the fear of disclosure and possible discrimination, causing social distancing and limiting the social support they receive (Li et al., 2017). As social support plays a protective role in reducing distress, this would indirectly impair caregivers' psychological well-being (Choi et al., 2016). In this study, social support had a moderate negative relationship with distress. Another study showed that enhanced social support could reduce caregivers' stress and improve their coping skills (Mendoza, Fruhauf, & MacPhee, 2019). Thus, support-oriented interventions should be implemented to help the caregivers to gain more help from their family members and peers.

Limitations

Our study had several limitations. First, owing to the cross-sectional design, we failed to investigate the different stages of distress for each primary caregiver, which might have helped identify the pattern of psychological distress change over time. Second, owing to the correlational relationships between the variables, the directions of the associations cannot be firmly established. For example, social support may also ease stigma. Future studies must develop more detailed, multi-dimensional measures of stigma.

Implications for clinical practice

The caregivers of children of IA were under high level of distress. Interventions focused on stigma should be developed, such as presentations, videos program and group discussions to help caregivers change their attitudes of stigma, thus reduce perception of stigma. Also, medical professionals should help enhance social support among caregivers, especially those with the potential to increase the subjective social support and social support utilization of caregivers of patients with IA. For example, we can provide caregivers with more approaches for contacts with medical staff, and encourage them to seek help from their family members and other parents who with similar situations (Sari et al., 2014). These measures could benefit the psychological health of both caregivers and children (Grano, Bucci, Aminoff, Lucidi, & Violani, 2013; Marieke et al., 2013).

Conclusions

On average, primary caregivers of children with IA experienced high levels of distress, with the majority experiencing high or very high distress. Moreover, stigma could increase caregivers' distress. Social support was confirmed to potentially mediate the relationship between stigma and distress, and it could decrease the effect of stigma, thus alleviating caregivers' distress. Assessing and reducing stigma are crucial for enhancing social support and mitigating distress among caregivers of children with IA. Future studies should further explore the stigma of caregivers of children with IA, including the assessment and coping strategies of their stigma. Also, more protective factors associated with distress should be studied to help decrease caregivers' distress and benefit their mental health.

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Declaration of Competing Interest

None.

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