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Stigma in youth with Tourette's syndrome: a systematic review and synthesis

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Abstract: Tourette's syndrome (TS) is a childhood onset neurodevelopmental disorder, characterised by tics. To our knowledge, no systematic reviews exist which focus on examining the body of literature on stigma in association with children and adolescents with TS. The aim of the article is to provide a review of the existing research on a) social stigma in relation to children and adolescents with TS, b) self- stigma and c) courtesy stigma in family members of youth with TS. Three electronic databases were searched: PsycINFO, PubMed and Web of Science. Seventeen empirical studies met the inclusion criteria. In relation to social stigma in rating their own beliefs and behavioural intentions, youth who did not have TS showed an unfavourable attitude towards individuals with TS in comparison to typically developed peers. Meanwhile in their own narratives about their lives young people with TS themselves described some form of devaluation from others as a response to their disorder. Self- degrading comments were denoted in a number of studies in which the children pointed out stereotypical views that they had adopted about themselves. Finally, as regards courtesy stigma, parents expressed guilt in relation to their children's condition and social alienation as a result of the disorder. Surprisingly however there is not one study that focuses primarily on stigma in relation to TS and further studies that examine the subject from the perspective of both the 'stigmatiser' and the recipient of stigma are warranted.

Keywords: Tourette's syndrome, tics, stigma, bullying, discrimination, social rejection

Introduction

Tourette syndrome (TS) is a childhood onset neurodevelopmental disorder characterised by tics, namely sudden repetitive movements or vocalisations. In order for a diagnosis to be ascertained, multiple motor and at least one vocal tic must be present for at least one year [1, 2]. Tics have been classified into simple and complex types.

The former are abrupt and brief in duration. Characteristic examples of simple motor tics include excessive eye-blinking while sniffing, and throat clearing can be embedded in the category of simple vocal tics. In contrast to simple tics, complex motor tics involve groups of muscles and their duration is longer. They may appear as purposeful movements, but they serve no function. Typical examples of complex tics are unusual smelling objects and inappropriate offensive gestures with hands or tongue (copropraxia) [3]. Complex vocal tics can also be mistakenly perceived as purposeful utterances. Thus, in this category, words and phrases are verbalised. Echophenomena, such as echolalia (repeating the words of others) and paralilia (repeating his/her own words) are quite frequent. It is within this group that the controversial feature of coprolalia, thus the utterance of obscenities, is embedded. Studies also suggest that individuals with TS have an urge to make socially inappropriate and disruptive comments (NOSIS non-obscene complex socially inappropriate symptoms) [4].

There is a large body of evidence that suggests that TS is not a unitary condition and individuals with TS exhibit a multifaceted and complex clinical picture, since the majority also receive an additional neuropsychiatric disorder diagnosis, with ADHD and OCD comprising the two most common comorbidities [5].

Studies have reported that the complexity of tics, their duration and their intensity as well as the presence of comorbidities may affect the social and adaptive functioning of an individual with TS. Thus, complex tics like coprolalia or even NOSIS may be more salient, socially unacceptable and therefore linked to social stigmatisation, rejection and diminished quality of life (QoL) [6].

Indeed, beside the hurdles children and adolescents with TS endure because of the manifestation of tics, narrative testimonies of youth with TS provide extensive accounts of stigmatisation, harassment, bullying and teasing [7]. It is therefore the aim of the current systematic review to identify the relevant publications in the literature relating to stigma in TS.

Conceptualisation of social stigma

The word stigma is originally derived from the ancient Greek verb $\sigma\tau\acute{\iota}\zeta\omega$ which referred to the creation of a scar upon the bodily flesh by a sharp instrument through the process of pricking, cauterizing or carving [8]. According to Herodotus it either represented a mark of ownership in slaves (Hdt 5,35) or signified disgrace (Hdt 7, 35). Since the publication of Goffman's milestone monograph (1963) "Stigma: Notes of the Management of Spoiled Identity", the concept of stigma has been subject to elaboration and conceptual variation in contemporary literature. Goffman [9] defined it as a visible or invisible mark or attribute that reduces the bearer from a whole and "usual" person to a tainted one. This "blemish" [9] prohibits the stigmatised from being fully accepted in a specific social setting [10] which underlines the dynamic, relational and contextual nature of the "scar" [11].

Stigma can be viewed as a complex social construct that can exist when cognitive, affective and behavioural components of the 'stigmatizer' (the person who is likely to stigmatize another) converge [12]. The cognitive

component refers to negative stereotyped knowledge and beliefs. Stereotypes are sets of cognitive schemas that reflect the assumed attributes and distinctive characteristics that a category of people possess [13]. They are perpetuated by ignorance and a lack of knowledge [14]. The affective domain, prejudice, refers to the negative feelings and emotions that the stigmatiser engenders as a response to the endorsement of the negative stereotypes [15]. The behavioural dimension is associated with the enacted discrimination against the object of stigmatisation, not solely actively harming and disadvantaging the targeted individuals but also segregating or coercing them. During childhood and adolescence bullying fits under the wider umbrella of discrimination [16]. The term bullying refers to both proactive types of aggression and covertly manipulative forms of victimisation [17]. The former category involves overt physical and verbal aggression from the bully to the victim. Indirect bullying on the other hand involves the presence of a third party. This category encompasses relational forms of bullying, such as spreading rumours about somebody or excluding them intentionally from activities in an attempt to negatively impact their social status.

Self-Stigma

Self-stigma refers to the internalisation of social stigma by an individual who has been devalued and dehumanised. Thus, those who experience self-stigmatization tend to accept negative stereotypes that are propelled by society. In turn the individual experiences shame, a loss of self-esteem which leads to hiding their “spoiled social identity” by withdrawal from social opportunities [18]. Self-stigma also involves the anticipation of rejection by others. It should be emphasised however, that not all individuals who endure public stigma internalise the negative societal messages [19]. In the case of Tourette’s a number of people are energised and empowered by their individuality, which at times they exhibit through the media

(e.g. The Undatable, <http://www.channel4.com/programmes/the-undateables/profiles/all/ruth>, Touretteshero <http://www.touretteshero.com/>).

Courtesy and Affiliate stigma

Stigma may also affect people who are closely associated with a person who has been discredited, such as immediate family and friends, who may be blamed or considered responsible for the condition of their relative. A considerable amount of literature has documented that family members, especially caregivers, tend to internalize the devaluing views of others and feel guilt or self-blame in relation to their relative’s condition [20]. The self-stigmatisation of family members has been referred to as affiliate stigma [21]. As a consequence caregivers tend to conceal their status and isolate themselves.

Stigma and Tourette’s syndrome

The six dimensional approach to stigma proposed by Jones et al. [22] may suggest that the very nature and symptomology of TS could result in an individual with the disorder being stigmatised. The first dimension is concealability. In the case of Tourette’s, tics may be highly visible so that in contrast to other disorders, individuals with TS cannot hide their condition for a prolonged period of time. Another dimension of stigma is

the origin of the mark which has a direct impact on the perceived responsibility the marked person has over their condition. The abrupt movements and vocalizations might be perceived by the observer as controllable and the observer may also feel that people with TS are accountable for their actions. This may be especially relevant to swearing tics and NOSIS. Perceived deliberate actions may attract angry responses and may justify withholding prosocial behaviour from the stigmatised. The disruptiveness of the tics and the manner in which they hamper, strain and interrupt interpersonal interaction and communication can be associated to the dimension that Jones et al. [22] have identified as disruptiveness. Lastly the abrupt, odd and sometimes painful movements of an individual with TS may not appear visually pleasing which highlights the dimension of aesthetics [23].

Tourette's is also a syndrome that has mostly been misunderstood by the public and mocked by the media [24-25]. The stereotypical image of a person with unpredictable and unstable behaviour who has the irresistible urge to utter obscenities has been perpetuated within popular culture through comedic films like "Not another Teen movie". Thus, despite the fact that an unprecedented volume of information is available to the public about TS, the unintentional and involuntary basis of these symptoms have not been adequately pointed out and accurate information about the syndrome remain scarce.

Some researchers have proposed that it is also possible that people with Tourette's have some cognitive vulnerabilities that make socially inappropriate and stigmatising behaviour more likely. For example, Eddy and colleagues have considered the possibility that people with TS may have social cognition impairments and more specifically deficits in relation to theory of mind [26-27]. In particular TS may be associated with hyper-mentalizing, (i.e. over- thinking and making exaggerated assumptions about the mental state of others based on the information that's available). This deficit in combination with impulse dyscontrol and urges to exhibit dangerous behaviour could result in individuals with TS being more likely to exhibit NOSIS. Thus, over-thinking about others' metal state and a compulsion to exhibit 'socially dangerous actions' may possibly lead to socially inappropriate behaviour which could reinforce public stigma towards individuals with TS [28].

Aims and objectives of the current systematic review

To our knowledge, there have been no systematic reviews that focus on examining the body of literature on stigma relating to children and adolescents with Tourette's syndrome. It is therefore the aim of this study to systematically review the existing research on a) social stigma in relation to children and adolescents with TS b) self- stigma, and c) courtesy stigma in family members of children and adolescents with TS. More analytically, the aims of this review are as follows:

- To identify publications which explore the public's stereotypical knowledge about TS, including unfavourable attitudes and discriminating behaviour towards youth with TS.
- To understand the origins of any negative attitudes and behaviour from the point of view of the individuals who exclude and marginalise youth with TS.
- To identify if stigma is associated with age or gender in youth with TS and to ascertain if gender and age could be identified as characteristics that could predict the perpetrators' behaviour towards individuals with TS.
- To provide an overview on how stigma is experienced and managed by youth with TS and to point out the overt and subtle forms of discrimination they endure.
- To evaluate the self-stigma in individuals with TS and identify coping mechanisms.
- To assess courtesy and affiliate stigma in family members.

The objective of this review is to summarize, evaluate and provide a comprehensive overview of the current knowledge and evidence and to indicate future research directions.

Methods

Research strategy

To identify studies for inclusion in the current systematic review three electronic databases that included a wide range of journals were searched: PsycINFO, PubMed, and Web of Science. Search terms focused on two areas: Tourette* and stigma. Each of the keywords of Tourette's was searched alongside each of the stigma words.

[Table 1 here]

Inclusion and exclusion criteria

Only empirically based studies that used explanatory, observational, experimental or survey study design and were published in peer review journals after 1994 and before 2015, were included. As the degree of stigma tends to change over time, these date limits were set to guarantee that the literature accurately reflects society's conceptualisation of TS through the past two decades.

Predefined eligibility criteria identified as relevant any study that directly evaluated the public's awareness of Tourette's syndrome or examined the attitudes, the behavioural intentions and exclusionary practices of other groups towards youth with TS. Studies were also included if they assessed stigma as seen from the perspective of the children and adolescents with TS or their families. Experiences of stigma and bullying during childhood and puberty that were retrospectively reported by adults with TS were also considered eligible for this review. Studies were also included if they explored the experience of self-stigma in children and adolescents with TS or courtesy/ affiliate stigma of family members.

Studies have demonstrated that the findings pertaining to how stigma is enacted and experienced among adults cannot always be generalized to younger population [29]. Since the aim of this study was to explore the potential effects stigma may have on the well-being and ongoing development of children/adolescents with TS, articles that related to adults experiences of stigma were excluded. Studies were also eliminated if they only measured the attitudes and behavioural intentions towards individuals with TS after a form of intervention had been employed without evaluating the views of the participants before that intervention was implemented. Grey and non-English language literature were excluded, as were book chapters, reviews and dissertations.

Results

Included studies

The research from the three chosen database searches yielded 3635 results of which 19 peer reviewed articles met inclusion criteria. Only 17 of these reported data from independent projects as two sets of articles discussed data from the same study [30-48]. (Figure 1 about here)

The 17 relevant studies were diverse in their primary research focus and design (Table 2). Thus, 6 studies explored the experience of having TS in relation to everyday life [41-45, 47-48]. Four studies used an experimental study design to explore the social acceptability of individuals with TS in comparison to individuals that did not have TS or the participants' knowledge about the disorder before and after an educational intervention had been implemented [30, 31, 34, 39]. Four studies that used survey designs explored the knowledge that particular sections of the population had about TS and attempted to estimate the rates of victimization among youths with TS [32, 33, 35, 36, 40]. One large survey explored the impact of TS on the lives of individuals with the disorder [37-38]. Finally, one study delved into the clinical characteristics of a population of people with TS in Costa Rica [46].

Population and countries

The study sample in the reviewed studies (n=2,322) were either individuals who had been diagnosed with TS (n=793, 34.2 %), their parents (n=861, 37.1 %) or some segment of the general population (n= 668, 28.7%). The latter category consisted of healthcare students and professionals (n= 174), school teachers (n=137) and school aged children (n=357).The majority of the studies were conducted in the United States of America (9) followed by the United Kingdom (3) Spain (2) Australia (1) Denmark (1) and Costa Rica (1).

Data synthesis

Regarding the qualitative studies data were extracted mainly from the results section of the publication by the first author. Thematic synthesis was performed in the qualitative and mixed- methods studies according to the approach described by Thomas and Harden [49].

Quantitative studies

Awareness of TS

Four studies explored knowledge about TS among different types of people [32, 34, 39, 40]. Teachers and elementary school children revealed only a basic knowledge about TS [39, 34], with teachers displaying a better knowledge of management strategies than etiological causes of TS. This indicates the need for disorder specific training. The surveys on professionals [32] and healthcare students [40] suggested an adequate knowledge on TS. The generalizability of the rates is questionable however due to the small sample sizes and the low response rates; only 6.6% of the targeted sample participated in the survey of Katona [40] and 17.5% of the professionals in the study of Marcks [32].

Questions of prognosis, treatment and contagiousness were not addressed in any of the aforementioned quantitative research with studies based on questionnaires that were developed by the authors to measure knowledge about Tourette's. Researchers also failed to report any details of the validity of the measures. Most importantly, the articles did not highlight the myths and misconceptions that are endorsed by the aforementioned populations in order for more realistic and tailor- made educational interventions to be developed.

Attested attitudes and behavioural intentions towards children and adolescents with TS

The studies that pertained to the attitude [30, 31, 34, 40] and behavioural intentions of the participants towards an individual with TS [30,,34] indicated the negative impact that tics have on social interaction. In rating their own views, youth without TS showed less positive attitudes towards youth with TS in comparison to typically developing peers [30,31]. However, in comparison to other studies that utilise the same assessment tool (Adjective Checklist) individuals with TS were rated more positively than children with ADHD [50], autism [51] or obesity [52] by their peers. With respect to gender, boys appeared to have a more positive attitude towards children with TS than did girls [30] and adolescent females with TS were seen less favourably than females without tics [31].

Only one study that evaluated the behavioural intention towards a child with TS suggested that youth showed no reluctance to engage in social activities with the individual with TS [30]. However, only 60% of the initial sample completed the specific questionnaire and low response rates may indicate misleading results.

The findings of the studies should also be interpreted with caution due to the fact that in some the artificially created stimuli which introduced the participants to the symptomology of TS lacked authenticity [30,,31]. Thus videos used a child/ adolescent actor portraying solely motor tics although phonic tics which could be more destructive to the interpersonal interaction in comparison to the motor ones [35] are basic components of TS [1]. By presenting only one element of the symptomology of TS, the participants' evaluation may have been positively affected. Moreover, Holtz & Tessman [34] did not use a visual stimulus or a written vignette to introduce the participants to the symptomology of TS. Thus, the measures were completed without the

participants having any clear indication of the manner in which the disorder manifests itself. Therefore the results may not truly reflect the real attitude they would have towards a peer with TS but rather their fear of an unknown syndrome that is considered socially unacceptable. In addition none of the studies controlled for level of contact with individuals who exhibit non-typical behaviour through close family members or friends so as to investigate the variables or the combination of variables that could predict more or less stigmatising attitudes.

Discriminatory behaviour and Bullying

Discrimination was only addressed by one survey, in which the informants were parents [37]. Four questions focused on the subject of prejudice and disadvantageous treatment and the results revealed that 75% of the respondents felt that their child had been treated differently because of their tics.

Four studies attempted to estimate the rates of peer victimisation in children with TS [33, 35, 36, 38]. The discrepancies between the findings, which ranged from 26% [38] to 44.7% [36], can be interpreted by the variety of measures that were employed and the different informants that were used for the studies. In the two studies that measured self-reported peer victimisation approximately one fourth of the individuals with Tourette's reported being the target of bullying [35, 38]. The studies that based their estimates on the reports from the parents of individuals with TS [33, 36] reported extremely high rates of bullying although they focused solely on one distinct aspect of victimisation, namely teasing. It should be noted however that a detailed definition of teasing had not been provided to the research participants in these studies and the overestimation may be a result of poor adherence to the basic meaning of the concept. Furthermore, in cross-national studies the comparability of the terminology is tenuous [53]. The results of the studies are further confounded by ambiguous response options. Taking into consideration that one of the basic characteristics of bullying is repetitiveness, by measuring the frequency of the incidents with the use of vague terms such as "sometimes" or "most of the time" the respondents are endorsed to use their personal frame and interpretation [54]. In other words there is no specific reference period provided e.g. the past 30 days, to which the participants should relate their assessment. Moreover, the cut-off point that differentiated the victims from the non-victims appeared to be fairly arbitrary. However, it was pointed out by Solberg and Olweus [55] who examined the psychosocial adjustment of students from the general school population that had admitted to past experience of being bullied, that the lower cut off point that separated the victim status from the non-victim was being bullied two to three times a month. Students who were bullied more than 2-3 per month exhibited a different psychosocial profile and were less socially integrated, experienced depression and had lower self-esteem than less bullied students. In reference to the forms of bullying individuals with TS endure, solely Zinner [38] examined every specific form of bullying separately, suggesting that individuals with TS were more likely to endure verbal and relational bullying than they were to experience physical aggression.

Although there are discrepancies between the findings the limited available data suggests that the prevalence rates of bullying among individuals with TS is significantly higher than the estimates among the typically developing school age population (9%) or youth with other disabilities (9%)[34].

Another confounding issue absent from the studies in the review was cyberbullying in relation to individuals with TS which is pertinent as it ensures the anonymity and unaccountability of the perpetrators [56].

Qualitative Studies

Surprisingly, no single qualitative study currently exists that focuses primarily on the experience of stigma in relation to TS. Nevertheless, by studying the articles providing narratives of individuals with TS, the concept of stigma or the components of it emerge [41-48]. It should be noted however, that the perceived public stigma in the reports of individuals with TS may also reflect and be influenced by self-stigma depression, anxiety [5] or anticipated rejection [57].

Misconceptions and stereotypes about the disorder

The majority of participants described some form of devaluation as a response to their tics. This is perhaps unsurprising given that in many parts of the world causal beliefs of TS are shrouded in misconceptions, and sometimes linked to demonic possession. For example, a teacher within a Christian school in Australia suggested that tics were caused by possession of the devil [47]. Similarly in Indiana under similar circumstances an improvised exorcism was undertaken which further contributed to the isolation and withdraw of the individual with TS [42].

Such misconceptions are further complicated by the stereotypes that have been linked to TS and most specifically coprolalia. The individual with TS is deemed to always be associated to the uncontrollable fits of profanity. “When you say *I’ve got Tourettes* people will-have been-are like waiting for you to swear and stuff, and I’m saying no, it’s not swearing. Every person, apart from a couple, have said oh you don’t swear, you don’t swear. And it just get right on my- right – gets gets yeah – *it’s really annoying* (Hayden)[44].

Discrimination within school settings

Although it should be pointed out that this is not the norm and that in general, at least in the UK, children with TS will be provided with additional support within educational settings including extra time for exams [48] a subtle form of discrimination has been found to take place in some schools where the teachers have been unwilling or unaware of how to make allowances in order to cater to the individualised needs of children with TS [45]. Characteristically, a number of educators were reluctant to repeat questions, and/or allow the students to leave the room in order to take their medication or make reasonable adjustments to accommodate the needs of

individuals with TS. The educators justified this form of inflexibility by arguing that the greater demands of the classroom should exceed the individual needs of a single student [47].

Bullying and the aftermath

Qualitative descriptions of the behavioural experience of bullying were very poorly reported, insufficiently descriptive and did not provide a thorough insight into the experience of victimisation. However, despite the diverse settings and populations the theme that prominently emerged was the positive relation between name calling, ridicule and individuals with TS [41, 45, 46 47]. The subsequent aggressive and sometimes violent responses of the victims were also recorded. *“They like call me Tourettehead and stuff, I just end up kicking them or something, as I just get really angry and start kicking them and they start punching me, doing that back. And I end up falling out with them”* (G2, P7) [48]

Moreover, victimisation was linked to social isolation .I feel like somehow get left out of all the games, and I feel like a dork and that stuff, and *I really don't have much friends...and mostly I get picket on at school”*(Kyle)[47].

Only one study examining the experience of adolescents with TS reported that the participants had never endured victimisation [44]. Taking into account the small sample number and their involvement in TS organisations it is unclear to what extent this group represented individuals with TS in general.

Self-stigma in children and adolescents with Tourette's syndrome

Self- degrading comments about their limited mental capacity dominated a number of the interviews in which the children pointed out stereotypical views that they had adopted about themselves, their diminished confidence in their future and the impact their tics may have in future employment opportunities [44]. For example:

Interviewer: Why haven't you had any luck do you think [in finding a job]?

John: Too dumb I suppose (laugh followed by a flurry of tics) Yeah, too stupid! (John)[47].

By masking their tics, concealing their “spoiled” [7] identity, avoiding being in the company of strangers and withholding the distressing fact that they had TS because of the anticipated fear of being socially rejected, an underlying self-stigma is evident [44]. *“If I do noises they're like ‘oh that's Tourettes isn't it’ and I think [few words inaudible] and I'm scared they'll start thinking I'm weird and my relationships will go. I'm scared of that”* (Pilot, P2) [48]

Courtesy and Affiliation stigma

Rivera- Navarro [43] found that a number of caregivers felt they were discredited as parents having been accused by medical professionals of magnifying the symptoms of their children. . Some parents also reported constrained relationships with friends and relatives and marginalisation as a response to their children's TS. This form of intolerance impelled them to become socially isolated and alienated from both their friends and their extended family [45].

A number of qualitative studies also indicated affiliate stigma in families [43, 45]. Parents' guilt and self-blame after learning about the inheritability of the syndrome may have inhibited them from seeking professional help and guidance [43].

Discussion

Eddy [6] has documented that youth with TS have diminished Quality of life (QoL) in comparison to typically developing peers or children and adolescents with epilepsy. The lower scores in the relationship domain suggest that restricted social life and interaction difficulties in particular may be mediated by the negative impact of social rejection and stigmatisation [58]. Thus, low social activity engagement which creates lower QoL and poorer emotional functioning [59], may be due to the stigma of socially unacceptable behaviour. Yet, this systematic review revealed a relative scarcity of data concerning the stigma, public's awareness, attitudes, and behavioural intentions towards individuals with TS. Also, convenience and highly educated samples might have presented an underestimation of the misconceptions about the disorder [32, 39]. Furthermore, there is a dearth of research concerning the way stigma is constructed and interpreted by the perpetrators in relation to TS. Hence, the studies to date do not give the participants the opportunity to freely express their thoughts and feelings in order to gain rich qualitative data on the origins of stigma. By turning the attention away from the producers of the exclusion, stigma is seen as something within the individual, rather than a designation that others affix on the person. There is also a lack of research that has delved into the motives, the mechanisms and reasoning involved in the exclusion of individuals with TS from social interaction. In other words, the psychological functions that are being served by the exclusion of an individual with TS from a group have not been revealed and interpreted from the perspective of "typical" children. Moreover, whether specific peers, or rather peer groups in general, stigmatise and subsequently exclude individuals with TS has not been assessed. Hence, research needs to investigate if the devaluation of individuals with TS is a socially sanctioned behaviour among children and adolescents. An understanding of the aforementioned subjects could provide a basis for developing effective and tailor-made interventions aimed at reducing the misconceptions which lead to the social stigma associated with the condition.

Because of the variety of assessment methods and the small sample sizes used, no gender or age distinction in stigma amongst children and adolescents with Tourette's could be determined. Furthermore, gender and age could not be identified as characteristic in ascertaining attitudes and behaviour towards individuals with TS. Similarly, the different assessment methods utilised did not permit an exploration of the kind of victimisation that is prominently associated to TS. No association between age and victimisation was revealed in any of the studies and only one report suggested that boys with TS reported higher levels of peer victimisation [35].

Another issue arising from this review is that studies are warranted that explicitly address public stigma and the specific ways discrimination is enacted through the perspective of the individuals with tics themselves.

Surprisingly, there is not a single study that focuses on the experience of stigma in relation to TS; the fragmentary testimonies available paint a vague and partial picture. By examining the subjective views of those who are exposed to stigmatising attitudes and discriminatory practices, valuable insight could be provided about how the virtual social identity of TS affects everyday life and the personal identity. As Sartorius [59] argues, interventions that aim to combat stigma should be based on what stigmatised individuals highlight as more deprecating and devaluing in the way they are viewed and treated by the society.

Internalised stigma is an important subject to address since it is associated with hopelessness and may therefore have adverse effects on the individual's subjective quality of life [60]. Future research is also required into courtesy-stigma in families, especially siblings and extended family because research so far is limited.

Limitations

The studies included in the review were conducted mainly in the U.S.A and in the UK, thus mostly reflecting stigma of TS in relation to western society. No data on stigma associated to TS in Eastern and African countries was reviewed although this is likely to be due to our exclusion of non- English language studies. Although the tendency to stigmatise is universal and omnipresent, it is not a static concept but a phenomenon that is dictated by cultural and historical forces [62]. Future research therefore needs to address these limitations. A further failing in the studies was the small and non-diverse sample of individuals with TS presented. Thus, a considerable amount of the population under study was drawn from speciality tic clinics, thus representing the most severe end of the TS spectrum and excluding the population for whom TS does not necessarily impact on daily living activities. In addition, a large proportion of the articles reported studies of individuals with ADHD or OCD and TS as a homogeneous group even though the additional presence of ADHD can result in added social difficulties for the individual with TS [63]. Finally, children and adolescents that had been involved in TS organisations and support groups represented a notable percentage of youth studied. These individuals may have de facto been more willing to request support compared to their counterparts who were not attached to such TS groups and subsequently the data collected was possibly skewed towards TS group/organisation influences.

This systematic review also has limitations. Because of the nature of the term 'stigma' precise search terms were difficult to identify, and although every effort was made to capture the relevant literature it is possible that some articles were missed. Also, due to publication bias, it is possible the literature included was biased towards studies with significant results. Also, due to publication bias grey literature was not included which impeded a comprehensive search for all relevant information. Finally, the exclusion of articles published in languages other than English limits the scope of the research.

Conclusion

The main results of the present systematic review suggest that although there is an association between youth with TS and stigma, due to the scarcity of studies this relationship is unclear. Although stigma can be associated

to a number of variables such as weight, height, economic status or other conditions (e.g. people with learning disabilities [64]) it may be more important to this specific population since misconceptions about the intentionality and lack of knowledge of TS still lie at the centre of the problem. Clearly, the impact of stigma on well-being of people with TS is such that effective anti-stigma interventions need to be developed which promote the knowledge about the particular syndrome of TS. Issues of stigma also need to be included as an integral part of a comprehensive treatment of individuals with TS in order to promote better quality of life.

Conflict of interest;

The authors declare no conflict of interest.

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