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ADOLESCENTS' PERCEPTIONS OF THEIR PEERS WITH TOURETTE'S SYNDROME: DOES A BRIEF ANTI-STIGMA INTERVENTION HELP?

A THESIS SUBMITTED TO
THE UNIVERSITY OF KENT
IN THE SUBJECT OF APPLIED PSYCHOLOGY
FOR THE DEGREE OF
DOCTOR OF PHILOSOPHY

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Abstract

Background and aim: Tourette's syndrome is a neurodevelopmental condition characterised by tics. It is a condition that has been stigmatised by the public. School age children and especially adolescents often experience rejection from their typically developing peers due to their symptomology which could hinder school inclusion and social participation. Therefore, there is a need for implementing interventions to target typically developing peers' inaccurate knowledge about TS, and their negative attitudes. There is also a paucity of studies that explore the motivation of peers who stigmatise against people with TS. Understanding stigma and its underlying causes, however, is vital to develop effective interventions that can minimise and help to eventually eradicate stigma.

The aim of the thesis is therefore three-fold: firstly, to understand how individuals with TS were perceived by typically developing adolescents. Secondly, based on these attitudes, to develop a tailor-made anti-stigma intervention; and thirdly to evaluate the effectiveness of the developed intervention within a secondary school classroom setting.

Method: A sequential mixed-method design was used. Qualitative data were collected to capture tic-free adolescents' motives, and reasoning involved in the exclusion of individuals with TS. These findings were then used to design a tailor- made intervention and research measures. A randomised cluster control trial with a mixed-model design was used to determine the impact of the developed intervention on tic-free adolescents' awareness of Tourette's syndrome, attitudes, and behaviours towards individuals with TS. Data were collected at three time points: pre-intervention, post-intervention and 9 weeks after its implementation. This is the first intervention that was specifically designed for adolescents and with an assessment at a follow-up stage.

Results: The qualitative study indicated that adolescents understanding about the condition was construed from misconceptions, unfamiliarity and unanswered questions. Furthermore, people with TS were either perceived as being deprived of agency and strength or as deserving pity, and in need of support. These findings helped design the intervention which was implanted on Year Ten students in a school in the South East. Upon completion of the intervention, positive changes were recorded relating to students' awareness, attitudes and behavioural intentions. Tic-free adolescents were more willing to embrace diversity within the classroom and rejected previously held notions of ableism. However, only changes in awareness were maintained at the 9-weeks follow-up.

Conclusion: Tailor-made interventions can contribute substantially to preventing consolidation of unfavourable attitudes towards individuals with TS. Future research should however place emphasis on how these positive outcomes could be maintained over time. Implications of these findings for school curricula and mental health policy, as well as suggestions for future research, are discussed.

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To my husband, Dr Fernando Otero, there truly are no words to express the level of gratitude that I feel towards you. Thank you for all your love and encouragement – it has meant the world to me.

To Phaedra

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Impetus for conducting the study

The idea for conducting the study first stemmed from my professional experience as a special educational teacher in Greece. During 2011-2013 I worked at a special and vocational school in a small town in Southern Greece. I was also hired by the government to home school a 13 year old boy who had Tourette’s syndrome (TS). Shortly after I started my job I realised that the reason he had been excluded from school and subsequently socially isolated was not so much associated with his behaviour but with the behaviour of his peers and the wider local community. Indeed, when he attended a mainstream school he had systematically endured teasing and bullying from his peers. The head teacher of the mainstream school also believed that his presence in the classroom and his tics distracted his classmates. Furthermore, his exclusion had been strongly advocated by the parents of his peers who maintained that the boy’s tics would inevitably interfere with the educational attainments of his classmates. It appeared then, that in order to serve the wider good of the school, the “problematic” student should be excluded. The school never considered making reasonable adjustments to accommodate the needs of the individual with TS but believed that the boy should adjust to the stringent criteria of the school. More importantly however, bullying and teasing appeared to be considered reasonable responses towards a person that was viewed as deviant. blaming the victim was part of the school ethos. There also seemed to be a culture of ableism that was supported by the wider community. Thus, there was a lack of understanding about TS and the implications it had on the individual. Most importantly, in line with the medical model it was believed that the boy should be “fixed” in order to be accepted by his peers who were seen as superior.

The alternative, viewing Tourette’s syndrome through the social model perspective, was never considered by the school. The social model suggests that the impairment is not found within the individual but in the systemic barriers, and negative attitudes society possess. In order to break these barriers interventions must be implemented that inform the wider society about the condition and therefore help ameliorate misconceptions, negative attitudes and stigmatisation. Since in my perspective the biggest obstacles were imposed by the peers of the individual with TS, it was important that they be informed about the condition and appropriate psychoeducational interventions be implemented in the school.

However, looking into the wider literature it was made clear that Tourette’s had only been approached through the medicalised perspective and that there was a paucity of interventions to ameliorate stigmatisation in relation to TS. The need to create appropriate empirically based interventions that would be addressed towards typically developing adolescents was imperative taking into account how stigmatisation can deteriorate one’s quality of life.

Thesis Overview

The idea for conducting the study first stemmed from my professional experience as a special educational teacher in Greece. During 2011-2013 I worked at a special and vocational school in a small town in Southern Greece. I was also hired by the government to home school a 13 year old boy who had Tourette's syndrome. Shortly after I was hired I realised that the reason that he had been excluded from school and socially isolated was not so much associated with his behaviour but with the behaviour of his peers. Indeed, when he attended a mainstream school he had systematically endured teasing and bullying from his peers.

Chapter 1 of the thesis reports on the current and past definitions on Tourette's syndrome (TS) as well as its characteristics and clinical course. Prevalence rates, past and present aetiological theories are discussed in depth. This chapter exemplifies the complexity of the condition, gives a context to the study, but also informs the anti-stigma intervention by providing accurate facts about a condition that has been mostly misunderstood by the public.

Chapter 2 is a systematic review that examined the literature in relation to stigma and youths with TS. The need for anti-stigma interventions to be based on how stigma is experienced by a specific population and on how that specific population is perceived by their peers, necessitated this literature review.

Chapter 3 is a systematic review that assesses and evaluates the effectiveness of school-based interventions that aim to reduce the stigma in behaviourally defined conditions. Such a review allowed an evaluation of the merits and limitations of previous interventions and in doing so, guide decisions about the feasibility, duration, intensity, and other essential components of this new anti-stigma intervention.

Chapter 4 of this thesis described the exploratory sequential mixed-method design used for this study. The scarce and insufficient findings of chapter 2 indicated the need for a qualitative study that examines typically developing adolescents' motives and reasoning for excluding individuals with TS.

Therefore, **Chapter 5** is an exploratory study that aimed to provide overarching theoretical framework for understanding the process by which individuals with TS are perceived by their typically developing peers. Free text writing and focus groups were used to elicit the views of twenty-two Year Ten students from a secondary school in South East England and grounded theory was used to develop an analytical framework. The studies limitations and implications for future research are also discussed.

The findings from Chapter 3 as well as the qualitative study from chapter 5 guided the development of the intervention which is presented in depth in **Chapter 6**. **Chapter 7** presents a cluster randomised control trial investigating the efficiency of the intervention in improving adolescents' knowledge of Tourette's syndrome, attitudes and behavioural intentions towards their peers with TS. Fifty-Nine Year Ten students participated in the study that assessed the immediate effects of the intervention, as well as 9 weeks after the intervention had been implemented. Limitations in relation to the study are discussed as well implications for future research, practice and policy. Finally, an overview of the studies is provided in **Chapter 8**.

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Introduction: Definitions and Characteristics of Tourette's Syndrome

In this chapter, a comprehensive overview of Tourette's syndrome (TS) is presented including the current definition of Tourette's, the characteristics and clinical course of the condition, the prevalence rates, and various aetiological theories. Comorbidities and the contemporary treatments are then discussed. The objective is to give a background and context to this study, and to illustrate the complexity of the condition. The information gathered in this chapter aim to inform the educational part of the anti-stigma intervention.

1.1. Tourette's syndrome: definition

Tourette's syndrome (TS) is a childhood onset neurodevelopmental condition, characterised by tics. It is at the severe end of the spectrum of tic conditions, with provisional tics being at the mildest end of the spectrum. According to the Fifth Edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-V) for a diagnosis to be assigned, multiple motor and at least one phonic/vocal tic must be present for a period of more than a year, during which time the severity of the tics fluctuate (American Psychiatric Association, 2013). Their manifestation however must not necessarily occur simultaneously. Another prerequisite for a diagnosis is that the onset of the tics occurs before the age of 18.

The diagnostic criteria for Tourette's have altered according to the changing editions of DSM and ICD (International Classification of Diseases) (World Health Organization, 1992). Characteristically, the criterion which suggests that the presence of the tics must impair everyday-life was omitted in the revised version of DSM IV-RT (American Psychiatric Association, 2000), broadening the population that can be embedded within the category. Moreover, the criterion of DSM IV-RT by which there should never be more than a three months tic- free interval, was excluded in the latest edition of DSM-V and ICD 10, since such a duration is arbitrary and not based on any clinical data (Walkup, Ferrão, Leckman, Stein, & Singer, 2010). Also, the age of onset has been subject to alteration; the upper age limit for developing the symptomology being moved from 15 to 21 and 18 years of age. Furthermore, although it is highlighted in ICD- 10 (World Health Organisation, 1992) that the symptoms initially appear during childhood or adolescence, there are no specific criteria which exclude adult onset tic disorder from being characterised as TS. Another apparent shortfall of the current diagnostic guidelines in both DSMV and ICD 10 is the requirement of multiple motor tics and at least one vocal tic. Since it can be argued that the distinction between vocal and motor tics is arbitrary, due to the fact that phonic tics are merely a result of a muscle contraction of the oropharynx or diaphragm (Leckman, Bloch, Sukhodolsky, Scahill, & King, 2013), it is debatable how scientifically valid this criterion is. It is further unclear whether an individual who exhibits one motor tic and several vocal tics, or one motor and one vocal tic, for example, would be assigned the diagnosis. It has been argued that these issues need to be addressed in the forthcoming revised edition ICD 11 (Woods & Thomsen, 2014).

1.2. Tics- definition and distinct phenomenology

Tics have been defined as abrupt, sudden non-rhythmic movements or phonations that involve distinct muscle groups (Cohen, Leckman & Bloch, 2013). They can mimic nearly all possible human movements but have no logical reasoning that underlies their occurrence. Furthermore, they appear in a different intensity and frequency than normal motions. Although the term “stereotypical” has been excluded in the definition of tics from DSM-V in order to avoid confusion with the movement characterising autism and schizophrenia (Roessner, Hoekstra, & Rothenberger, 2011), tics do have certain patterned sequence to them in the sense that they will be performed in the same manner every time they occur.

Tics have been classified into simple and complex. Simple motor tics are sudden, abrupt and brief in duration. Characteristic examples include excessive eye-blinking, head-jerking, facial twitching and shoulder shrugging while sniffing, snorting, throat clearing and yelping can be embedded in the category of simple vocal tics (Fahn, 2005). Complex motor tics involve a group of muscles and their duration is longer. They may appear purposeful movements, but they serve no function. Due to this, they bear resemblance to compulsive behaviour. However, the underlying reason for which they are performed does not appear to be associated with thoughts of catastrophe or anxiety and distress (Worbe et al., 2010). Typical examples of complex tics are brushing hair back, smelling objects, skipping and hopping, obscene gestures with hands or tongue (copropraxia) and self-injurious acts (Jankovic, 1997). Complex vocal tics can also be mistakenly perceived as purposeful utterances. Thus, in this category, words and phrases are verbalised. Echo phenomena, such as echolalia (repeating the words of others) and paralilia (repeating one’s 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 (Eddy & Cavanna, 2013).

Recent research suggests that the frequency and severity of the tics are not necessarily directly linked to an individual's functioning. On the contrary, it is the complexity of tics, their duration and their intensity that may affect the social and adaptive functioning of an individual. Thus, complex tics such as coprolalia and NOSIS may be more salient and socially unacceptable (C. M. Eddy & Cavanna, 2013; Specht et al., 2011).

1.3. Characteristics

Although the manifestation of the condition varies in individuals, distinct features characterise the phenomenology of the tics that are encountered in Tourette's syndrome and these elements differentiate them from other movement disorders. Tics occur in episodes, they are not always present but manifest in a paroxysmal pattern and their anatomic location varies through the course of the condition. Moreover, the severity of the symptoms, their frequency and duration change during the course of the day and they exhibit variability through time (Leckman et al., 1998). Most importantly, they are presided by a premonitory urge and considered suppressible and suggestible. They are also dependent on environmental contingencies.

1.3.1. Premonitory urges

Individuals with Tourette's syndrome report a dysphonic sensation allocated in a specific anatomic region before a tic occurs that can be temporarily allayed by the performance of a tic. These sensory antecedents that have been referred to in the literature as premonitory urges have been characterised as the core element of TS have been regarded as more disturbing than the tics themselves (Cohen & Leckman, 1992). The conviction that the performance of a tic will temporarily relieve the distressing sensation may be an underlying reason for the manifestation of the tics and for the reinforcement of the negative cycle (Piacentini et al., 2006). This idea which came to academic attention through case reports (Bliss, Cohen, & Freedman, 1980; Kane, 1994) has been verified by a number of studies that suggest that 79 to 92% of individuals with TS report this inner aversive feeling which needs to be satiated (Cohen & Leckman, 1992; Kwak, Dat Vuong, & Jankovic, 2003). In light of the aforementioned literature tics cannot merely be described as involuntary motor discharges. Thus, the vast majority of the individuals with TS report that the tics are a voluntary response to an irresistible urge. They have therefore been characterised as 'involuntary', or semi-voluntary, thus movements between the boundaries of voluntary and involuntary. This characteristic differentiates motor tics from other hyperactive movement disorders (Leckman, Walker, & Cohen, 1993). As the study of Lang (1991) has demonstrated in comparison to the high percentage of individuals with Tourette's who reported that their tics were a voluntary response to an urge, only eight of a hundred and ten individuals with other hyperkinetic tic conditions asserted that their tics can be considered as voluntary acts.

It must be noted however that the aforementioned studies have solely focused on clinically referred cases of TS rather than on population-based samples. It should therefore be ascertained if this sensation accompanies the population of TS which do not meet the criterion of distress or impairment in everyday life. Since the definition of TS has drawn away from cases that only require clinical attention, it is necessary to verify the premonitory urge findings on the population whose symptom's severity have not compelled them seek clinical attention.

1.3.2. Suppresibility

Another salient characteristic of the tics encountered in Tourette’s syndrome is their suppressibility. Case studies, anecdotal self-reports, and case design studies have demonstrated that individuals with TS have the ability to suppress their tics for a short or prolonged period of time. Koller’s (1989) study indicated in contrast to individuals with other movement disorders, all individuals with TS managed to suppress their tics for a period of approximately 2.5 minutes. Moreover, in contrast to the commonly held belief that the suppression of tics will result in a rebound that exceeds the average frequency, a number of more recent single case designs have demonstrated that after a period of robust tic suppression, tic frequency returns to pre-suppression level (Meidinger et al., 2005; Specht et al., 2013). It should be noted however, that the majority of the aforementioned studies (except Meidinger et al. 2005) used small samples of less than 13 participants each, monitored the post-suppression period for a limited time and used highly artificial settings. Thus, the reaction of people with TS after the tic suppression within a secluded room in which they were isolated from their accustomed environment and from the reaction of their peers has low ecological validity and it is questionable if these findings could be replicated in naturalistic settings.

The suppression of tics however requires attention resources and energy from an individual. The rigorous single case study of Conelea and Woods (2008) demonstrated that during a period of tic suppression the accuracy on tasks was reduced in comparison to a ‘free to tic’ period. Thus, the attention that is devoted to suppressing the tics impacts the performance of the individual on a task.

1.3.3. Dependant on contextual factors

Another distinctive characteristic of the tics encountered in TS is that they are sensitive to a number of environmental contingencies. There is a large volume of published self-report studies describing the role of internal and external contextual factors such as stress, and their effects on the frequency and intensity of the tics (Robertson, Banerjee, Eapen, & Fox-Hiley, 2002; Silva, Munoz, Barickman, & Friedhoff, 1995). These assertions were contradicted by the prospective longitudinal study of Hoekstra (2004) in which no correlation was observed between small anxiety-provoking events of an individual's life and the exacerbation of tics. However, the study relies solely on the self-rating of tic severity. Although self-reports are useful, discrepancies commonly occur between the frequency of the tics that are reported by individuals with TS and the one reported by the practitioners, with the former reporting lesser than the later (Pappert, Goetz, Louis, Blasucci, & Leurgans, 2003).

Moreover, the case design study of Conelea, Woods & Bradt (2011) suggested that stress does not directly affect the frequency of the tics but rather, it negatively impacts the suppression efforts of an individual. More specifically, ten individuals with TS were exposed to four conditions: a baseline, where they were allowed to tic freely, a reinforced tic suppression condition; a reinforced tic suppression condition plus a stress induction task; and a stress induction task alone. It was observed that tic frequency did not differ between the stress task and the baseline condition. However, a reduced frequency was observed during the tic suppression compared to the suppression condition plus a stress task. The main limitation of this study, however was that the stress induction task consisted of a number of timed maths tests that were not only stressful but also require an individual's attention. It is therefore arbitrary to presume that the exacerbation of tics is solely a result of a stress demanding task and not of the attention required for the completion of the test or an outcome of the combination of both.

Furthermore, specific environmental incentives have been perceived as factors that exacerbate tics. Indicatively, it has been demonstrated that tic related conversations (Woods, Watson, Wolfe, Twohig, & Friman, 2001) and paying attention to one's own tics (Brandt, Lynn, Obst, Brass, & Münchau, 2015; Woods & Himle, 2004) escalate tic frequency, while contingent rewards may help the remission of tics (Woods & Himle, 2004).

1.4. Clinical course of Tourette's syndrome

Tourette syndrome is a chronic condition. There is a large and growing body of literature however that indicates that the severity of the tics diminishes in frequency and severity with age (Bloch & Leckman, 2009; Leckman et al., 1998). Leckman et al's (1998) study on 36 individuals with TS suggests that the mean age of tic onset is 5.6 years of age. These findings appear to be consistent with those of Freeman et al's (2000) international study which showed that the onset of symptoms for 41% of the sample began before the age of 6 and for 93% the symptomology of the syndrome emerged prior to the age of 10. Lately Burd's (2005) study which is based on the 3,500 subjects of the Freeeman report (1,235 individuals with TS in total) suggests that although the average age of symptoms onset is 6.36 years the mean age of diagnosis is 13.43 years, indicating that a correct diagnosis may be delayed by many years. Typically, the first tics that occur are simple movements which progress with time to more complex ones. Facial tics and tics involving the head and the shoulders precede the ones occurring at the lower trunk. Moreover, motor tics occur approximately one or two years before the manifestation of the vocal tics (Leckman et al. 1998).

The severity of the tics reaches their peak in the majority of cases in early adolescence. According to the prospective longitudinal study of Bloch et al., (2006) individuals experienced the most distressful symptomology of TS between the ages of 10 and 11. This period is followed by attenuation in tic severity and frequency. The longitudinal studies indicate a significant remission in puberty and adulthood. In one half to two thirds of the cases, the severity of the tics is reduced during puberty and only approximately one quarter of the individuals continue to experience moderate or mild tics through adulthood (Bloch et al., 2006).

However, the data concerning the remission of the tics during puberty and adulthood should be interpreted with caution. The longitudinal studies that have been conducted to date are not based on clinical diagnoses and there may be evaluation bias implications. Thus, the clinical evaluation on the study of Leckman et al (1998) relied solely on participants and parent interviews and the study of Bloch et al. (2006) used information provided by the parents. However, it has been pointed out that self-assessment may not always be precise and accurate. In the longitudinal study of Pappert et al. (2003) it was clearly demonstrated by using recordings of the interviews that 50% of the participants who declared that they were tic-free still had tics. Furthermore, it was demonstrated that in contrast to their subjective reports, the videos indicated 90% of the participants with TS still exhibited tics during adulthood. The same could apply to the parents of the individuals with TS who habituate to the tis. It has also been pointed out that adults manage to sustain, suppress and modify their tics in a more successful manner and for a longer period of time than children and young adolescents (Specht et al., 2011). It is therefore of pivotal significance to determine to what extent the remission of tics is a result of extensive suppression and habituation to the effort in sustaining the tics.

1.5. Prevalence

The occurrence of TS was thought to be a rarity until the late 1980's. Indicatively, the study of Lucas et al (1982) – which examined the prevalence of TS during a twelve-year period in Rochester, Minnesota – estimated, through cases of individuals seeking medical attention, that the annual number of TS cases in the U.S.A would be approximately 1000.

A number of systematic reviews have been conducted recently to estimate the prevalence of TS amongst the school age population. A rigorous meta-analysis by Knight et al. (2012), which was based on 13 studies that have focused on the general population aged mainly between the ages of six to fifteen, indicated a prevalence of 0.77% amongst school age children. This finding is consistent with the recent meta-analysis conducted by Scharf (2015), which was based on both clinically referred cases and population-based samples. A male preponderance over female (4-4.1:1) was also pointed out.

The estimate of prevalence between various community studies are inconsistent, with some ratings indicating up to 5.26% (Cubo et al., 2011) and others as low as 0.5 per 1000 (Apter et al., 1993). This is due to the fact that different studies use different definitions and classifications of TS, utilise different assessment methods and vary in the age population that is being assessed. Moreover, the demographic characteristic of the population that is being used defines the results. The clinical characteristics of TS and a lack of awareness about the syndrome further complicate the research.

An important compounding issue is the fact that diagnostic criteria relating to TS have changed over time (as stated in the Definition section above). The impairment criterion, which suggests that for a diagnosis to be asserted, the tics must impair the person's life, was first included in DSM-III (American Psychiatric Association, 1987), and then removed in DSM IV-TR and DSM V, yetit is still included in the Chinese diagnostic criteria. Since impairment is a notion subjectively and culturally defined, this criterion, which was used in most of the recent studies, may present deflated estimates of the syndrome (Robertson, Eapen, & Cavanna, 2009). The two-stage study of Cubo et al. (2011) estimated that the number of children that fit the criteria for TS with the impairment were 12, while the identified cases without the impairment were 27. The study of (Kurlan et al., 2001) however, which omitted the impairment criterion, estimated that TS is encountered in 3.8% of the population. Although the high frequency is mainly due to the biased sample, the impairment criterion may be an additional parameter that defined the results. Therefore, differences may be observed in future epidemiological studies of TS that use DSM- V to define the criteria.

Studies conducted hitherto have also used a variety of different assessment methods, which consequently influence the estimate of the research. On the one hand, studies that use self- evaluation or are based on individuals who are seeking medical attention may deflate the estimates of the syndrome. This is due to the fact that some individuals are not aware of their tics. The study of Kurlan et al. (1987) indicated that in a kindergarten population of 159 children, 30% were unaware of their tics. Moreover, many individuals may not seek medical support because of their lack of awareness about the syndrome or may be erroneously diagnosed because numerous practitioners still hold the notion that TS is linked to coprolalia (Robertson, 2008). On the other hand, using direct observation without a follow-up interview and multiple sessions may lead to an overestimation of the prevalence rate (Tanner, 2005).

Epidemiological studies, which rely on paediatric populations, are bound to produce higher prevalence rates than the ones that use adolescent or adult samples, since the symptoms of the syndrome manifest at the age of 7 deteriorate during early adolescence and subside during puberty. Thus, the study of Apter et al. (1993), which examined the frequency of TS amongst 18,364 male and 9,673 female Israel adolescences aged from 16 to 17, may represent the remitting course of TS rather than some form of ethnic differentiation. As has been mentioned, a pivotal characteristic of the tics is their remission and exacerbation through time and the individual’s ability to suppress them. It is therefore possible that many cases are overlooked if the diagnosis is based on a single observation (Tanner, 2005).

The characteristics and phenotype of TS are parallel across countries and cultures, as the study of Freeman et al. (2000) indicates. Thus, the data from 3500 individuals with TS, gathered by practitioners from 65 sites in 22 countries, suggests that a variation of symptomatology does not exist among different nations. However, there appears to be a differentiation of prevalence rates among different ethnicities. Characteristically, studies to date indicate that TS has a lower prevalence among Asian and South African populations (Robertson, 2008). However, the low figures in the former population could be a result of the usage of different criteria for TS diagnosis. According to the Chinese Mental Disorder Classification and Diagnostic Criteria (CCMD: 3) in order for a child to be diagnosed with TS, she/he must display more than two tics at the same period, a suppressibility over the tics and must fit the impairment criterion. These rigorous criteria may be accountable for the deflated estimates among Chinese population (Robertson, 2008). The sole study that examines the epidemiology of TS among South African population (Pelzer, 1994) exhibits low prevalence rates. These findings have been attributed to a lack of awareness as well as environmental factors (Robertson, 2008).

1.6. Aetiology

1.6.1. Past theories

Since the late 19th century and the publication of Gilles de la Tourette concerning the “maladie des tics” (De La Tourette, Georges Gilles, 1885) literature abounds with contradictory publications denying the entity of the “disease”. The aetiology of the tics has also been controversial. Through the historical review of the studies four explicit but broad categories can be constructed which attribute the manifestation of tics to specific causes or link the symptomology of TS to other various mental or organic diseases.

1.6.1.1. The manifestation of tics as part of the hysteria spectrum

Immediately after Gilles de Tourette’s initial publication the distinct entity of the condition was refuted. George Guinon, firmly asserted that the symptomology described by Gilles Tourette’s was nothing more than a subset of hysteria and therefore should be treated with hypnotism (Guinon, 1886). Sigmund Freud also ascertained that tics and vocalisations were part of dramatic hysteria which was provoked by repressed childhood trauma (Kushner, 1999).

1.6.1.2. The expression of tics as a form of psychosexual disturbance: the psychoanalysis approach

The first theory that tied the expression of tics to sexual disturbance emerged in the late 1890’s. The symptomology of a fifteen-year-old boy, whose tics resembled to those described by Gilles de Tourette (1897), was regarded as a result of masturbation. Therefore, a circumcision was performed and the physician ordered for the child’s arms to be put in splints at night (Wilson, 1897).

It was not however until the 1920’s and the Freudian era of psychoanalysis that the concept of tics as an expression of repressed sexuality gained wider acceptance. Sandor Ferenczi (1921), ascertained that all tics bear an unconscious sexual component. They were viewed as an expression of repressed childhood sexuality and narcissism as well as a symbolic act of masturbation. Coprolalia was therefore conceived as an expression of the same erotic emotion. The theory of repressed sexuality was further advocated and built upon by Margaret Mahler (Mahler & Gross, 1945). Her views on tics would dominate the medical community until the 1960's because of the large number of individuals she treated.

1.6.1.3. Tics as an expression of habit

The influential study of Meige and Feidel at the beginning of the twentieth century maintained that tics were infantile bad habits (Meige & Feindel, 1902). Based on observations of their patients these scholars maintained that tics were voluntary acts that turned into automatic habits. They also argued, that it was the weakness of the patient's character and the little or lack of effort to inhibit the tics that permitted their manifestation; it was therefore the infantile nature of the individuals' determination that prohibited them from eradicating the tics.

1.6.1.4. Manifestation of tics as part of other conditions

It was first argued immediately after Tourette's essay that the abrupt motor movements and vocalisations that were described were merely components of chorea which was caused by rheumatic fever. In other words, it was asserted that although these two conditions manifested in a different manner, they had the same underlying cause.

By the 1920's the occurrence of tics was viewed by many physicians as a sequel to previous encephalitis infection and characterised as a "post -encephalitis neuropsychiatric tic disorder" (Straus, 1927). In response to this theory and the assentation that with the removal of the organ that had been infected the tics would be eradicated (Billings, 1912), a surgical era for the treatment of tics arose between the 1920 and 1930's. Characteristically, tonsillectomy and the removal of sinuses and adenoids were common practices (Kushner, 1999).

The current understanding of the aetiology of TS began after haloperidol, a neuroleptic antipsychotic drug that was mostly used to treat schizophrenia, was found to be effective in ameliorating tics (Stevens & Blachly, 1966). Although the drug's effectiveness proved the neurobiological nature of TS, the precise aetiology is still unknown.

1.6.2. Current understanding of aetiology

Aetiological factors behind TS are largely unknown, but it is quite complex and linked to genetics, development, neurobiology, and neurochemistry.

1.6.2.1. Genetics

Despite the heterogeneous phenotype of the condition studies suggest a genetic contribution to TS. However, no single genomic locus has been identified. Based on the therapeutic response to neuroleptics, all genes that encode neurotransmitters have been viewed as candidates for TS aetiology (Paschou, 2013). Although the exact hereditary phenotype is poorly understood, the familial pattern of TS had been observed by Gilles de la Tourette himself and is well documented through family studies. The study of Price et al. (1985) that evaluated 43 twin pairs ascertained concordance to be between 50% to 77% in monozygotic pairs whereas only 10% to 23% in dizygotic pairs. Furthermore, the study of Hyde et al., (1992) suggests that 94% of the 16 pairs of monozygotic twins that were examined exhibited concordant for some form of a tic condition. The vulnerability of biological relatives has also been demonstrated by the study of Pauls et al. (1991) that examined 338 first degree relatives of 86 TS subjects and ascertained at least a 10-fold increase risk for exhibiting TS in comparison to the control group.

1.6.2.2. Developmental factors

Prenatal and perinatal adversities have been implicated in predisposing an individual to TS. Twin studies (Hyde, Aaronson, Randolph, Rickler, & Weinberger, 1992; Pauls, 1992; Price, Kidd, Cohen, Pauls, & Leckman, 1985) demonstrate that not all monozygotic twin pairs were concordant for TS, thus providing evidence that non-genetic factors mediate the expression of TS. The study of (Burd, Severud, Klug, & Kerbeshian, 1999) that compared the birth hospital records of 92 individuals with the one of 460 controls suggest that older paternal age and early pregnancy complications were associated to TS. Furthermore, Motlagh et al., (2010) (with a cohort of 180 women) implicated heavy maternal smoking and maternal stress during pregnancy to the emergence of TS. The latter finding is corroborated by Leckman et al. (1999) who associates TS to severe nausea and/or vomiting during the first trimester of the pregnancy. However, this study is confounded by a lack of a control group. A significant association is also observed between the TS cohort and a number of prior induced pregnancy terminations (Klug, Burd, Kerbeshian, Benz, & Martsolf, 2003). Yet again, the current literature is confounded by small numbers of participants and a lack of systematic study that examines all of aforementioned risk factors in TS cohort.

The literature also abounds in the association of TS to infections and most specifically to the Paediatric Autoimmune Neuropsychiatric Disorder (PANDAS). Group A beta-haemolytic streptococcal (GAS) (Martino, Defazio, & Giovannoni, 2009) has also been linked to TS by a large proportion of the literature, but the exact nature of the relationship is poorly understood (Robertson, 2011).

1.6.2.3. Neurobiology

A third factor that may contribute to the expression of TS is neurobiology. Tourette’s syndrome has been linked to physiological abnormalities within the Cortical- Striatal-Thalamic-Cortical circuits (CSTC). However, the exact location of the lesion within these circuits is unidentified. A number of radiographic studies suggest that cortical abnormalities are associated with the pathogenesis of TS. It has also been associated to basal ganglia dysfunction (Peterson et al., 2003).

1.6.2.4. Neurochemical imbalances

Neurochemical imbalances have also been associated to the expression of TS. The therapeutic response of tics to blockage of the dopamine receptors suggest that abnormal dopamine neurotransmission is implicated in TS pathology. This is corroborated by post-mortem and neuroimaging studies (Peterson et al., 2003; Singer & Minzer, 2003). Furthermore a number of post-mortem (Anderson et al., 1992) studies and blood analysis (Leckman et al., 1995) have implicated an association between low levels of serotonin with the occurrence of tics.

1.7. Comorbidities

Tourette’s syndrome has been linked to a wide but specific range of neuropsychiatric disorders, most common of which being Attention Deficit/ Hyperactive Disorder (ADHD) and Obsessive-Compulsive Disorder (OCD). A large and growing amount of studies have demonstrated that TS as an isolated condition is the exception, rather than the rule (Freeman et al., 2000; Khalifa, Dalan, & Rydell, 2010). The study of Freeman that relies on the data from 3500 individuals who were clinically ascertained, suggests that merely 11 to 12% of individuals with TS experience solely TS. In congruence with these findings, community based samples suggest that up to 80% of individuals with TS are affected by a co-occurring condition (Kurlan et al., 2002; Scahill et al., 2013). However, the recent epidemiological study of Scharf (2012) has questioned this high frequency of comorbidity in community samples. Through longitudinal data from 6768 children gathered by mother-completed questioners, it was estimated that approximately 70% of the participants did not endure to have other co-morbid psychiatric conditions. However, the study relied solely on maternal screening questionnaires and there was a lack of clinical assessments. Furthermore, the researcher only examines the most profound disorders that have been associated to TS, (i.e. ADHD and OCD), and omitted to take into account other conditions such as Self-Injurious Behaviour (SIB) anxiety disorders etc. Thus, the study failed to give an accurate estimate and image of the holistic spectrum of the comorbidities encountered in the TS population (Scharf, Miller, Mathews, & Ben-Shlomo, 2012).

The high rates of comorbidity suggest that a multifaceted and complex clinical picture exists. Thus, despite the fact that TS is still been viewed as a unitary condition by the World Health Organisation and the American Psychiatric Association, a considerable number of studies have identified a behavioural spectrum in TS. Cluster analysis, principal component factor analysis and latent class analysis have all been utilised to distinguish various phenotypes in terms of both tics and psychopathology with homogeneous characteristics (Cavanna & Rickards, 2013; Eapen & Robertson, 2015). Individuals with TS alone, without the existence of any other co-morbid conditions, are considered to be unencumbered from major behavioural and cognitive difficulties. On the contrary, induvial with TS, ADHD and or and OCD, may be challenged with psychosocial functioning and cognitive impairments.

1.7.1. Attention Deficit / Hyperactive Disorder (ADHD)

ADHD is characterised by inattention and impulsivity which causes impairments in social, academic or occupational functioning (American Psychiatric Association, 2013). The symptoms must occur before the age of 7 years old and therefore the diagnosis commonly precedes the emergence of the tics by two or three years. Epidemiological studies have indicated the occurrence of ADHD exhibited in 50 to 80% of individuals with TS (Roessner et al., 2007; Zhu, Leung, Liu, Zhou, & Su, 2006). These figures however should be considered with caution because of the overlap between the symptomology of the two conditions. It has been argued that inattention, the core element of ADHD, may be observed in individuals with TS due to the fact that strenuous effort is put into suppressing the tics. Therefore, the disentanglement between the two conditions may be challenging (Cavanna & Rickards, 2013).

A large and growing body of literature suggests that the co-occurrence of the two conditions predicts a number of severe additive psychophysiological and cognitive effects in comparison to individuals with TS alone (Carter et al., 2000; Sukhodolsky et al., 2003). The international study of Roessner et al. (2007) that examined the clinical course of 5060 youths with TS, suggested that children with TS plus ADHD endure more internalising and externalising behavioural problems in comparison to individuals with TS alone. Furthermore, the presence of ADHD suggests high rates of other comorbid conditions. However, the study is weakened by a lack of age-matched healthy control groups and ADHD groups only which would serve as point of reference. A comparison to the developmental course of children with ADHD alone in such a large sample would provide insight into whether the clinical profile of the children with TS plus AHDH is unique.

The diagnosis of ADHD in individuals with TS further perplexes the developmental course for the individual. In contrast to TS, in which older age is associated to a remitting and a more stable course of symptomology, in ADHD although the hyperactive symptoms decline, the inattention symptoms appear to persist through adulthood (Faraone, Biederman, & Mick, 2006). However there is a paucity of longitudinal studies assessing the clinical course of individuals with TS plus ADHD. The recent study of Rizzo, Gulisano, Calì, & Curatolo, (2012) evaluated the clinical course of 100 people with TS and comorbid conditions ten years after their first clinical assessment. It was observed that the subgroup of TS plus ADHD (n=48) presented a spectrum of various clinical phenotypes on the second assessment. Thus, in the majority of the individuals (65%) all symptomology relating to

ADHD had disappeared and they were diagnosed as having only TS. 35% of them endured TS plus OCD while only 2% experienced the combination of TS plus ADHD plus OCD. This clinical picture is superior to the one encountered in individuals who were initially diagnosed with TS alone; after 10 years, 43% of them developed OCD and 58% carried on with the same phenotype. Conclusively, in the area of co-morbidities, the research suggests better outcomes for people with TS plus ADHD compared to TS alone. These findings are in congruent with the study of Roessner (2007) that suggests that over time general comorbidity is more pronounced in the TS alone group. However, the study of Rizzo et al., (2012), is beset with methodological limitations including a small sample size. Furthermore, taking into account that ADHD is a chronic condition, it may be argued that some participants were initially misdiagnosed as having ADHD because of the overlapping symptomology of the two conditions. Conclusively, further and broader research is warranted.

The high rate of incidence between the two conditions and the unique behavioural and cognitive profile that is encountered in individuals with TS raises the question as to whether the combination of TS and ADHD reflects a separate entity or is merely the aggregation of two independent conditions (Yordanova, Heinrich, Kolev, & Rothenberger, 2006).

1.7.2. Obsessive-Compulsive Disorder (OSD)

Obsessive-Compulsive disorder can be defined as reoccurring and invasive involuntary thoughts that cause distress and discomfort to a person’s life. Although in most cases the individual is aware of the hollowness of their actions, the performance of a repetitive behaviour may briefly relieve the individual from the anxiety caused by the initial obsession. This temporary alleviation however, reinforces the symptomology (American Psychology

Association, 2013). The aforementioned process can be compared to the sensory phenomenon which proceeds the volitional occurrence of a tic. However, the former is characterised by anxiety and a multifaceted cognitive procedure, whereas the latter is not (Cavanna & Richards, 2013).

TS had been associated with compulsions and obsessions since the initial description of the maladies des tics of Gilles de la Tourette. The French neurologist described obsessive thoughts that anguished the individuals (Kushner, 1999). The epidemiological studies examining the rates of OCD in individuals with TS which vary greatly from 11% to 80% (Cavanna, Servo, Monaco, & Robertson, 2009; Robertson, 2000), but prominently exceed the prevalence in the general population (1.9-3.2%) suggesting an intrinsic association between the two conditions. Recent studies have attempted to disentangle the obscure relationship between the two conditions, suggesting that the combination of the two syndromes form an OCD subtype with specific characteristics and familial pattern.

It has been argued that "pure" obsessive-compulsive disorder should be distinguished from tic associated OCD, the phenomenology of which is linked to male preponderance, younger age of onset and the presence of antecedent sensory phenomenon. The obsessive thoughts experienced by individuals with TS plus OCD form their own subcategory and differ from the ones encountered in individuals with pure OCD, with the former being prominently related to violent, inappropriately sexual and symmetrical thoughts (Worbe et al., 2010). Moreover, the obsessive thoughts of germs, cleaning and contamination, which is characteristic in individuals with OCD, are rarely observed in individuals with the combination of the conditions. It has also been argued this phenotypic subtype is a highly familial condition. A considerable number of studies have delved into the genetic relationship between the two conditions in several families. The comprehensive study of Pauls et al., (1992) which examines the frequency of OCD among first degree relatives suggests that the rates of TS and OCD in families of TS were the same as in families with the combination of the conditions. Furthermore, in the study of Rosario it was observed that relatives of individuals who experience both OCD and TS had much higher rates of OCD in comparison to relatives of individuals who only endured OCD, making the diagnosis of a tic the best predictor for the presence of OCD. It has therefore been argued that TS and OCD are different expressions of the same gene, which may be gender related. The aforementioned inconsistencies between the two groups, OCD and TS plus OCD, are also mirrored in differences between responses to medication. A commonly used treatment for OCD, selective serotonin reuptake inhibitory (SSRI) did not have effective results on individuals with TS plus OCD (Mc Dougle et al., 1993). It has therefore been suggested that OCD is a heterogeneous condition and a tic related subtype exists. Despite the recommendation that it be entered as a separate subtype in the DSM V, it has not.

1.8. Contemporary treatment

The multidimensional nature of Tourette syndrome complicates and perplexes the treatment of the condition and the analysis of the efficiency of the used treatments. Because of the organic nature of TS the first line of treatment has been a pharmacological approach although a number of side effects have been reported. Behavioural therapy is currently a commonly used method in reducing tic severity with benefits being well documented. Furthermore, surgical interventions have been utilised when other forms of therapeutic modalities seam to fail and the severity of the incapacitating tics are not eradicated. In the following review the most prominently used methods of each category will be revised.

1.8.1. Pharmacotherapy

Pharmacological treatment has been used for the reduction of tics in people with moderate to severe tics. It should be emphasised that this form of treatment does not eliminate the tics but typically 25-50% reduction of symptomology is observed (Roessner & Rothenberger, 2013). However, they work in a more rapid manner than behavioural therapy.

The novel atypical antipsychotics seem to be detached from the adverse side effects of their ancestors (e.g haloperidol). Currently risperidone is the first choice for the treatment of tics with recent studies suggesting similar efficiency in the reduction of tics as haloperidol and primozide (Huys et al., 2012). Despite the lack of double blinded control trials to demonstrate its efficacy it is favoured by practitioners throughout Europe (Rickards, Cavanna, & Worrall, 2012). Olazapine and apriprazole have also been prescribed to patients with TS with a number of studies presenting encouraging results (Huys et al., 2012).

Alpha adrenergic agonists are commonly used to treat people with Tourette's. In a recent European survey that involved a range of clinicians treating children with Tourette's it was demonstrated that the clonidine was one of the pharmacological treatments of choice (Rickards, Cavanna & Worall, 2012). Yet, there is paucity of double-blind controlled studies comparing the effectiveness of the different psychopharmacological agents and evaluating the tolerability of the different medication. Moreover, the long-term side of pharmacotherapy is unclear (Whittington et al., 2016).

1.8.2. Behavioural therapy

Recently non-pharmacological treatment of tics has been a subject of salient controversy. The most prominent technique is Habit Reversal Training (HRT) which was initially described and suggested for the reduction of tics and other disorders that comprised of repetitive movements by Arzin and Nunn. Although there is evidence to suggest that HRT is effective (Himle, Woods, Conelea, Bauer, & Rice, 2007) it is less often used than pharmacotherapy possibly due to lack of availability or limited financial means. Furthermore, the mechanism behind the efficacy of HRT is poorly understood. It was initially conceptualised as an effective method to strengthen the opposed muscles to the tic motion that were unused and therefore incapable of inhibiting the erroneous movement. Tics were therefore conceptualised merely as learned responses that were perpetuated by reinforcement. The basic argument against utilising and disseminating this method to eliminate tics is the reconceptualization of tics as a psychological condition (Scahill et al., 2013). Finally, a recent systematic review assessing the effectiveness of different treatments for Tourette’s suggests that there is no evidence that HRT is effective without the use of medication (Whittington et al., 2016).

1.8.3. Deep Brain Stimulation (DBS)

In Deep Brain Stimulation electrical signals are used to modulate neuronal activity but in contrast to other forms of surgery the outcomes are reversible. It has been accentuated however that this form of therapeutic approach should only be accessible to adult patients who have unsuccessfully received other standard, less-invasive therapies. Since 1999 a number of surgical procedures have been conducted in severely affected, treatment resistant patients (Porta et al., 2016).

The overall results suggest encouraging outcomes in the inhibition of tics as well on the associated conditions. The efficacy of the treatment is documented by the reduction of the tic frequency by at least 24% (Servello, Porta, Sassi, Brambilla, & Robertson, 2008) with some studies revealing a 90% improvement (Vandewalle, van der Linden, Groenewegen, & Caemaert, 1999). However, it should be emphasised that the measurement instrument of the tic severity that is being utilised is not the same in all studies. Thus, a number of studies relied solely on the observations of tics by investigators (Vanderwalle et al. 1999) while others (Diederich, Kalteis, Stamenkovic, Pieri, & Alesch, 2005) used more multidimensional instruments for assessing the severity of the tics, such as the Yale Global Tic Severity Scale (YGTSS) which relies on observation and semi-structured interviews. Therefore, a more uniform approach is warranted for the outcome measures. A sustained tic reduction has also been observed in long term follow up examination (Ackermans et al., 2010).

However, a number of side-effects and a lack of beneficial outcomes have been reported. Fatigue and lethargy has been associated with DBS and visual disturbances have been reported by patients (Ackermans et al., 2011). Servello et al. (2011) reported a number of infectious complications in people with TS that lead to the removal of the stimulator. Furthermore, the more recent study of Duits et al. (2012) describes the case of a twenty-year-old female who underwent thalamic DBS. Beside TS the individuals suffered from pervasive developmental conditions, depression and self-injurious behaviour. After the surgery, she developed severe hypertonia that did not dissolve after the stimulator was switched off. Disturbances in consciousness were observed as well as mutism, impaired swallowing and intolerability in keeping food down. Subsequently she died three years after the operation.

Most reports hitherto are merely case studies, with only two double blind studies has been conducted (Ackermans et al., 2011). Furthermore, the validity of the studies to date is compounded by the small sample size.

1.9. Summary

This chapter has provided detailed information about Tourette’s syndrome and the nature of the condition. Tourette’s syndrome is a multidimensional condition, encountered in approximately 1% of the school aged population. The tics, which are the essential characteristic of the condition are reported to be susceptible to contextual factors and suppressible. This complicates the phenomenology of the condition. Also, the high rates (80–90 %) of comorbidity found in clinic and community-based TS populations, contribute to its heterogeneous presentation.

All of the information gathered for this chapter helped with the development of the anti-stigma intervention used in the current thesis as well as aiding the understanding of the complexity of the condition.

Moving on, in order to create a tailor-made intervention, it was considered to be important to understand how people with TS are perceived by their peers, why negative attitudes might have been formed, and how stigma may be experienced for people with TS. This is the focus of the next chapter.

2

Stigma in Youths with Tourette's Syndrome: A Systematic Review and Synthesis[*]

2.1. Introduction

Beside the hurdles children and adolescents with Tourette's syndrome endure because of the manifestation of the symptomology, narrative testimonies of youths with TS provide extensive accounts of stigmatisation, harassment, bullying and teasing (Mansley, 2003). Although research has applied the concept of stigma in a variety of marginalised groups (e.g. mental illness), no previous systematic reviews that explored the difficulties people with Tourette's experience due to stigma were found. Different conditions are however, subject to distinct types of negative attitudes, necessitating examination of stigma that threat specific conditions individually. It is therefore the aim of the systematic review presented in this chapter to provide an overview of the existing research on social stigma in relation to children and adolescents with TS, self-stigma, and courtesy stigma in family members of youths with TS.

The objective of this chapter is to reveal why and how stigma is enacted on youths with Tourette's syndrome. Understanding why people with TS are stigmatised by their tic-free peers and identifying how stigma is imposed on them is essential for developing an anti-

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stigma intervention and strategies to eliminate discrimination. Indeed, the root of developing an effective intervention lie in examining and understanding public conceptions about TS.

The chapter begins with an introduction to the concept of public stigma and the components that converge in its development. The notions of self-stigma and courtesy-stigma are also explored. The chapter further identifies the features that render Tourette's syndrome a stigmatising condition. The aims and methodology of the systematic review are explored and the results are reported. Limitations and issues that have yet to be addressed are discussed and promising avenues for further study are indicated.

2.2. Conceptualisation of social stigma

The word stigma is derived from the ancient Greek verb στίζω. It originally referred to the creation of a scar upon the bodily flesh by a sharp instrument through the process of pricking, cauterizing or carving (Onions, Friedrichsen, & Burchfield, 1966). According to Herodotus (Hdt 5, 35) it could be associated to a mark of ownership in slaves but could also signify disgrace and subsequently denote the bearers' compulsory segregation from general society (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 a subject of elaboration and conceptual variation in contemporary literature. Goffman (1963) 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" prohibits the stigmatised from being fully accepted by society. Conceptualisations of the term have also emphasised the dynamic, relational and contextual nature of stigma (Link & Phelan, 2001).

Despite the divergent views in both psychological and sociological approaches in identifying stigma as a complex social construct that can exist when cognitive, affective and behavioural components of the ‘stigmatiser’ (the person who is likely to stigmatise) converge (Fiske, 1998). It should be emphasised that these three components may occur in any order. Thus, cognitive processes may be triggered by affective reactions in order to justify an initial emotion and may be followed by a behavioural aversion. Alternatively, cognitive processes may affect emotional reactions which in term could lead to discriminating actions.

2.2.1. Cognitive component of stigma

The cognitive component refers to negative stereotyped knowledge and beliefs. Stereotypes can be conceptualised as sets of cognitive schemas that reflect the assumed attributes and distinctive characteristics that a category of people possess (Hilton & Von Hippel, 1996). The process of stigmatising involves ascribing a set of inaccurate traits to individuals because they are believed to be affiliated with a specific social group. In reality this “virtual social identity” that is imposed on the stigmatised may bear little resemblance to the actual identity of the individual (Goffman, 1963).

Research indicates that individuals that have a thorough understanding about a stigmatised condition are less likely to endorse stigmatisation (Corrigan & Penn, 1999). Though providing information alone is not expected to eliminate stigmatisation, lack of awareness is considered to be an important factor in the inflammation of prejudice (Haghighat, 2001). Most interventions that aim to combat stigmatisation provide factual information in order to demythologise a condition and promote favourable attitudes and behaviours (Corrigan & Penn, 1999).

2.2.2. Affective component of stigma

The affective domain, prejudice, refers to the negative feelings and emotions that the ‘stigmatiser’ engenders as a response to the individual with a “spoiled identity”. These feelings could fluctuate between fear, irritation, discomfort, pity and anger. The affective responses are more primitive and presumably faster than the cognitive ones that require effort. Furthermore, they are more detectable and therefore perceptible by the individual that is being stigmatised (Link, Yang, Phelan, & Collins, 2004).

2.2.3. Behavioural component of stigma

The behavioural dimension refers to the enacted discrimination against the object of stigmatisation. Discrimination can be viewed as biased behaviour which does not solely mean actively harming and disadvantaging the “spoiled” individual but also segregating or coercing them. Childhood and adolescent peer exclusion and bullying could fit into the wider umbrella of discrimination (Spears Brown & Bigler, 2005). According to Olweus’ well-established definition, bullying can be interpreted as an intentional and repetitive act that requires an imbalanced dynamic between the victim and the perpetrator, with the latter establishing dominance and subordination over the former. This relationship is therefore created by using some form of aggression and it is formed in order to extinguish any form of willpower the victim might have to fight back (Olweus, 2011).

There are different categorisations of bullying, all of which fit into the umbrella of discrimination. Traditionally literature has distinguished between the proactive types of aggression and covertly manipulative forms of victimisation (Mynard & Joseph, 2000). The former category involves overt physical and verbal aggression from the bully to the victim. Indicatively pushing, hitting and name calling can be embedded in this category. Indirect bullying on the other hand requires 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 the social status of an individual. In contrast to the overt form of bullying the perpetrator is less obvious and less easily identifiable in this category (Scheithauer, Hayer, Petermann, & Jugert, 2006).

2.3. 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-stigma, 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 (Corrigan, 1999). 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 message (Corrigan & Watson, 2002). In the case of Tourette's, a number of people are energised and empowered by their individuality which they exhibit through the media (see, for example The Undatables[1], Touretteshero[2]).

2.4. Courtesy and affiliate stigma

The concept of courtesy-stigma, which was initially introduced by Goffman (1963), highlights that the individuals with the "spoiled" identity are not the only ones who endure stigmatisation. Stigma may also affect people who are closely associated with a person who has been discredited e.g. immediate family and friends. They are therefore treated as one

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[1] http://www.channel4.com/programmes/the-undateables/profiles/all/ruth
[2] http://www.touretteshero.com/

unit in a social situation. Courtesy stigma is associated with difficulties in interacting with other people, social rejection and disrupted social relations.

This form of stigma is identified in families of children with autism (Mak & Cheung, 2008), ADHD and mental conditions such as schizophrenia (Schulze & Angermeyer, 2003). Furthermore, a considerable amount of literature has documented that family members, in particular caregivers, tend to internalise the devaluing views of others and feel guilt or self-blame in relation to their relative's condition (Muhlbauer, 2002). The self-stigmatisation of family members has been referred to as affiliate stigma (Mak & Cheung, 2008). As a consequence, caregivers tend to conceal their status and isolate themselves.

2.5. Stigmatising features of Tourette's syndrome

It has been pointed out that the name Tourette, which has been bestowed on the syndrome, does not bear the stigmatising and invalidating effects of other terminologies used with other conditions like schizophrenia (Ramirez-Bermudez & Perez-Rincon, 2010). Notwithstanding this, the six-dimensional approach to stigma proposed by Jones et al. (1984) may suggest that the very nature and symptomology of TS could result in an individual with the condition being stigmatised. Thus, tics could be highly visible, perceived as controllable and they distort interaction and communication. They may also cause an unpleasant aesthetic effect on the observer. Finally, it is a syndrome that is misunderstood by the public, and mocked and sensationalised by mass media.

The first dimension of stigma is concealability (Jones, 1984). In the case of Tourette's, tics may be highly visible so that, in contrast to other conditions, individuals with TS cannot hide their condition for a prolonged period of time. Goffman (1963) has pointed out that, in contrast to concealable forms of attributes, the ones that are visible have more negative effects on one’s identity.

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. Thus, in accordance with Weiner’s attribution theory, anger, unfavourable views and punitive reactions may be associated with supposed controllability over the syndrome. On the contrary, sympathy may be the affective reaction towards an individual that is not considered at fault for his/her misfortune (Weiner, 1993). Taking into account that tics violate our prior knowledge and understanding on what could be conceived as deliberate and unintentional as far as gestures, vocalisations and movements are concerned (Buckser, 2008), without adequate knowledge the observer may feel that the people with TS are accountable for their action and lack of willpower. Moreover, the paradoxical capacity of many individuals to consciously exercise some form of control over the tics further exacerbates the stigma formation. Thus, the symptomology of TS may appear as a lack of control and attract angry responses and may justify withholding prosocial behaviour from the stigmatised because of the presumed responsibility in engendering the mark.

The disruptiveness of the tics and the manner in which they hamper, strain and disrupt the interpersonal interaction and communication can be considered one more dimension that inflates the stigmatisation mechanism (Jones, 1984). The loss of control over the individual’s movements and the body gestures may intensify the feeling of discomfort the non-stigmatised may experience during their contact with people with TS. This fear of unexpected behaviour may also alarm the non-stigmatised.

Aesthetics and the negative affective reaction to something that is considered visually unpleasant is another dimension that Jones et al. (1984) has linked to stigma inflammation. The movement of the body, which can be viewed as a means of interaction and has an expressive value, conveys our character to the world. In accordance to this concept, Souriau (1983) identifies that the basic characteristic of the aesthetically attractive movement is the easiness and the effortlessness by which it is performed by the actor. The abrupt, odd and sometimes painful movements of an individual with TS are radically antithetic to what is considered an aesthetic pleasing movement (Davis, Davis, & Dowler, 2004). In other words, the normalised human motion is disrupted by the apparently irrational movements of individuals with TS and the semantic dimension of the motion hold no bearing as it fails to conform to what the individual is thinking.

Tourette's is also a condition that has mostly been misunderstood by the public and mocked by the media. The stereotypical image of a person with unpredictable and unstable behaviour who has the irresistible urge to utter obscenities is further perpetuated by mass media and popular culture through comedic films such as "Wedding Crashers" (Calder-Sprackman, Sutherland, & Doja, 2014; Fat, Sell, Barrowman, & Doja, 2012). In the aforementioned movie TS is attributed to individuals that compulsively swear, but have no other tics. In other words, despite the fact that an unprecedented volume of information is available to the public about TS, accurate information about the syndrome is scarce. In the entertainment industry TS is attributed to a character in a mistaken way and has been misused by comedians, columnists, films and even cartoons by associating Tourette's to individuals that have a tendency to swear frequently (Monaco, Servo, & Cavanna, 2009). Although people with TS do not employ profanities to convey an emotion or emphasise an opinion but use them out of context, TS has been linked to aggressive expression through vulgar language and been referred to as the "cursing disease" (Kushner, 1999).

2.6. Aims and objectives of the systematic review

Despite the growing number of studies that address the social acceptability of individuals with TS, to my knowledge, there have been no systematic reviews that focus on examining the body of literature on stigma in association with children and adolescents with Tourette's syndrome. Through a systematic review we could obtain a complete, exhaustive summary of current literature and therefore provide a comprehensive picture of the available evidence. The objective of the study to 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 were as follows:

- To identify publications that explore the public's stereotypical knowledge about TS, including unfavourable attitudes and discriminating behaviour towards children and adolescents with TS;
- To understand the origins of any negative attitudes and behaviour from the point of view of individuals who exclude and marginalise youths with TS;
- To identify if stigma is associated with age or gender in children and adolescents with TS and to ascertain, if gender and age could be identified as characteristics that could predict the stigmatisers' behaviour towards individuals with TS;
- To provide an overview on how stigma is experienced and managed by youths 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 investigate courtesy and affiliate stigma in family members.

Furthermore, this review aimed to summarise, evaluate and provide a comprehensive overview of the current knowledge and evidence and to indicate future research directions.

2.7. Methods

2.7.1. Research strategy

To identify studies for inclusion in the systematic review three electronic databases were searched: PsycINFO, PubMed, and Web of Science. Search terms focused on two areas: Tourette's and stigma. All terms in the left hand column were combined with all the terms in the right hand column (Table 1, p. 36). Thus, 96 separate searches were conducted.

2.7.2. Inclusion criteria

This systematic review was restricted to empirically based studies that used explanatory, experimental or survey study design and were published in peer review journals after 1994 and before 2015. Using both quantitative and qualitative studies was imposed by the questions raised by this research. Languages of publication were restricted to English, German, and Greek, which correspond to the researcher's language acquisition.

Table 1 - Table of the literature review search terms.

Condition	Stigma
Tourette *	Stigma, self- stigma, courtesy-stigma, knowledge, ignorance, misconception
tic *	peer evaluation, public opinion, stereotypes, social rejection, prejudice, bully*, victim*, discrimination*, teas*, peer problem*

Limits were set on the database to exclude articles that were published before 1994. As the degree of stigma is known to change through time, this date limit was set to ensure that the literature accurately reflects society's conceptualisation of Tourette's syndrome through the past two decades.

Predefined eligibility criteria identified as relevant any study that directly evaluated the public's awareness of Tourette syndrome or examined the attitudes and behavioural intentions of other groups towards children and adolescents with TS. Studies were also included if they assessed stigma, discrimination and victimisation 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 the courtesy and affiliate stigma of family members.

2.7.3. Exclusion criteria

Studies have demonstrated that the findings pertaining to how stigma is enacted and experienced among adults cannot always be generalized to younger populations (Sirey et al., 2001). Since the aim of this study was to explore what 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 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. Theses, dissertation papers, editorials, letters, conference presentations, review articles, book chapters were excluded due to potential bias.

2.7.4. Quality appraisal

Although the studies that have been included are assessed throughout the results section, a specific quality assessment tool has not been used in this systematic review. This is due to the fact that it was not the aim of present study to provide a hierarchy of evidence and quantitatively weight the evidence of the studies but to provide comprehensive understanding of the current literature (Petticrew & Roberts, 2006).

2.7.5. Research strategy

The research from the three database searches yielded 3635 results. The remaining 2445 articles that were identified after the duplicates were removed were screened for inclusion based on their title. After eliminating articles that clearly lacked relevance to the current systematic review, 38 studies were assessed based on their abstract and against each of the inclusion criteria. Of these papers, 11 articles were not empirically based studies, and 2 were comprised of participants who were older than 18 years old and were therefore excluded. Full copies of the 25 remaining articles were retrieved and assessed for eligibility. In 5 articles there was insufficient data on stigma or bullying during childhood and puberty; in 2 articles measures about the attitude towards individuals with TS were taken only after a psychoeducational intervention was employed; in 3 studies only, attitudes towards adults with TS was sought. The reference lists and bibliographies of the 15 final retrieved articles were also hand-searched to identify studies that were not found in the databases. Four relevant articles were recovered that fitted the inclusion criteria (see Figure 1, p. 39) using this process.

This left a total of 19 peer-reviewed articles. Only 17 of these reported data from independent projects as two sets of articles discussed data from the same study.

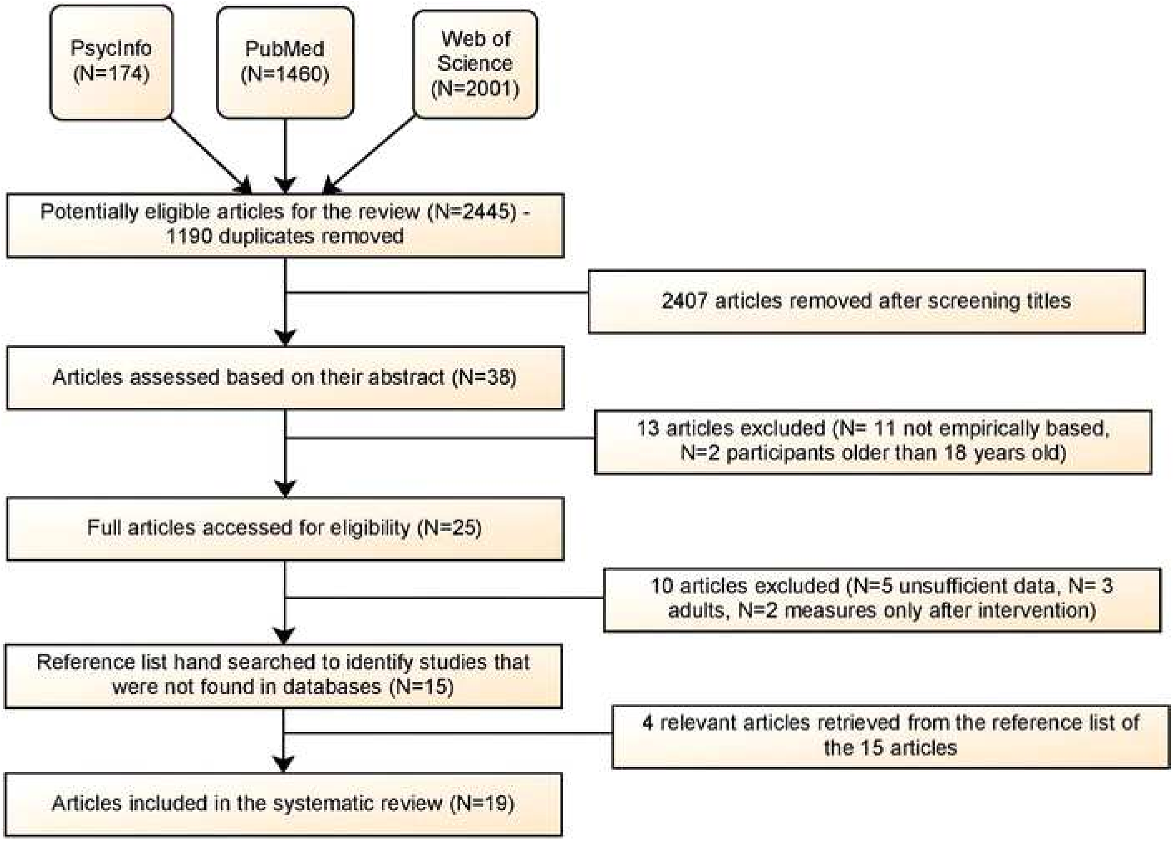

Figure 1 - Flowchart of the literature search and retrieve process.

2.7.6. Data extraction

Information collected from studies included location, sample approach, design and methodology, aspects of stigma studied and measures used as well as results obtained. Personal contact was sought with all of the main/corresponding author of the studies via email in order to clarify ambiguous points in the articles and analyse the questionnaires that had been developed by the authors and used in the studies. Four of the scholars responded with additional information. A surprising number of scholars could not retrieve the questionnaires that they based their studies on and therefore an overall evaluation of their measures was not made possible.

2.8. Results

Seventeen empirical studies were relevant to the aims of the systematic review (see Table 2, p. 58). The studies were diverse in their primary research focus and design. Six used qualitative measures and eleven used a quantitative research approach.

Six studies explored the experience of having TS in relation to everyday life (Buckser, 2008; Buckser, 2009; Cutler, Murphy, Gilmour, & Heyman, 2009; Grace & Russell, 2005; Rivera-Navarro, Cubo, & Almazán, 2009; Rivera-Navarro, Cubo, & Almazán, 2014; Wadman, Tischler, & Jackson, 2013). Four studies used an experimental study design to explore the social acceptability of youths with TS in comparison to individuals that did not have TS or participants’ knowledge about the condition before and after an educational intervention had been implemented (Boudjouk, Woods, Miltenberger, & Long, 2000; Friedrich, Morgan, & Devine, 1996; Holtz & Tessman, 2007; Storch et al., 2007). One large survey explored the impact of TS in the lives of individuals with the condition (Conelea et al., 2011; Zinner, Conelea, Glew, Woods, & Budman, 2012). Finally, one study explored the clinical characteristics of a population of people with TS in Costa Rica (Mathews et al., 2001).

2.8.1. Population and countries

The study sample in the reviewed studies (n=2,218) 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), and Denmark (1) Costa Rica (1).

2.9. Data synthesis

For the qualitative studies data was extracted mainly from the results section of the publications. Thematic synthesis was performed in the qualitative and mixed method studies according to the approach described by Thomas and Harden (2008). Firstly, line to line coding was employed to identify codes and themes across the included studies. In the second stage descriptive themes were developed by relationships articulating between the themes. Lastly, analytic themes were developed by comparing similarities and differences across the studies in the descriptive themes between the studies. These themes provide new conceptualisations beyond the initial explanation of the studies.

2.10. Quantitative studies

2.10.1. Awareness of Tourette's syndrome

Four studies explored knowledge about TS among professionals (Marcks, Woods, Teng, & Twohig, 2004), health care students (Katona, 2013), in service teachers (White et al., 2011) and elementary school children (Holtz & Tessman, 2007). Teachers and elementary school children revealed only a basic knowledge about TS, with teachers displaying a better knowledge of management strategies than etiological causes of TS. This indicates the need for condition-specific training.

The surveys on professionals and healthcare students suggested an adequate understanding of the syndrome. However, the generalisability of the rates of knowledge is questionable due to the small sample size and the low response rates. Only 6.6% of the targeted sample participated in the survey of Katona and 17.5% of the professionals in the study of Marcks. Moreover, all of the professionals in Marcks study (2004) that completed the questionnaire had either treated an individual with some form of tic condition or had worked directly with a patient with TS. Although only speculations can be made regarding the reasons for this high non-response rate it is possible that this problem may have influenced the substantive results of the research. Therefore, it is unlikely that these findings are representative of the general group of professionals as a whole.

With the exception of the survey of Marcks et al. (2004), questions of prognosis, treatment and contagiousness were not addressed. They were also based on questionnaires that were developed by the authors to measure knowledge about Tourette's and researchers failed to report any details of the validity of the measures. Most importantly, the articles did not highlight the myths and misconceptions that were endorsed by the aforementioned populations in order for more realistic and tailor- made educational interventions to be developed.

2.10.2. Attested attitudes and behavioural intentions towards children and adolescents with Tourette's syndrome

The studies that pertained to the attitude (Boudjouk et al., 2000; Friedrich et al., 1996; Holtz & Tessman, 2007; Katona, 2013) and behavioural intentions of the participants towards an individual with TS (Friedrich et al., 1996; Holtz & Tessman, 2007) indicated the negative impact the tics have on social interaction.

In rating their own views, youths without TS showed less positive attitudes towards youths with TS in comparison to typically developing peers (Boudjouk et al., 2000; Friedrich et al., 1996). However, despite the adverse impact the tics had on the evaluation of an individual, a comparison to other studies that utilise the same assessment tool (Adjective Checklist) suggested that individuals with TS are rated more positively than children with ADHD (Law, Sinclair, & Fraser, 2007) autism (Swaim & Morgan, 2001) or obesity (Bell & Morgan, 2000). One possible explanation for this could be that the attributes that are included in the Adjective Checklist do not adequately represent the aberrant behaviour of a child with TS.

With respect to gender, boys appeared to have a more favourable attitude towards children with TS (Friedrich 1996), whereas adolescent females with TS were seen less favourably than females without tics (Boudjouk, 2000).

Only one study that evaluated the behavioural intention towards a child with TS suggested that youths showed no reluctance to engage in social activities with an individual with TS (Friedrich et al., 1996). On the contrary the study of Holtz suggests that youths would avoid interacting with individuals with TS. The discrepancies between the results of the two studies could be interpreted by considering the composition of the samples. The evaluators of the Holtz study also consisted of adolescents. Recent literature suggests that adolescents exhibit a less positive attitude towards peers that could be perceived as different, in comparison to children and the need for homogeneity and order within the group is more prominent during puberty Furthermore, only 60% of the initial sample (n=82) completed the specific questionnaire in Friedrich's study regarding the behavioural intentions and low response rates may indicate misleading results (O'Driscoll, Heary, Hennessy, & McKeague, 2012).

The findings of both Friedrich's and Boudjou's studies should also be interpreted with caution due to the fact that the artificially created stimuli used to introduced the participants to the symptomology of TS lacked authenticity. Both videos used a child/ adolescent actor portraying solely motor tics. However, in contrast to other tic conditions, a basic component of Tourette's syndrome is vocal tics (American Psychiatric Association, 2013). Storch's research (2007) has demonstrated that the effects of the phonic tics could be more destructive in interpersonal interaction in comparison to the motor ones. Therefore, by presenting only one element of the symptomology of TS, the participants' evaluation may have been positively affected. On the other hand, Holtz & Tessman (2007) did not use a visual stimulus or a written vignette to introduce the participants to the symptomology of TS. Thus, initial measures were completed without the participants having any indication of the manner in which the condition manifests. However, it can reasonably be assumed that a sizeable proportion of children and adolescents who took part in the study had no awareness of the syndrome. They may therefore have been influenced by the mere fact that it is a condition that is stigmatised and an intervention is warranted to promote better attitudes towards people who have TS. In other words, the results of the measures may not truly reflect the attitude they would have towards a peer with TS but rather their fear of an unknown syndrome that is socially inacceptable. In addition, none of the studies controlled for level of contact with individuals who exhibited 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.

2.10.3. Discrimination and bullying

Discrimination, the behavioural component of stigma for children and adolescents with TS was only addressed by one survey, in which the informants were the parents (Conelea et al., 2011). Four questions of the Tourette's syndrome Impact Questionnaire for Children (TSIS-C) focused on the subject of prejudice and disadvantageous treatment of their children. The vast majority (75%) of the participants felt that their children had been treated differently because of their tics. The study also concluded that discrimination was affirmatively associated to tic severity as well as comorbid conditions.

Four quantitative studies attempted to estimate the rates of peer victimisation in children with TS (Debes, Hjalgrim, & Skov, 2010; Packer, 2005; Storch et al., 2007; Zinner et al., 2012). The discrepancies between the findings, which range from 26% (Zinner et al., 2012) to 44.7% (Debes et al., 2010), 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 (Storch et al., 2007; Zinner et al., 2012). In the study of Zinner at al. (2012), the ambiguous response options and the arbitrary cut off point that distinguished victims from non-victims limited the reliability of the results. Taking into consideration that one of the basic characteristics of bullying is repetitiveness the respondents are encouraged to use their personal frame and interpretation, by measuring the frequency of the incidents with the use of vague terms such as "sometimes" or "most of the time" (Bryman, 2015). In other words, there is no specific reference period provided (e.g. the past 30 days), to which the participants should relate their assessment (Solberg & Olweus, 2003). Moreover, the cut-off point that differentiated the victims from the non-victims appears to be fairly arbitrary. However, it has been pointed out by the study of Solberg and Olweus (2003), who examined the psychosocial adjustment of students from the general school population that have admitted to past experience of being bullied that the lower cut off point that separated the victim status from the non-victim is being bullied two to three times a month. These students 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 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.

Taking into account that the study of Zinner (2012) and Conelea (2011) were part of an online survey (Tourette Syndrome Impact Survey), sampling issues should be taken into consideration. It is impossible to calculate the response rate and the non-response bias since the size of the potential population is difficult to determine. Recent studies have suggested that the respondents of online surveys are people who have exhibited a predominant interest on the subject and therefore are more willing to complete the online questionnaire (Graefe, Mowen, Covelli, & Trauntvein, 2011). We could presume that in the current study, the self-selected respondents experienced some sort of negative impact due to TS. It is therefore possible that rates of victims and people that have been socially excluded because of their tics are overrepresented.

The studies that based their estimates on the reports from the parents of individuals with TS (Packer, 2005; Debes et al. 2010) reported extremely high rates of bullying (Packer, 39% of the individuals have been teased due to their tics; Debes, 44.7 % of the cohort endured teasing because of their tics) in comparison to the self-report studies. This is surprising taking into consideration that victims are not always inclined to disclose their experiences to their parents (Houndoumadi & Pateraki, 2001) or divulge that they've been bullied to any adult (Unnever & Cornell, 2004). Therefore, this data source may be less suited for bullying estimation since both studies 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 problematic. It is therefore possible that the Danish term “modning” that was used in the Danish questionnaire (Debes et al., 2010) may incorporate a wider meaning of teasing which may have led to an overestimation (Smith, Cowie, Olafsson, & Liefooghe, 2002). Finally, ambiguous concepts such as “a bit”, “a lot” and “a little” confuses the answers of the informants and impedes the accuracy of the findings.

Although there are discrepancies between the findings, the limited available data suggest that the prevalence rates of bullying among individuals with TS are significantly higher than the estimates among the typically developing school-age population (9 %) or youths with other disabilities (Storch et al., 2007).

Another confounding issue that has not yet been addressed is cyberbullying in relation to individuals with TS. Thus, little attention has been paid to this recent form of bullying that ensures the anonymity and unaccountability of the perpetrator in relation to individuals with TS (Spears Brown & Bigler, 2005).

2.11. Qualitative studies

Surprisingly no single qualitative study currently exists that focuses primarily on the experience of stigma in relation to TS. However, by studying the narratives of individuals with TS the concept of stigma or the components of it emerge. Through these studies, the perspectives of the individual with TS, their parents and the specialists were revealed.

2.11.1. Misconceptions and stereotypes about the condition

The majority of participants described some form of devaluation as a response to their tics. Even in the present day and in the developed world, causal beliefs of TS were shrouded in misconceptions. Surprisingly, the manifestation of the symptoms of TS was still linked to demonic possession. These beliefs that stem from ignorance and archaic religious prejudice still traverse societal boundaries and contributed to the formation of stigma. More specifically, a teacher within a Christian school in Australia suggested that tics were caused by the possession by the devil (Grace & Russell, 2005). Similarly, in Indiana under the similar circumstances an improvised exorcism was employed which further contributed to the isolation and withdraw of the individual with TS (Buckser, 2009).

Such misconceptions were further complicated by the stereotypes that have been linked to individuals with TS and most specifically coprolalia. The “virtual social identity” (Goffman, 1963) of the individual with TS, which may be quite different from the actual social identity, is deemed to possess the attribute of uncontrollable fits of profanity.

> *“Coprolalia is an important cause of social maladjustment because of the lack* of understanding of this sympto*m in most schools or workplaces” (Health* professionals) (Rivera-Navarro et al., 2014).

> *“When you say you 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) (Wadman et al., 2013).

People with TS and their families suggest that the very term “neuropsychiatric” which is used to define the essence of Tourette is in itself stigmatising. More specifically, this ambiguous and unfamiliar clinical term correlates to negative outcomes in their mind.

> *“The quality of life of subjects and families who are diagnosed is, at the moment* terrible, so we say now what should I do? We see a very dark future on the *horizon. Why do they say neuropsychiatric disease?”* (Rivera-Navarro et al., 2009).

2.11.2. 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 (Cutler et al., 2009) a subtle form of discrimination was found to take place in some schools. Teachers were unwilling or unaware of how to make allowances in order to cater to the individualised needs of children with Tourette’s syndrome (Rivera-Navarro et al., 2014). 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 simple classroom modifications 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 (Grace &Russell, 2005). Thus, instead of removing the barriers that inhibit the learning process as the Disability Discrimination Act posits, some teachers through their acts suggested that the problem is within the individual and not in the structures of society. As a professional said:

> *“Somebody would have to speak with some teachers with the objective that they* see the lack of understanding of the conditions/symptoms; this is the lack the pu*blic administration has related to this condition”* (Rivera-Navarro et al., 2014).

2.11.3. Bullying and the aftermath

Qualitative descriptions of the behavioural experience of bullying were very poorly reported. Taking into consideration that most of the studies examined the experience of individuals with TS in general, the accounts concerning discrimination and bullying were limited, non-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 bullying and individuals with TS and the emotional distress they endured (Buckser, 2008; Cutler et al., 2009; Grace & Russell, 2005; Rivera-Navarro et al., 2014).

> *"In class I felt embarrassed, I couldn't pay attention because I heard a laugh* and I thought my colleagues were laughing at me and I always kept my eye on *what my colleagues thought, said or did and I had a bad time."* (Riverra-Navarro et al. 2014).

The subsequent aggressive and sometimes violent responses of the victims were also recorded.

> *"They like call me Tourettehead a*nd 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"* (Cutler et al., 2009).

Moreover, victimisation was linked to social isolation.

> *"I feel like I 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 picked on at school"* (Grace &Russell, 2005).

Only one study examining the experience of adolescents with TS reported that the participants had never endured victimisation (Wadman et al., 2013). Despite the fact that it was acknowledged by the participants that the tics negatively affected their relationship to their peers, bullying was not reported. However, 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.

In the study of Matthews (2001) the participants disclosed incidents of bullying, discrimination and social rejection. However, they reported no impairment in their social lives due to the tics. This further confirmed the cultural character and perception of social rejection. Moreover, another form of discrimination was attested within the society of Costa Rica. By conceptualising the tics as a form of bad behaviour or voluntary habits individuals with TS were denied access to the appropriate medical health care services that were warranted. This form of discrimination that stems from ignorance results in the individuals with TS not receiving the appropriate treatment.

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

In the personal testimonies of individuals with TS self-stigma is also perceptible. In contrast to the enacted stigma described above, self-stigma refers to the devaluation of one's self. This emotion could stem from prior personal experience or could be built upon the perceived social unacceptability of TS (Scambler & Hopkins, 1986). 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 (Wadman et al., 2013). 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 fury of tics) Yeah, too stupid!
> (Grace & Russell, 2005).

By masking their tics, concealing their "spoiled" identity, avoiding situations where they were in the company of strangers and withholding the distressing fact that they had TS because of the anticipated fear of being socially rejected, the underlying self-stigma is evident (Buckser, 2009).

> *"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"* (Cutler et al., 2009).
>
> *"when you're meeting new* friends you wanna stop tic-you wanna stop ticcing *as hard as you can"* Kurt (Wadman et al. 2013).

2.13. Courtesy and affiliation stigma

Rivierra- Navarro (2009) found that caregivers felt they were discredited as parents having being accused by medical professionals of magnifying the symptoms of their children. Some parents reported constrained relationships with friends and relatives. The latter often refused to acknowledge the existence of the condition and therefore to admit that the parents were in need of additional support. This form of intolerance impelled parents and family members to become socially isolated, and alienated from both their friends and their extended family.

> *"to* invite people to my house is not possible-neither family, nor friends, nor *anybody; it is impossible…"* (Rivierra-Navarro, 2014).

A number of qualitative studies indicated affiliate stigma in families (Rivera-Navarro et al., 2009; Rivera-Navarro et al., 2014). Parents' guilt and self- blame after learning about the inheritability of the syndrome may have inhibited them from seeking help and guidance.

> *"I think that many families stopped visiting doctors because they were afraid of* the fact that TS is an inhe*rited disease"* (Rivierra-Navarro, 2009).

In response to courtesy stigma parents sometimes adopted the stigmatising views held by the public and impelled their children to conceal, control or mask their tics in public places. Thus, the embarrassment triggered by observing deviant behaviour that had been mandated from society had been adopted by the parents. Some parents in the studies came to act as “stigma couches” (Goffman, 1963) teaching them that the tics represent an undesired differentness. They emphasised the restrictions the condition imposed and coached their children to conceal their tics from others (Conrad & Joseph, 1983).

> *“…what’s valued in our society are confident people who keep their business to themselves and their voices quiet…I am conscio*us all the time of having to be *the one who pulls her down”* (Australian mother-Jill) (Grace & Russell, 2005).
>
> *“When I am going to my school, she[mother] says to me David, do not show* your tics and I look at her with displeasure and tell her if you tell me that again *I will leave”* (Rivierra-Navarro, 2014).

2.14. Discussion

Studies assessing the Quality of Life (QoL) of individuals with TS (Eddy et al., 2011) have documented that youths with TS have diminished 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 (Elstner, Selai, Trimble, & Robertson, 2001). Thus, low social activity engagement which creates lower QoL and poorer emotional functioning (Conelea, Busch, Catanzaro, & Budman, 2014), may be due to the stigma of socially unacceptable behaviour. Yet, this systematic review revealed a scarcity of data concerning the public’s awareness, attitude, and behavioural intentions towards individuals with TS. Also, convenience and highly educated samples presented an underestimation of the misconceptions about the condition (Marcks et al., 2004; White et al., 2011).

Furthermore, there is a dearth of research concerning the way stigma is constructed and interpreted by the stigmatisers in relation to TS. Hence, the studies to date are based on questionnaires with restricted answers that do not give the participants the opportunity to freely express their thoughts and feelings to gain rich qualitative data on the origins on stigma. By turning away on the producers of the exclusion, stigma is seen as something within the individual, rather than a designation that others affix on the person. Indeed, it is common practice for the stigmatisers to justify their behaviour by bestowing the title of the "odd student" to the victim (Teräsahjo & Salmivalli, 2003). However, there is 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. Thus, although data exists on how stigma and bullying is perceived and experienced by the individuals with TS, a framework for understanding the factors that shape stigma and exclusionary practices towards individuals with TS has not been formed. 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 the "typical" children. Moreover, it has not been assessed if specific peers, or rather the peer groups in general, stigmatise and subsequently exclude individuals with TS. Hence, research needs to investigate if devaluation of individuals with TS is a socially sanctioned behaviour among children and adolescents. An understanding of the awareness, attitudes, stereotypes and exclusionary practices with respect to TS could provide a basis for developing effective and tailor-made interventions aimed at reducing the misconceptions and the social stigma associated with the condition.

Because of the variety of the assessment methods and the small sample sizes used no gender or age distinction in perceived 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 that have been used do not permit us to explore the kind of victimisation that is prominently associated to TS. No association between age and victimisation is revealed in any of the studies and only one report suggested that boys with TS reported higher levels of peer victimisation (Storch et al., 2007).

Another issue arising from the review is that studies are warranted that explicitly address public stigma and the experienced way 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 that are 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 actual identity of a person. Also, as it has been argued, 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 (Sartorius, 2008).

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 (Mashiach-Eizenberg, Hasson-Ohayon, Yanos, Lysaker, & Roe, 2013). Future research is also required into courtesy stigma in families, especially siblings and extended family because research so far is limited.

2.15. Limitations

The limitations of this study should be pointed out. The studies were conducted mainly in the U.S.A and in the United Kingdom, thus mostly reflecting stigma of TS in correlation to western civilization. There are no data on stigma associated to TS in Eastern and African countries in the current review. One explanation for this could be the 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. Furthermore, because conditions that could be devalued at a specific time within a given culture could be accepted later, it is of great importance to investigate it from a cross- cultural perspective (Pescosolido, Olafsdottir, Martin, & Long, 2008). Future research needs to address these limitations.

It is also important to consider some limitations in the TS sample presented in the studies. Firstly, there is a dearth of research on population based samples and the population is overrepresented by clinically referred cases. Secondly, there is a lack of heterogeneity in the sample population which consists mostly of Caucasians from Western cultures. Moreover, children and adolescents that have been involved in TS organisations and support groups represent a notable percentage of the children and adolescents studied. These individuals however may have been more willing to ask for support and subsequently the data they provided possibly has been influenced by their involvement in such groups and organisations. Furthermore, participants that have been ascertained through specialist services only represent individuals that have impaired lives due to the tics. However, DSM-IV-R broadened the TS population by omitting the criterion of everyday-life impairment (American Psychiatric Association, 2000). Also, in the majority of the studies the participants with ADHD and TS have been studied as a homogeneous group. However, a large number of studies suggest that children with TS and ADHD face greater social difficulties in comparison to individuals without ADHD (Carter et al. 2000). Lastly, it should be emphasised that the study that relies on retrospective reports of the experience of bullying and stigma is confounded by the fact that the heterogeneity of the age group of participants does not permit us to assess if the participants' reports of their experiences represent a different generation.

2.16. Summary

The systematic review on stigma and Tourette's syndrome indicated that the subject has not been sufficiently examined and there is scarce empirical research which investigates how stigma is enacted but also experienced by individuals with TS. Most importantly, there is insufficient data on how people with TS are perceived by the public and why exclusionary practices might take place.

Table 2 - Studies reviewed in the systematic review.

Authors Names and year of publication	Location	Study sample	Sample Approach	Design and methodology	Aspect of stigma studied- measures used	Key findings in relation to stigma
Friedrich et al., 1996	United States of America	153 children without TS (8-11 years old) from schools were randomly assigned to rate individuals with TS (n=49), individuals without TS (n=53) and individuals with TS and information (=51)	Self-selected sample from mainstream school	Between group design- 3 groups a) TS b) no TS c) TS and information	Assessed attitudes and behavioural intentions towards children with TS. Adjective Checklist Activity Preference Scale, Foley Questionnaire, Modified Foley Questionnaire	• Youths rated children with TS less favourably than individuals without tics. No distinction was made in behavioural intentions between the child with TS and the child without. No effects of information.
Boudjouk et al., 2000	United States of America	25 adolescents without TS (13-14years old) were assigned to rate peers with TS and without TS	Self-selected sample from a mainstream school	Within group design -1 group -TS condition, no TS condition	Assessed attitudes towards individuals with TS- Social Acceptance Scale	• Youths who displayed tics were rated less favourably than youths who did not exhibit tics
Marcks et al., 2004	United States of America	67 professionals (35 physicians, 32 psychologists)	Postal Questionnaire	Cross sectional survey. Questionnaire derived by the authors	A 2-page survey assessed knowledge about TS, beliefs in untested claims about TS	• 77% responded correctly to the general knowledge questions

						• 6.1% thought that coprolalia was present in the majority of cases of TS • 46.1% endorsed the myth that the tics deteriorate during adulthood.
Packer, 2005	United States of America	69 parents or guardians of 71 adolescents or children with TS (6.6 -17.8 years old)	Survey advertised by TSA	Cross sectional survey. 15-page parent-report questionnaire derived by the author.	One multiple choice question regarding teasing	• One third of the parents of the children reported that their children had never been teased • 29% of them were either teased a bit but never rejected • 10% were teased a lot but never rejected • 18% rejected a bit, 10% rejected a lot because of their tics
Holtz & Tessman, 2007	United States of America	179 (7-15 years old) elementary school children from 6 schools.	Self-selected sample from mainstream schools without TS	RCT	Assessed knowledge about TS, attitude and behavioural intentions towards individuals with TS- 8 item knowledge questionnaire, Modified	• The measures taken at the pre-intervention stage revealed that individuals with TS were rated more positively after a psychoeducational intervention

					CATCH, Foley Questionnaire	
Storch et al., 2007	United States of America	59 youths with TS and CTD (8 - 18 years old) - comorbidities included ADHD, OCD, MD, GAD, ODD, SP, AD and PD	Convenience sample of outpatients from the University of Florida – Department of Psychiatry Child and Adolescents OCD and Tic Clinic	Cross sectional survey	Assessed bullying in youths with TS- Schwartz Peer Victimization Scale	• 27% of individuals with TS and CTD reported peer victimisation • Higher rates of bullying in comparison to individuals with Type 1 Diabetes and healthy population • Peer victimisation is associated to loneliness
Debes et al., 2010	Denmark	314 youths with TS (5.3 – 20 years old) comorbidities included ADHD and OCD	Convenience sample- Tourette syndrome clinic at Glostrup University Hospital, Denmark	Cross sectional survey study	Assessed teasing in youths with TS. Structured interview answered by both parents and children-one question regarding teasing	• 44.7 % of the participants reported being teased • The study suggests that a positive correlation exists between co-morbid disorders and victimisation (TS+ comorbidity 52.9%, TS only 31.4%)
Conelea et al., 2011	United States of America	740 parents of individuals with TS Same sample of children as Zinner 2012	Web based survey advertised by TSA, emails to members, newsletter announcements	Cross sectional survey study.	A 4-item questionnaire derived by the authors to measure discrimination	• The majority of parents reported that their child endured some form of discrimination • Discrimination is positively correlated

						to other comorbidities
Zinner et al., 2012	United States of America	N= 211 youths with TS and CTD (10-17 years old) -no comorbidities		Cross sectional survey study.	An 8 item questionnaire was derived by the authors to assess bullying	• 26% of individuals with TS and CTD reported peer victimization • Symptom severity is positively associated to victimisation
White et al., 2011	United states of America	137 in service elementary school teachers (20.5 years old; 78 females)	Self-selected sample from 5 elementary schools in south-western Connecticut	Between group and within group design	27 item questionnaire assessed knowledge about TS, OCD and ADHD. 8 items specifically concentrated on TS	• TS knowledge (56%) was higher than ADHD (38%) but poorer than OCD (61%) • Myths regarding the causes of TS still linger among the teachers • Classroom experience did not correlate to awareness of TS
Katona, 2013	United Kingdom	94 healthcare students	Web based Survey advertised through St Georges’ student newsletter and individual emails	Survey study design	A 16-item questionnaire derived by the author Assessed knowledge and attitude about TS and epilepsy.	• Less knowledge & more unfavourable attitude (5% would object to their child associating to people who showed signs of the disease) towards TS in comparison to epilepsy

Buckser, 2008	United States of America	17 adult participants aged from 21-62 years old	Self-selected sample recruited through the Tourette Syndrome association of Indianapolis	Qualitative study. Ethnographic approach Interviews and informal interaction in support groups with the participants.	Bullying, discrimination	• Participants disclosed incidents of bullying that occurred during their childhood and puberty
Buckser, 2009						• Bullying was prominent during primary and secondary education. • Stigma was more apparent in restrictive environments.
Rivera- Navarro et al., 2009	Spain	6 adolescents with TS (11-17 years old 6 parents 5 doctors	Convenience sample from informal support group in Madrid	Qualitative descriptive design. 5 Focus Groups on Diagnosis of TS and the subsequent stigma.	Courtesy stigma	• Stigma stems from the term neuropsychiatric
Wadman et al., 2013	United Kingdom	6 adolescents with TS (14-16 years old)-comorbidities included ADHD, OCD, excluded -ASD	Convenience sample recruited from Tourette's Action UK as a part of a bigger study	Qualitative study. Semi structured interviews-interpretative phenomenological analysis	Self-stigma, stereotypes, social avoidance	• The importance of friendships • Concerns about the future • No incidents of bullying reported
Rivera-Navarro et al., 2014	Spain	12 adolescents with TS (11-18 years old) 12 parents 8 health professionals	Convenience sample recruited from support group in Madrid	Qualitative study. Separate focus groups for adolescents, parents and professionals. Delved into the	Discrimination, courtesy stigma, self-stigma	• Stigma, social maladjustment, victimisation among adolescent with TS

				impact of TS in school and family		
Mathews et al., 2001	Costa Rica	85 participants with TS (aged from 5-29 years old)	The clinical characteristics of the population of TS in Costa Rica	Mixed method study. Qualitative data from informal interviews with the 85 participants	Bullying, discrimination	• Although the majority of participants disclosed incidents of bullying and harassment, they reported no impairment due to the tics
Grace & Russell, 2005	Australia United States of America	26 children with TS (8-15.5 years old)- comorbidities included ADHD and OCD 34 parents	Not stated	Mixed Method study. Separate interviews between parents and children with TS focusing on the school experience. Ecocultural approach.	Self-stigma, bullying, courtesy stigma, affiliate stigma, discrimination	• Three groups of families emerged that were respectively mostly satisfied, essentially dissatisfied and entirely dissatisfied from the school experience. A form of stigma and exclusion was experienced by all groups
Cutler et al. 2009	United Kingdom	57 youths with TS (8-17 years old) – comorbidities included ADHD and OCD	Self-selected from a specialist TS clinic at a national children's hospital	Mixed method study. Focus groups delving into the quality of life for young people with TS	Bullying Self-stigma	• Anticipation of rejection • Bullying and frustration • The social unacceptability of TS

* ADHD Attention Deficit Hyperactivity Disorder, AD Aspergers Disorder, CATCH Chedoke-McMaster Attitudes Towards children with Handicapped Questionnaire, CTD Chronic Tic Disorder, MD Major Depression, GAD Generalised Anxiety Disorder, ODD Oppositional Defiant Disorder, SP Social Phobia, PD Panic Disorder, OCD Obsessive Compulsive Disorder.

3

School-based Interventions that Aim to Reduce Stigma in Behaviourally-defined Conditions: A Systematic Review

3.1. Introduction

Chapter two of the thesis pertaining to stigma and youths with TS clearly demonstrates that children and adolescents with TS may experience devaluation from their peers as a result of their condition. A large and growing body of literature highlights the detrimental consequences of bearing the visible or invisible “mark” of shame (Goffman, 1963) and the negative effects on self-concept for those belonging to a “blemished” group (Rüsch, Angermeyer, & Corrigan, 2005). Devaluation by others may have pervasive psychological and emotional impact on the stigmatised. Demoralisation, social withdrawal and isolation in order to avoid rejection may also heighten people’s vulnerability to emotional disorders (Corrigan & Watson, 2002; Link & Phelan, 2001).

In order to prevent the social isolation and marginalisation of “stigmatised” individuals, interventions that disseminate factual information and promote positive attitudes towards this population are warranted. A suitable delivery platform for these programs would be the school environment. Indeed, to design an intervention for schools was the aim of the thesis from when it was initially conceptualised. My professional experience and the literature to date have highlighted that adolescents need to be educated about the condition in order to prevent the marginalisation of individuals with TS

Classrooms provide a good opportunity to access a wide and diverse group of children and adolescents within a naturalistic setting. Furthermore, the aims of anti-stigma interventions coincide with the wider objectives of inclusive schooling that are orientated towards ensuring social participation for every pupil (Booth & Ainscow, 2002). Thus, it has pointed out that the aims of inclusive education extend beyond academic objectives and highlight the need for all pupils to be valued members of their school (Farrell, 2000). This means that every student should be accepted and experience supportive peer relations in order to develop a sense of belonging within the school. Anti-stigma/psychoeducational interventions aim to ameliorate or even prevent stigmatisation amongst adolescents and children by eliminating prejudice before it is deeply ingrained by challenging myths and stereotypes. Furthermore, they aspire to contribute to creating adults who do not discriminate and to ultimately prevent further prejudice in the society.

In order to develop an effective school-based intervention to help reduce stereotypes, and increase awareness of Tourette's syndrome, the broader literature relating to anti-stigma/ psychoeducational classroom interventions needed to be systematically reviewed. Such a review would allow an evaluation of the merits and limitations of previous interventions and in so doing, guide decisions about the feasibility, duration, intensity, and other essential components of a new psychoeducational intervention.

The systematic review of Nussey, Pistrang, & Murphy, (2013) demonstrated that only a few school psychoeducational interventions aimed at increasing awareness and understanding of TS exist, namely Friedrich, Morgan, & Devine, 1996; Holtz & Tessman, 2007; and Nussey, Pistrang, & Murphy, 2014. Furthermore, hitherto, there has been a lack of interventions aimed at tackling the stigma of TS among secondary school students and they have all been developed in order to target primary school children. This suggests that the TS interventions to date only provide a fragmentary account of the possible elements of anti-stigma interventions and so can only provide minimum evidence as to their effectiveness in increasing understanding about TS.

Due to the scarcity of TS interventions, it was considered appropriate to holistically assess interventions that aim to reduce stigmatisation against ‘behaviourally defined conditions’, that is, conditions that cannot necessarily be confirmed by any biological tests, that their diagnosis is based solely on behaviourally defined criteria and so there are no biological markers or definitive medical tests for these conditions although in general there is evident genetic underpinning. This broad category encompasses both non-syndromic developmental conditions as well as mental illnesses (Pennington, 2008). TS is a condition that can only be diagnosed based on the history of the symptoms and can therefore be embedded in this broad category.

A large and growing body of literature has systematically reviewed interventions that aim to ameliorate the stigma of mental illness and their effectiveness within educational settings (Mellor, 2014; Sakellari, Leino- Kilpi & Kalokerinou- Anagnostopoulou, 2011; Schachter et al., 2008). These reviews however focus solely on psychiatric and mental conditions and exclude the assessment of interventions that focus on developmental disorders and learning disabilities. Nevertheless, neurodevelopmental conditions such as Tourette’s syndrome, and developmental conditions like ADHD, and Autism are highly stigmatised and linked to misinformation (Gray, 2002). Indeed, it has been pointed out that condition that manifest in form of deviant and disruptive behaviour which are not linked to physical abnormalities may engender stigmatisation and pejorative views. They may also be considered more controllable than physical conditions and therefore arouse more punitive and discriminatory behaviour (Weiner, 1993).

It was therefore decided that a review of all anti-stigma school-based programs for behaviourally defined conditions would enable a more holistic evaluation of the broad variety of anti-stigma interventions that have been implemented within classroom settings. Since previous systematic reviews did not explore the specific elements of the reviewed interventions, whether the content and the length of the programmes influenced outcomes, or assessed the design and methods used in relation to the aims of the interventions, the current review would address these gaps. Finally, a secondary aim of this systematic review was to guide the current intervention development, helping to identify the characteristics that might determine the effectiveness of a psychoeducational intervention.

3.2. Aims and objectives of the current systematic review

The overall aim of the current review was to identify and describe anti-stigma/ psychoeducational interventions for behaviourally defined conditions implemented within school settings, and their effectiveness in reducing stigma. It further aimed to assess the content and aims of the interventions in relation to the methodology chosen to implement it, and the measures used to determine its efficiency.

More specifically concomitant aims were to:

- Categorize the various intervention strategies and identify the different components that were utilised in order to combat stigma, foster knowledge and positive attitudes;
- Identify an element or a combination of key components which contributed to the efficacy of an intervention;

- Determine if there was a certain time that the interventions needed to last or be implemented in order for them to be successful and produce stigma reduction. In other words, to identify if more information about a condition, and/or more exposure to individuals with the condition resulted in more positive outcomes;
- Review the methodology and measures used in the target studies, evaluating their appropriateness in relation to the aims of the research;
- Identify the limitations of studies and make recommendations for future behaviourally defined anti-stigma interventions.

3.3. Methods

3.3.1. Research strategy

In August 2014, systematic searches were performed using three electronic literature databases, PhycInfo, Pubmed and ERIC (Education Resources Information Centre). These specific databases were selected because they cover literature relating to psychology, life science and education. Search strategies were built around four sets of keywords: Intervention, school, population and behaviourally defined condition. The lists of search terms used can be seen in Table 3, (p. 69). Search strings were composed of one term of each column. Every combination of terms was included in the search strings.

3.3.2. Inclusion and exclusion criteria

3.3.2.1. Inclusion criteria

Eligible studies were school-based-interventions aiming to eliminate stigma, to raise awareness and/or to promote positive attitudes and/ or behavioural intentions. The intervention must have been implemented within a classroom environment and addressed to a group of school age children or adolescents (i.e. older than 5 and younger than 19 years old). This wide age range was chosen in order to encapsulate all of the interventions that had been implemented in both primary and secondary schools. Interventions also had to be delivered as part of the school curriculum, or at least during the students' regular school timetable. There was no restriction on the length of the intervention or the research design utilised. Thus, all studies using qualitative, quantitative and mixed-method research design were identified for inclusion. Studies must have been published in a peer-reviewed journal and publication languages were restricted to English, German and Greek, which correspond to the researcher's language capacity; this also eliminated English-language bias to some extent (Morrison et al., 2012). Reference lists that had been cited in relevant reviews and studies that met the inclusion criteria were also included, so as to encompass a comprehensive range of articles (Mellor, 2014; Sakellari et al., 2011; Schachter et al., 2008).

Table 3 - Search terms used to identify possible studies.

Intervention	**Setting**	**Population**	**Behaviourally Defined Conditions**
Stigma	School*	Peer*	Disorder*
Prejudice*	Classroom*	Classmate*	Disabilit*
Attitude*			Illness*
Education program*			

3.3.2.2. Exclusion criteria

Interventions that aimed to eliminate self-stigma were excluded as were programmes that had been implemented as an after-school activity or in non-school settings since individuals who had agreed to dedicate their free-time to participate in the research might have generally held more positive opinions about behaviourally defined conditions than their counterparts, thereby biasing the results. Interventions that used “Circle of friends” programmes to promote social inclusion were also excluded due to the aforementioned reasons (Frederickson & Turner, 2003). Programmes that were solely internet- based and did not take place in a classroom were also excluded. The search period was restricted to articles from 1994 onwards because social phenomena such as stigma changes over time, and it was judged that data from studies published before 1994 would be less relevant for today.

Interventions that aimed to eliminate stigma in any non-behaviourally defined condition i.e. medical conditions, such as HIV, and epilepsy were excluded. Physical disabilities were also excluded in addition to programmes that aimed to reduce stigma linked to demographic characteristics such as race, and sexual preference.

Articles that did not specify the instruments used to measure the independent and dependant variables were also excluded; this factor deemed the papers to be of a low methodological quality. Similarly, studies that had not been published in a peer reviewed journal were excluded. Whilst it was acknowledged that grey literature can help counterbalance publication bias, this was excluded in the current study since the methodological validity of such studies can difficult to evaluate. Book chapters, and dissertations were also excluded for the same reasons.

3.3.2.3. Data extraction and quality assessment

The Cochrane data extraction form was adapted to generate a standardised pro-forma for extraction in order to capture key features of the studies. Abstract items included: authors, date, details on study design, participant characteristics, intervention design, intervention duration, number of follow-ups, aims of the intervention, measures used, and study outcomes.

The quality of papers that were included were appraised by reviewing their designs and methodologies using the Mixed Methods Appraisal Tool (MMAT) (see Table 5, p. 108). This instrument's validity and reliability have been verified (Pace et al., 2012), and the MMAT offers the best and most comprehensive tool for assessing studies of multiple method types. It has also been used in numerous complex systematic literature reviews that include qualitative-, quantitative-, and mixed-method studies. The four criteria used to determine the quality score varies by design. For quantitative randomised controlled studies, randomisation, allocation concealment, completeness of data, and response rates was assessed. For non-randomised quantitative studies, the appropriateness of the measures, the recruiting procedure, the comparability of the groups, and completeness of data was assessed. The data sources, data analysis, research contexts, and researchers' influences were assessed in the qualitative studies. For a mixed methods study, both the appropriate section for the quantitative component and the qualitative component are used. The results from the tool provided an overall methodological score calculated as a percentage ranging from 25% for 1 criterion met to 100% for 4 criteria met with higher percentages indicate better methodological quality. For mixed methods studies, the overall quality score is the lowest score of the quantitative and qualitative study components.

As opposed to the systematic pertaining to stigma in youths with Tourette's syndrome (Chapter 2) a quality assessment tool was deemed important since it was the aim of the study to quantitatively weight the evidence and assess the interventions' effectiveness and efficiency in eliminating stigma. Thus, the aim of this systematic review was not only to provide a narrative but to weight the evidence.

3.4. Results

The research from the three chosen database searches yielded 4476 results from which 690 duplicates were removed. The titles of 3786 articles were assessed and 3318 were excluded leaving 468 papers to be assessed based on their abstract. Of these 468 papers, 321 articles did not meet the inclusion criteria, for the following reasons: the studies used participants that were older than 18 years of age (n= 228); the interventions focused on physical disabilities (n=80); the articles were theoretically based papers or systematic reviews (n=13). A further 118 articles were excluded after they were retrieved and read in full to assess their eligibility. Fifty-five of the articles provided insufficient data; 22 included participants of an inappropriate age range (over 18-year-old); 32 evaluated interventions that were implemented as an afterschool activity; 8 focused on non-behaviourally defined condition s. Scanning of reference list elicited an additional 8 articles resulting in 38 peer-reviewed articles being retained for inclusion in the review. Only 37 of these reported data from independent projects as two discussed data from the same study (see Figure 2, p. 73).

The sample size of the included studies ranged from 59 to 3538. The majority of the studies were conducted in the USA (14) followed by Australia (5), United Kingdom (5), Germany (3), Greece (3), Canada (2), Hong Kong (1), Italy (1), Austria (1), Spain (1), and Norway (1). Only 8 of the 37 studies included primary school students as participants.

In terms of the focus of the intervention, 28 aimed to combat the stigma of different types of mental illnesses and 9 focused on developmental disorders (5 on autism, 3 on

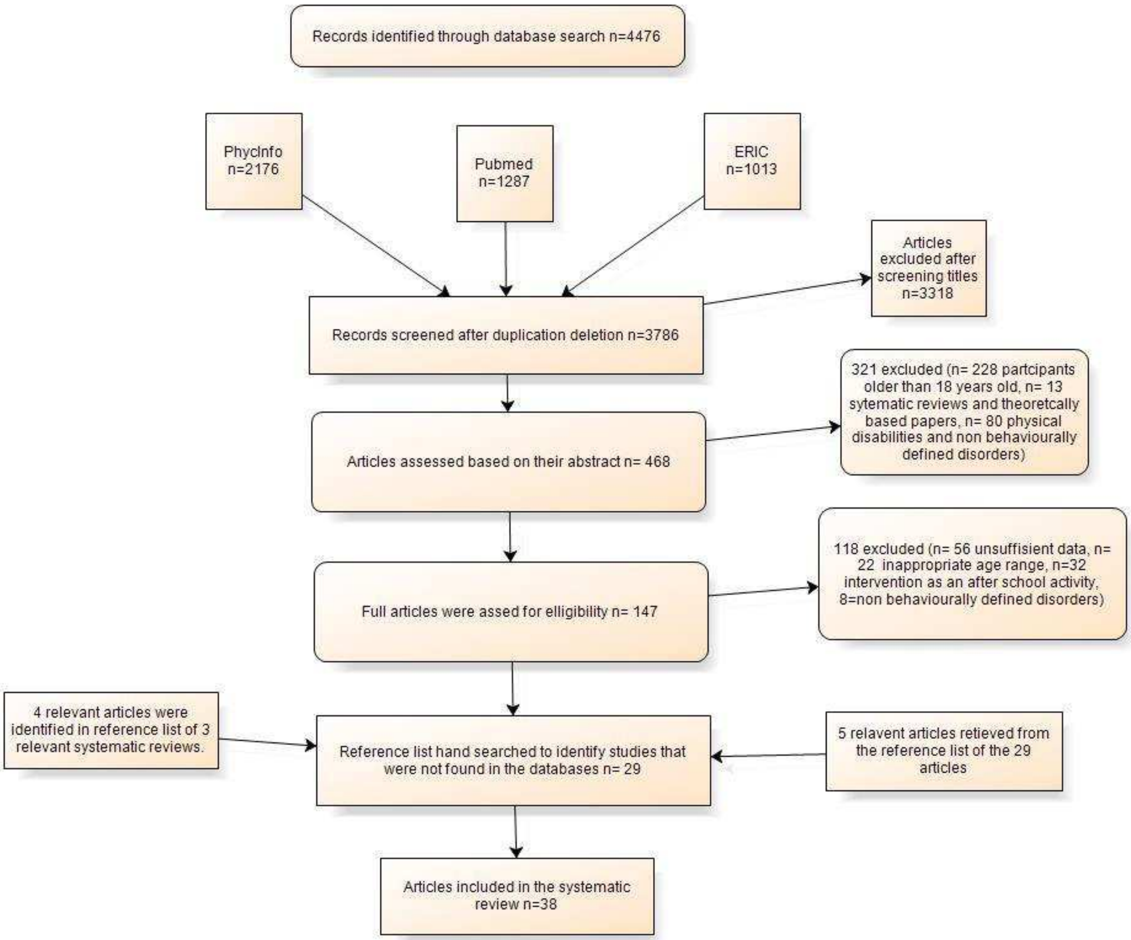

Figure 2. Flowchart of literature search and retrieval process.

Tourette’s syndrome, and 1 on ADHD). Only one study aimed to raise awareness about learning disabilities. Four studies compared different interventions types while 34 studies evaluated the effectiveness of a single programme in ameliorating stigmatisation. 21 of the studies (57%) reported significantly positive improvements while 16 showed mixed results (see Table 4, p. 91).

3.4.1. Content of the interventions

Two main forms of interventions that aimed to ameliorate stigmatisation were evident within the included studies; those that relied solely on education, and those that combined educational elements and contact with a person with a stigmatised behaviourally defined condition. None of the studies solely used contact in order to improve attitudes and reduce stigmatisation.

Only five of the included articles stated the theoretical framework on which the intervention was based upon or provided a rational for the choice of a specific approach taken (Campbell, Ferguson, Herzinger, Jackson, & Marino, 2004; Campbell, 2006; Campbell, 2007; Holtz & Tessman, 2007; Morton & Campbell, 2008). Rather, they were mainly based on the general principle of anti-stigma campaigns and failed to take into consideration the characteristics, the culture of the participants, and the ways they could be approached based on their cognitive abilities and interests. Thus, they represent 'universal interventions' and are not tailored to potential factors that influence stigma for that specific population and age group.

3.4.1.1. Educational components of the programmes

Thirty-five of the articles included in the systematic review used educational factual lectures in order to ameliorate stigma. Eighteen of these studies reported to have included interactive classroom activities to activate student learning (e.g. role playing, poster painting, web-based activities, discussion etc.) with only eight utilising passive learning strategies instead of active ones which help the learners engage with content of the lecture and each other. Only three interventions used unconventional mediums, namely performance arts (theatre, and hand puppets) to convey information to the students and reduce stigma toward mental illness among teenagers (Essler, Arthur, & Stickley, 2006; Pitre, Stewart, Adams, Bedard, & Landry, 2007; Roberts et al., 2007).

Three of these lecture-based interventions were based on the medical approach (Ruble, Leon, Gilley-Hensley, Hess, & Swartz, 2013; Swartz et al., 2010; Watson et al., 2004), the onus being on the biomedical causes underpinning the condition, with less emphasis placed upon the contextual factors that contributed to the condition. The interventions associated with mental conditions also mentioned treatment options and possible recovery outcomes. A major criticism of such an approach is that heightening focus on the genetic basis of a condition can create a wider gap between the student ('us') and the person with the condition ('them'). By pointing out possible inheritable and genetic aetiologies and thereby stressing incurable/permanence of conditions (Mehta & Farina, 1997), a sense of helplessness may also be perpetuated, emphasising the differences between the stigmatised and the audience. Arguably this approach also cultivates 'benevolence stigma' with the assumption that people with these conditions are less capable and in need of a responsible figure who can help them with their everyday and life's decisions (Brockington, Hall, Levings, & Murphy, 1993). Studies have suggested that explanatory information about a condition should be accompanied by data that makes the audience believe that the individuals who share the devalued label are not fundamentally different from them (Campbell et al., 2004).

Three studies were based on demythologizing stereotypes (Chan, Mak, & Law, 2009; Economou et al., 2014; Martinez-Zambrano et al., 2013). In the latter two programmes the facilitators of the educational programme asked the participants to mention the most commonly held stereotypes in relation to schizophrenia and subsequently provided and evidence in order to dispel them. In Chan et al.'s intervention, the participants were not asked about their understanding of the condition and the researchers appeared to make assumptions concerning the participants' level of awareness. This reflects the dearth of studies which have attempted to understand the myths and misconceptions endorsed by the specific target group. Rather, in the included studies, data obtained from the participants pre-intervention was solely used as a comparison to the post intervention figures. This means that such studies were not tailor- made to consider the specific needs of the target study group, or to propose the most appropriate ways for more positive messages about behaviourally defined conditions to be communicated.

Most of the interventions cited (n=18) gave the students the opportunity to actively learn through participation in interactive educational classroom activities including games (Essler et al., 2006), roleplaying (Economou et al., 2014; Naylor, Cowie, Walters, Talamelli, & Dawkins, 2009; Stuart, 2006), artwork poster painting (Schulze, Richter-Werling, Matschinger, & Angermeyer, 2003), and web activities (Ranson & Byrne, 2014; Staniland & Byrne, 2013). However, the majority of the aforementioned articles were found to be of poor reporting quality (n=10). Indicatively, Del Casale et al., (2013); DeSocio, Stember, & Schrinsky, (2006); Essler et al., (2006); and Ventieri, Clarke, & Hay, (2011) all suggested that their interventions included various elements including interactive activities without defining their content. Whilst role playing was a component of interventions, the authors failed to define the duration of the activity and the roles of imaginary characters. Furthermore, it is questionable as to whether or not there was enough time within short single-session interventions for the whole class to participate given the large number of components of the interventions (Economou et al., 2014).

A substantial number of the interventions that aimed to reduce stigma in relation to Autism, ADHD and Tourette's syndrome (n=9) used the technique of transferring knowledge through a direct manner to a passive, non-participating, listening audience (Campbell et al., 2004; Campbell, 2006; Campbell, 2007; Friedrich et al., 1996; Holtz & Tessman, 2007; Morton & Campbell, 2008; Saecker, Skinner, Skinner, Rowland, & Kirk, 2010; Silton & Fogel, 2012; Swaim & Morgan, 2001). This form of receptive or passive learning (Ausubel, 1978) which appears to resemble conventional teaching requires no active engagement from the audience. In fact, the majority of interventions which aimed to tackle the stigma of Autism and TS were inadequately developed to actively engage the students (Mezirow, 1997). This differed from interventions concerning mental illness in which more “transformative learning” has been utilised to engage the audience in improvisational activities including designing, researching and reporting.

A limitation of most of the reviewed studies (n=20) was that that they failed to report the details of who delivered the intervention, under what capacity they introduced themselves to the audience, and the influence this may have had on the intervention outcomes. Only three articles had specified that the intervention was delivered by a psychologist or a team of psychologists or an appropriately trained class tutor (Economou et al., 2012; Meise et al., 2000; Naylor et al., 2009). As Campbell (2006) pointed out, existing interventions have tended to focus on the message and its appropriateness to the target audience rather than the characteristics of the source that delivers the persuasive de-stigmatizing proposal which can enhance or limit the message’s influence. It is also argued that expertise and high trustworthiness of the key speaker can lead to greater attitudinal change (Pornpitakpan, 2004). Physically attractive individuals that are also likeable and have more similarities to the target audience have also found to have persuasive advantages over their less attractive counterparts (Chaiken, 1987). The study of Morton & Campbell (2008) suggests that a different source can have a different persuasive influence on different age groups. Therefore, the included studies may be critiqued for failing to acknowledge the influence of the characteristics of the communication source on the intervention outcome.

3.4.1.2. Contact-based components of the programmes

Twenty-seven studies explored the impact that contact with a “tainted” individual could have on reducing stigma. For the purpose of this review “contact” is defined as any form of direct, indirect or imagined interaction between the target audience of an intervention and a person with a stigmatising condition. Face to face, hence, direct contact, includes personal narratives of individuals who have been stigmatised followed by active discussions with the target audience. Video presentations had mostly been derived from interviews or speeches in which individuals described their condition or their journey through life. These videos do not hold the advantages of real life contact; thus an active two-way informal dialogue is not possible.

Only nine of the reviewed articles were based on onsite face-to-face contact between the typically developing research participants and individuals diagnosed with behaviourally defined conditions (Kohlbauer et al., 2010; Martinez-Zambrano et al., 2013; Meise et al., 2000; Nussey, Pistrang, & Murphy, 2014; Pinfold et al., 2003; Pinto-Foltz, Logsdon, & Myers, 2011; Schulze et al., 2003; Staniland & Byrne, 2013; Stuart, 2006). Yet the length and the quality of the interaction between the 'inflicted' individuals and the student-participants were not explicitly specified in any of them. Furthermore, whilst it was stated in all of the reviewed articles that dialogue between the two parties was encouraged, nothing was stated as to whether or not children and adolescents actively participated in the discussion, if questions were raised by the participants, or the duration of the whole process. This is despite the fact that it has long been established that the quality of the interaction in intergroup contact can determine the intervention outcomes and as such, the interaction's quality should be described rigorously (Allport, 1979). In the current interventions, if open dialogue and interaction was not facilitated, "negative contact" could have occurred which in turn could have reinforced misconceptions and stereotypes (Chen, Koller, Krupa, & Stuart, 2016). Instead of utilising direct contact with people with BDD, most of the studies (n=18) used indirect, one-way contact to attenuate stigma resulting in limited opportunities for the participants to directly interact with the 'stigmatised' and the 'stigmatised' to know whether or not the message had been accurately received. It could also be argued that indirect contact allows both groups to maintain their anonymity as well as overcoming the practical obstacles of implementing direct contact.

In the majority of these contacted based interventions the emphasis is placed on the personal narrative of the stigmatised individual and their life experience and journey through the condition. In the case of Autism, ADHD and Tourette's young people with the condition simply described the conditions symptomology (Friedrich et al., 1996; Holtz & Tessman, 2007). Similarities between the audience and the targeted individuals are highlighted. Most campaigns relating to mental illness also tried to delve into the journey of recovery and the presenters' determination to overcome the difficulties that they were faced with as well as their willpower to live a fulfilling life. In much of the publicised interventions on Autism and Tourette's syndrome the experience of having the condition is not discussed and it is questionable whether the audience have the opportunity to empathise with the target individuals.

3.4.2. Duration of the interventions

The duration of the educational interventions varied in length, from brief videos that lasted a few minutes to whole semester courses. Of the 38 studies, 21 implemented single-session interventions, some of which ran for a couple of minutes (Chan et al., 2009) whilst others lasted for two hours or more (Pinfold et al., 2003; Rickwood, Cavanagh, Curtis, & Sakrouge, 2004; Roberts et al., 2007; Swartz et al., 2010). The majority of the interventions that lasted less than an hour aimed to reduce stigma against specific conditions including Autism, TS and ADHD. This indicates that approaches to tackle prejudice against developmental conditions are still evolving (Friedrich et al., 1996; Hotz & Tessman, 2007).

The majority of the single session interventions (66%) reported positive results and appeared to be effective in improving the participants' intentions towards their peers with the target disorder. Nevertheless, only four of these studies evaluated the longitudinal effects of the intervention, and the study designs aimed to capture only the ephemeral reaction of the participants (Chan et al., 2009; Economou et al., 2012; Pinfold et al., 2003; Pinto-Foltz et al., 2011).

The multi-session interventions also varied in length and intensity. Some of the programs were delivered during two hourly sessions (Ventieri, Clarke, & Hay, 2011) while others became a part of the students' curriculum and were implemented once a week during the course of a semester (Ranson & Byrne, 2014). Other interventions carried out a more intensive approach through the course of three to five consecutive days (Conrad et al., 2009; Skre et al., 2013). The findings suggest the programmes that have been implemented for a longer time period do not guarantee better outcomes compared to shorter interventions. Indeed, 50% of the multi-session intervention studies failed to achieve a number of their goals. There did not therefore appear to be a threshold of length of intervention time that would guarantee a better impact.

3.4.3. Methodology and study design

Thirty of articles reviewed employed quantitative questionnaires in order to evaluate the effectiveness of the interventions and arguably this has limited our understanding of certain aspects of the phenomenon that could be investigated through a qualitative approach. Whilst two studies solely relied on qualitative methodology (Sakellari, Sourander, Kalokerinou-Anagnostopoulou, & Leino-Kilpi, 2014; Sulzenbacher, Schmid, Kemmler, De Col, & Meise, 2002), five articles opted for mixed-methodology to assess the perplexing issue of the stigma process (Economou et al., 2014; Kohlbauer et al., 2010; Nussey et al., 2014; Pinfold et al., 2003; Roberts et al., 2007). Yet, in the aforementioned studies the qualitative data were analysed in a quantitative manner, by applying content analysis. This meant that the onus was placed on the frequency in which certain words, phrases or concepts were repeated in the data. Such descriptive analysis risked overlooking what is not stated and what was omitted in a particular context. Taking a word or phrase in isolation of other parts of the text, can also result in a loss of meaning, and difficulties in reaching explanations for social phenomena since nuanced understanding of the participants' reasoning is impossible to achieve (Holsti, 1969).

Thirty of the reviewed studies utilised a combination of questionnaires that related to knowledge, attitude and behaviour; the three key elements of stigma. However, eight of the studies did not include a knowledge questionnaire (Campbell et al., 2004; Campbell, 2007; Friedrich et al., 1996; Martinez-Zambrano et al., 2013; Morton & Campbell, 2008; Pitre et al., 2007; Silton & Fogel, 2012; Swaim & Morgan, 2001). This could be considered a shortfall, since an important outcome of an intervention should be enhanced knowledge. Moreover, in 'within subject' and 'quasi experimental' designs with no control groups, the questionnaires covering knowledge could be used to assist a manipulation check by strengthening the assumption that better attitudes or behavioural intentions at the post-test are directly linked to increased awareness about the condition. Concurrently, it minimizes the chances that the observed changes are merely the result of the demand characteristics effect, namely a consequence of the participants' awareness of what is being investigated in the study and subconscious change of their behaviour to what they think is being expected of them (Orne, 1962). Thus, a failure to include knowledge questionnaires further weakens the external validity of the study.

The majority of questionnaires pertaining to attitudes and behavioural intentions used pre-existing validated instruments. Several studies used general population measures, adapting them for the cognitive development and age of the participants (Stuart, 2006; Economou et al., 2012). Opinions about Mental illness, (OMI) (Martínez-Zambrano et al., 2013; Pitre, Stewart, Adams, Bedard, & Landry, 2007) and the Adjective Checklist (AC) were the two most commonly used instruments to elicit participants' attitudes. The Shared

Activities Questionnaires was a commonly used tool among primary school children and adolescents in order to gauge behavioural intentions in academic, social and recreational activities (Campbell et al., 2004; Campbell, 2007; Morton & Campbell, 2008). Ten studies used social distance scales, (Schulze's (2003) scale being the most popular). These scales assessed participants' willingness to interact with individuals with stigmatised conditions by evaluating discriminatory behaviours and intent to engage in a relationship of varying levels using a Likert scale. Only a limited number of studies constructed questionnaires for their specific study (DeSocio et al., 2006; Kohlbauer et al., 2010; Rickwood et al., 2004; Skre et al., 2013; Ventieri et al., 2011) based on user-defined criteria. Arguably, these questionnaires may have captured a more precise and accurate depiction of individuals' attitudes since they included youths' preconceived ideas.

A major limitation of using predetermined questionnaires is that they are at risk of missing cultural factors linked to current participants as well as shifts in normative behaviours (for example the rise of social networking (Tsitsika et al., 2014); a number of the questionnaires used were thus rather outdated having been developed in the late 70's and 80's (e.g. The Foley Questionnaire used in Holtz & Tessman, 2007).

Twenty-eight of the reviewed studies evaluated the behavioural intentions of participants which may be defined as the consciously designed plans to engage in a specific action or behaviour (Ajzen, 1991). Thus, they were based on an individual's belief system, encompassing the notions of effort and motivation. Such questionnaires phrase questions in a manner which allows the participants to project their general belief system (e.g. "If one of my friends developed schizophrenia, I would go and see him/ her at the home"). These may be distinguished from self-expectation questionnaires, in which a person subjectively estimates whether she/he will be acting or behaving in the targeted way. In contrast to intentions in predicting her/his behaviour one also takes into consideration the various environmental facilitators or impediments that could influence the attempt to carry out the intention. It is therefore a broader construct.

Consequently, although the motivational driver to perform an act is charted, this is not sufficient for predicting subsequent behaviour. Shephard et al, (1988) have argued that self-predictor measures are more closely correlated to the subsequent behaviour. More specifically in the relevant aforementioned metanalysis, measures of self-predictions were found to have stronger correlation to behaviour than did behavioural intentions. It is therefore recommended that emphasis should be placed on the predictions one makes about themselves rather than their motivation (e.g. If one of your friends developed schizophrenia, how likely is it that you would go and see him/her at the home?)

Only 35% of the reviewed studies included control groups to assess the impact of the intervention (Economou et al., 2014; Economou et al., 2012; Holtz & Tessman, 2007; Paulus et al., 2009; Pinto-Foltz et al., 2011; Pitre et al., 2007; Ranson & Byrne, 2014; Rickwood et al., 2004; Rillotta & Nettelbeck, 2007; Skre et al., 2013; Spagnolo, Murphy, & Librera, 2008; Staniland & Byrne, 2013; Ventieri et al., 2011). All of the studies which utilised randomised control trials (RCTs) did not report important information regarding the randomisation methods thereby avoiding assessment as to whether or not all the necessary measures were taken to avoid selection bias. Furthermore, no information was provided regarding whether or not the control group continued with their usual activities or another activity while the intervention was implemented on experimental group. The majority of the RCTs had individuals as the unit of analysis (Holtz & Tessman, 2007; Ranson & Byrne, 2014; Rickwood et al., 2004; Rillotta & Nettelbeck, 2007). Whilst inevitably participants in the experimental and control groups were peers, the reviewed studies failed to take into account the spill-over effect of the intervention on the control group caused by mixing/communicating outside of the class room. If an intervention aims to change attitude, social behaviour, or transmit knowledge, it is wise to consider how interaction can be avoided between the control and the experimental group (Hutchison & Styles, 2010). Staniland & Byrne's (2013) was the only study designed to account for the spill over effect before the intervention was implemented; same grade classes being either allocated to intervention activities or control conditions with older grade students being used as the control group.

The majority of the interventions also did not utilise a waiting-list control group (excluding Naylor et al., 2009; Sakkelari, 2014). However, withholding a beneficial intervention from the control group may raise ethical concerns.

Furthermore, only 11 studies evaluated the longitudinal effectiveness of the interventions even though the basic objective of the interventions was for long term attitudinal changes of the participants. However, only a few of the interventions assessed if the initial changes maintained or decreased through the subsequent time, relative to the controls. Follow up data were mostly collected 3 to 6 months post intervention (Ranson & Byrne, 2014; Pinfold et al., 2003) with only one study examining the sustainability of the effects one year later (Economou et al., 2012). Rillotta's & Nettelbeck's (2007) study assessed the attitudes of past secondary students 8 years after they had participated in an awareness programme about intellectual disability. Although their attitudes in comparison to age-matched participants who did not receive training were significantly favourable, the absence of baseline measures meant that no meaningful comparison could be made. The convenience sampling strategy used also weakened the study.

The interventions themselves were not directly assessed by the participants, who were not directly asked about their opinion on the intervention and its impact. Questionnaires and interviews may be warranted in order for the participants to appraise the interventions and to point out changes, if any, which could potentially optimise their effectiveness

(Weinholtz, Kacer, & Rocklin, 1995). This may be even more pertinent to studies that appeared to have been unsuccessful in ameliorating stigma interviews (e.g. Swaim et al. (2001) and Staniland & Byrne (2013)). Interviews with the participants could have helped evaluate the weaknesses and the strengths of the programmes in order for them to be modified for further use.

Finally, it should be noted that only one study assessed the wider impact of an anti-stigma intervention on the participants. Kohlbauer et al. (2010) assessed the effects of an anti-stigma intervention about depression on the participants' attitudes towards individuals with schizophrenia. Thus, it moved beyond the evaluation of the ephemeral and narrow effects of an intervention and used a design in order assess if contact effects from a primary outgroup of target generalize to other secondary outgroup members (Pettigrew, 2009). A design assessing the wider effects of an intervention could exemplify the wider benefits of an intervention for behaviourally defined conditions.

3.5. Discussion

The current systematic review aimed to uncover the effective components for anti-stigma behaviourally defined condition interventions that guarantee optimal outcomes. This review found no specific element or combination of components to be associated with successful outcomes; studies featuring education and direct contact with stigmatised individuals having been both unsuccessful (Pinfold et al., 2003; Pintzo- Foltz et al., 2011; Paulus et al., 2009) and successful (Spagnolo, Murphy, & Librera, 2008) in eliminating stigmatising attitudes. Similarly, indirect contact in combination with education resulted in either improvement in measures or unsuccessful outcomes. The same inconsistencies are noted in interventions that do not use any form of contact with a stigmatised individual in order to eliminate unfavourable attitudes. It is therefore difficult to draw conclusions on which intervention elements were related to positive outcomes. No evident association could be made between the duration of the intervention and the transient and longitudinal effects on the audience. In other words, the intervention length appeared to be unrelated to the effectiveness of the programs.

In relation to the context of the interventions, with the exception of the studies of Robertson’s (2002), Essler, (2006) and Pitre et al. (2007) information about the target condition was delivered using a traditional approach of a lecture and persuasive message with no other stimulus provided for students to develop a spirit of curiosity or even empathy towards the target population. Although it has been clearly demonstrated that stereotyping and discrimination is perpetuated by a lack of knowledge, it is questionable if providing information alone is an adequate means of eliminating prejudice (Haghighat, 2001). This puts into question campaigns reliant solely on educational components which target children and adolescents as a means of overriding prejudice. Rather, both motivation to change, and ability to comprehend the arguments are needed in order for the message to be successful in altering attitudes (Petty, Heesacker, & Hughes, 1997). The interventions administered in the reviewed studies seemed to assume that the students were sufficiently motivated and interested in the arguments presented to them; the importance of a stimulus to engage the audience in the persuasion process and facilitate their motivation to demolish any pre-existing beliefs being lost.

More active learning approaches have been identified which engage students in the topic. For example, role playing has proved to be a very powerful way of strengthening empathy and enhancing learning experientially (DeNeve & Heppner, 1997). Only few of the studies used segments of popular educational films as tools to engage students’ interest and enhance the learning experience, yet films have been found to enable the viewer to enter a state of absorption and concentration, providing an opportunity for social learning and identification with a protagonist who is from a different background to them (Cape, 2003). Furthermore, Immordino-Yang, et al. (2009) demonstrated that when a pupil has an emotional reaction to new stimuli, lessons presented by that stimuli are more likely to be remembered over time. Ritterfeld & Jin, (2006) has examined the efficacy of entertainment education in order to promote accurate knowledge about schizophrenia and tackle stigmatising views, finding that a movie which depicts an individual with schizophrenia in an empathic manner, in combination with educational information can mediate stigma reduction. Nevertheless, the influence of films in ameliorating stigmatisation for adolescents and children within school settings has not been assessed.

Variables that could influence the outcomes are the characteristics and diverse needs of the audience. However, the audiences' characteristics were not analysed before the development of the interventions and in most cases their demographic characteristics (e.g. ethnicity, gender, and previous contact) were not examined to determine how these factors may have impacted the effectiveness of the interventions. It also appears that none of the reviewed studies involved any degree of tailoring or modification to address the students' cultures and attitudes. A clear, comprehensive understanding of the audiences' attitudes towards the target condition could help develop both the type and content of intervention. In these studies, though, more generic approaches were applied which did not take into consideration the cultural factors of the audience.

In relation to the methodology used by the reviewed studies it can be argued that the studies needed to employ more stringent criteria to determine if a cause-effect relationship existed between the intervention and the outcomes. The aims of the interventions were very narrow, mainly assessing the short-term and ephemeral impact of the interventions. Future studies need to assess the longitudinal effects of the intervention with aims of the interventions interlinked not only to creating an inclusive school environment but also developing adults who do not stigmatise in the future. The fact that waiting-list control groups were not used in the majority of the study designs also emphasised the artificiality of the studies and their cursory aims. There was also a paucity of studies assessing the overall effects of an intervention in changing attitudes towards stigmatising conditions in general. Despite the fact that most interventions placed onus on a specific condition associated with strong prejudice, the ultimate goal of every intervention is for participants to overcome prejudicial attitudes in general and accept a more inclusive environment. However, the studies that were reviewed have missed the opportunity for measuring the potential benefits that the intervention could have on tackling prejudice associated to other conditions with Kohlbauer et al. (2010) being the only study to evaluate “secondary transfer effects”.

Finally, it is clearly indicated that qualitative or mixed-methods research would allow more in-depth information about the participants’ experiences to be collected, especially in exploratory studies. Through examining the roles and functions of different intervention components, more efficient interventions could be designed.

In relation to the context of the interventions the findings further highlight that there is a paucity of interventions that focus on developmental conditions. The majority of the interventions that were reviewed tackle the stigma of mental illness and mostly focused on schizophrenia. Yet schizophrenia is not a common condition amongst school age children. On the contrary, it is extremely rare to be diagnosed before the age of 10 and although it exhibits a steady increase during adolescence (the prevalence amongst children in community settings assessed as 1 to 10 in 10000 (Thomsen, 1996), the mean age of onset for men and women is early adulthood (Hollis & Rapoport, 2011; Thomsen, 1996). By focusing on less common disorders among school age children, myths and misconceptions that students may have about more common disorders like ADHD are not being addressed and dispelled. Whilst interventions about schizophrenia may help develop more tolerant adults in the future, it is questionable whether or not they contribute to creating a more inclusive environment within the classroom for children with developmental condition.

More importantly this review also highlighted how different approaches to interventions worked better with different types of audiences. Clearly a ‘one size fits all’ approach is neither effective nor appropriate for the diverse needs of different school aged children and adolescents in reducing stigma. Rather, future research should focus on the population under study with emphasis given to how the interventions may be customised to their specific needs. These studies need to take into consideration both the local and broader societal influences on attitudes and develop tailor made interventions.

3.6. Limitations and implications for future research

This study's findings should be considered in light of its limitations. This review did not include grey literature which could have resulted in publication bias since studies with statistically significant results are more likely to be published than studies with non-significant or unfavourable results. It is therefore possible that related studies which have not been published due to unclear or insignificant results may have altered the present findings. It is also important to note that all of the studies included in this review were carried out in high and middle-income countries. It is questionable whether the findings may apply to low-income countries, where stigma towards mental illness and developmental conditions might be particularly strong.

A further limitation of this review is the necessarily reductive nature of MMAT, used to assess the methodological quality of the studies. The MMAT quality assessment tool was used as it is the best available measure for assessing quality of studies across a range of methodologies, which was necessary, given the heterogeneity of research approaches included this systematic review. However, it is unable to capture studies' nuances in order to give comparable nuances to all the reviewed study designs.

3.7. Summary and conclusions

Notwithstanding the above limitations of this study, the results provide information that will help professionals design and develop interventions that could potentially ameliorate stigmatisation. More innovative approaches to reducing stigma for behaviourally defined conditions should be developed that take into consideration the developmental stage of the participants. There is also a need to strengthen the evidence-base of the programs that are currently available by increasing the study sample size, including more diverse samples and including short-and long-term follow-ups where possible. More RCTs in combination with qualitative data on particular intervention components should be conducted in order to assess the effectiveness of different components of each intervention. Finally, with an increasing need to deliver cost-effective and easily delivered programs, it is important that cost and feasibility analyses are included in future evaluations of anti-stigma interventions.

The aim of the wider study was to design an anti-stigma intervention that aims to ameliorate discrimination for adolescents with TS within the classroom. Through this systematic review it was made clear that there are no hard and fast rules for designing and implementing an intervention. The key to an effective intervention may lay in understanding the characteristics of the audience and tailoring it to its needs.

Table 4 - Studies reviewed in the systematic review.

Authors Names and Year of Publication	Location	Conditions Targeted in the Intervention	Study Sample	Intervention-Description	Design	Aims	Main Measures used	Follow up	Main Findings
Campbell, 2007	U.S. A	Autism	233 students M age= 13.07. Four conditions Condition 1 boy with autism no information n = 68, Condition 2 descriptive information about boy with autism n =63 Condition3 explanatory information about boy with autism n=42 Condition 4 descriptive and explanatory information n =60.	3 interventions 1) Descriptive information about a boy with autism which does not mention the causes of his behaviour but describes similarities between him and the audience 2) explanatory information about boy with autism 3)combination of 1 and 2	Experimental design	To evaluate the effectiveness of the 3 interventions on middle school children's attitudes. To evaluate if gender and prior knowledge could be associated with children's attitudes.	Knowledge of Autism Adjective Checklist (ACL) Shared Activities Questionnaire (Campbell, 2005) Similarity Ratings Form Perceived (Campbell, 2005) Responsibility Questionnaire (Campbell, 2005)	n/a	- Females and students with prior awareness of autism reported more positive attitudes regardless of the intervention -Explanatory information resulted in more favourable conative attitudes than descriptive information
Campbell, Ferguson, Herzinger, Jackson, & Marino, 2004	U.S. A	Autism	576 students 7 -10 years old 2 experimental conditions Condition 1 no autism explanatory information- autism descriptive information n=283 Condition 2 no autism explanatory information-	2 interventions 1) Short Video with descriptive information about boy with autism (less than 2 minutes) 2) Short video descriptive and explanatory	Within and between group design		The Adjective Checklist (Siperstein, 1980), Shared Activities Questionnaire Similarity Rating Form	n/a	- Descriptive and explanatory information improved overall intentions to engage with children with ASD

			autism descriptive and explanatory information n=293	information about with autism (less than 2 minutes)					- Girls were more responsive than boys
Chan et al., 2009	Hong Kong	Schizophrenia	255 students from 3 secondary schools 13-18 years old (88= education, 94= education+ video, 73 =video +education)	Three conditions: 1) education -30 min demythologizing lecture 2) education-contact video 15 min “The same but not the Same” 3) video-education	Experimental design	Test the effectiveness of three interventions in improving knowledge, attitude, social distance towards individuals with schizophrenia	The Public Stigma Scale (Mak & Leung, 2008) Social Distance Scale- (Schulze, 2003) Knowledge about Schizophrenia, Familiarity with Schizophrenia	1 month follow up	The intervention 2) Education + contact video resulted in less stigmatising attitudes, social distance, increase in knowledge about schizophrenia. - The results sustained in the 1 month follow up.
Del Casale et al., 2013	Italy	Different types of mental disorders	44 students (16-18 years old)	4 sessions with didactic and interactive component (one and half an hour each) visual aids (slides, movie scenes) were used.	Quasi experimental design	Reduce stigma towards individuals with mental disorders	Haghighat’s Standardised Stigmatization Questionnaire (SSQ) (2005)	n/a	- Significant reduction of mental illness related stigma
DeSocio et al., 2006	U.S. A	Different types of health disorders	370 elementary and middle school children 10-12 years old	6 sessions that lasted 45 minutes each. Didactic and interactive components	Within group design (pre-test, post-test)	To evaluate the effectiveness of the programme in raising awareness about mental health disorders	16 -item questionnaire specially designed for the study that evaluated the knowledge about mental disorders	n/a	- Improvement in knowledge

Economou et al., 2012	Greece	Schizophrenia	580 students 13-15 years old n= 281 control group, n=299 intervention group, 11 schools.	2 hour semi structured programme led by an educational psychologist and a psychiatrist. Combating existing stereotypes. Didactic interactive component.	Randomised cluster control trial.	To evaluate the impact of the intervention on the students ‘attitudes and preferred social distance towards people with schizophrenia. To evaluate if the results could be sustained after 12 months.	Questionnaire derived from the Alberta Pilot Questionnaire Toolkit and adjusted to high school students	12-month follow-up	- The information provided improved the students’ attitudes, beliefs and desired social distance - The longitudinal results suggest that the desired social distance lessened with time.
Economou et al., 2014	Greece	Schizophrenia	1081 students, 13-15 years old (intervention group=551, control group=530) From 20 schools	2 hour semi structured programme (didactic and interactive components Creative activities i.e. poster painting	Randomised cluster control trial. Mixed method design. Post measures taken 2 weeks after the intervention. Control group received 2 hour lecture on vaccination	To evaluate the effectiveness of the intervention in reducing stigmatising beliefs and attitudes and decreasing preferred social distance levels towards individuals with schizophrenia	Questionnaire derived from the Alberta Pilot Questionnaire Toolkit and adjusted to high school students Qualitative Data (emotions, colours, words)	n/a	- Statistical differences were found in attitude beliefs and preferred social distance after the intervention.

Essler et al., 2006	U. K	Different types of mental disorders	104 students (13-14 years old)	Two phases. Games drama and quiz led by a theatre company	Within group design (pre-test, post-test) Post-test was taken one month after the intervention	Increase knowledge about mental health and reduce stigmatising attitudes	Mindout for mental health quiz (DoH, 2001)	n/a	- Increased knowledge and improved attitudes towards individuals with mental health disorders
Friedrich et al., 1996	U.S.A	Tourette's syndrome	153 students (8-11 years old) TS (n=49), individuals without TS (n=53) and individuals with TS and information (=51)	100 seconds educational video of an individual with Tourette explaining his condition	Between group design- 3 groups a) TS b) no TS c) TS and information	To evaluate if a brief educational intervention delivered by the individual with TS will impact children's attitudes and behavioural intentions	Adjective Checklist (Siperstein, 1980) Activity Preference Scale (Siperstein & Bak, 1977) Foley Questionnaire (Foley, 1979), Modified Foley Questionnaire	n/a	- Youths rated children with TS less favourably than individuals without tics. No distinction was made in behavioural intentions between the child with TS and the child without. -No effects on intervention.
Holtz & Tessman, 2007	U.S. A	Tourette's syndrome	179 (7-15 years old) elementary school children from 6 schools. n = 91 Intervention Group n= 45 control group	10 minutes video You've got a Friend video Control	Randomised control trial	To evaluate the impact on the intervention on knowledge about TS, attitude and behavioural intentions towards individuals with TS	The knowledge questionnaire- specifically designed for the study (Modified CATCH Rosenbaum et al., 1986), Foley Questionnaire (Foley, 1979)	n/a	The measures taken at the pre-intervention stage revealed that individuals with TS were rated more positively after an psychoeducational intervention

Kohlbauer et al., 2010	Germany	Depression	264 students (14-20 years old)	Two sessions-didactic and interactive components Direct contact	Within group design	Evaluate if an education focusing on depression changes the attitudes towards patients with depression as well as schizophrenia	Questionnaire designed for the study Emotional reaction towards individuals with schizophrenia (Angermeyer, 1997), Preferred social distance (Link et al. 2004) Endorsement of stereotypical characterisation (specifically derived for the study)	3-4 months follow-up	- Attitudes towards depression and stereotypical endorsements were improved after the intervention. - These results could not be sustained at the follow up. - Reduction of preferred social distance and fear was observed towards people with schizophrenia
Martínez-Zambrano et al., 2013	Spain	Different types of mental disorders	62 students (14-16 years old)	2 intervention sessions carried out over 2 weeks 1) didactic & interactive components 2) direct contact with individuals with mental disorders	Within Group design (pre-test, post-test)	To evaluate the effectiveness of the intervention in reducing stigma and to assess the variable of gender and prior knowledge of someone with a mental health disorder	Opinions on Mental Illness (OMI) (Cohen 1962)- subscales authoritarianism, benevolence-interpersonal aetiology scale-restrictiveness scale, negativism scale.	n/a	- Reduction on stigmatisation especially in the authoritarianism. - No changes were observed in the subscale of benevolence - The intervention had slightly greater impact on woman than it did on men

Meise et al., 2000; Sulzenbacher et al., 2002	Germany	Schizophrenia	114 students – 16-19 years old 57 psychiatrist contact- 57 only education)	Two conditions: condition 1 educational program from psychiatrist and contact with individuals with schizophrenia Condition 2 educational intervention by psychiatrist and social worker	Quasi experimental design. Qualitative study.	Evaluate the impact of the intervention in knowledge about schizophrenia endorsement of stereotypical characterisation and preferred social distance	Knowledge about schizophrenia questionnaire- designed for the study Emotional reaction towards individuals with schizophrenia Angermeyer, 1997) Endorsement of stereotypical characterisation Voges & Rossler, 1995) Preferred Social Distance Link et al. 1987) Qualitative Data Free text Writing for both groups both before and after the intervention.	n/a	- Significant decrease in the preferred social distance and more favourable emotional reactions were only observed in the group that came in contact with an individual with schizophrenia - More stigmatising views were observed in the group that received the intervention from a psychiatrist and a social worker - Better understanding of the core symptoms of schizophrenia in both groups
Morton & Campbell, 2008	U.S. A	Autism	296 children 8 -12 years old videotape=66 Classroom Teacher =58 hypothetical mother= 57 hypothetical father =55 hypothetical doctor= 60)	Descriptive and explanatory information about a child with autism shown in a short video was	Experimental design.	To evaluate if the information source affects attitudes towards autism	Adjective Checklist Siperstein & Bak, 1977), Shared Activities Questionnaire Short form	n/a	- Fifth graders had more favourable attitudes towards a child with autism when the information was

				provided by different sources			(Morgan et al., 1996)		provided by the doctor - Third graders had more favourable attitudes when the information was provided by the mother
Naylor et al., 2009	U.K.	Stress, depression, suicide/self-harm, eating disorders, bullying & intellectual disabilities	356 students 14-15 years old Intervention group =149, Control group= 207	6 sessions of 50 minute lessons (discussion, role playing, internet searching, video and films). Programme delivered by trained tutored.	Non-randomised pre-test – post-test control group design with a waiting list control group. Post-test administered 6 months after the intervention.	To evaluate the effectiveness in	Mental Health Questionnaire (MHQ)–specially designed for the study. Strengths and Difficulties Questionnaire (SDQ, Goodman, 1997) Questionnaire assessing the intervention	n/a	Improvement in knowledge and empathy
Nussey, Pistrang, & Murphy, 2013	U. K	Tourette’s syndrome	100 primary students that were in the same class with an individual with TS	A half an hour slideshow developed by ‘Tourettes Action delivered by children with Tourette’s syndrome	Mixed method design	To evaluate the effectiveness in knowledge, attitude about TS	Modified form of the CATCH, Interviews questionnaire	n/a	Improvement in knowledge and attitudes towards TS

Paulus et al., 2009	Germany	Different types of mental disorders	210 students 13 -18 years old from 4 schools (120 = intervention group, 90=control group)	"Crazy so what?" 5 days programme Education and contact	A quasi-experimental , longitudinal control study	Evaluate the effects of the programme in reduce stigmatising attitudes towards individuals with mental illness, increasing knowledge about help seeking strategies, students' competence in dealing with a mental illness. To identify variables that could be associated with prejudicial attitudes.	Self-efficacy scale (Schwarzer & Jerusalem, 1998) Help seeking scale (Wilson, 2005) Social distance Questionnaire (Schulze, 2003) Feedback and evaluation of school project	3 months follow up	- Short term effects on the preferred social distance towards individuals with mental disorders - The changes were not sustained in the follow up study - Boys had more stigmatising views than girls - Students with higher level of education had a lower preferred social distance - Help seeking behaviour increased
Pinfold et al., 2003	U.K	Different types of mental disorders	472 students (14-15 years old)	2-hour educational intervention workshop and Contact with individual with mental disorder.	Within group design (pre-test, post-test, follow up data) Mixed method design.	Evaluate the impact of intervention on knowledge, attitude and preferred social distance towards individuals with mental disorders.	World Psychiatric Association Questionnaire- Qualitative Data from descriptions of people with mental disorders.	6 months follow up	- Increase in knowledge about mental disorders. - Preferred social distance did not change significantly. - Changes were mostly marked for females attending grammar schools

						Identify variables that could be correlated to more stigmatising attitudes			and those reporting personal contact. - Attending grammar schools and personal contact was associated to higher baseline attitudes.
Pinto-Foltz et al., 2011	U.S. A	Different types of mental disorders	156 female students from 2 schools (13-17 years old) 95= intervention group, 61= control group	In our Own Voice (60-90 minutes) didactic & interactive components and direct contact with two consumers	Non-blinded randomised cluster control trial	To evaluate the effectiveness of the intervention in reducing stigma and increasing knowledge about mental health disorders	Revised Attribution Questionnaire by Watson et al (2004)	3-4 weeks follow up 4-8 weeks follow up	The intervention was not successful in reducing mental illness stigma. An increase in mental health literacy was observed in the in the 3-4, 4-8 week follow up.
Pitre et al., 2007	Canada	Schizophrenia, dementia and depression/anxiety	144 students (8-12 years old) from 5 schools. Does not mention how many participants were in either group.	Hand Puppet Program. 3 plays x 45 minutes. Puppets portrayed people with a mental illness	Cluster randomised control trial	To evaluate the effectiveness of the intervention in reducing stigma	Opinions about Mental Health Scale (OMI) adapted to age appropriate language. Benevolence Separatism, Stereotyping, Restrictiveness Pessimistic Prediction Stigmatisation.	n/a	Improvement in the experimental group in separatism, restrictiveness, stigmatization

Ranson & Byrne, 2014	Australia	Females with high functioning Autism	273 females- 12 – 15 years old (46= intervention condition; 66=no intervention peer condition; 283= no intervention no peer condition)	"Understanding our Peers" 8 weekly 50 minutes sessions Contact education online activities	Randomised Control Trial- two control groups	To evaluate the effects of the intervention in improving knowledge, attitudes and behavioural intentions towards females with High Functioning Autism.	AKQ-R, ACL, SAQ-R, SRF-R, PRQ-R, SIQ-R, SFS	Follow up after 3 months	- Better Knowledge Better attitudes marginally better behavioural intentions in the experimental group - Spill over effects - The results did were maintained until the end of the following school term
Rickwood et al., 2004	Australia	Different types of mental disorders	457 students (14-18 years old) 309=experimental group 148=control group	Mental Illness Education programme (MIE-ACT)- one session programme (50-90 minutes) Contact, discussion role-play, other interactive activities	Solomon four group design	Increase knowledge and help seeking intention, reduce stigma	Stigma Questionnaire- specifically designed for the study, Knowledge Questionnaire- Specifically designed for the study- General Intentions to Seek Help Questionnaire	n/a	- Increase knowledge in mental disorders - Moderate impact on stigma - Weak impact on help seeking intentions
Rillotta & Nettelbeck, 2007	Australia	Intellectual disability	259 past and present students (secondary students that has received a 3-week session or 8-week session ADP= 67 Past students that had participated in the 10 session ADP course 8	Awareness of Disability Programmes (ADPs)	Quasi experimental ; age matched control groups	To evaluate the effectiveness of the programmes in promoting inclusion	Attitude toward Persons with an Intellectual Disability Questionnaire	8 years follow up by accessing the views of individuals who	- Students that received the 8 session ADP programme had better attitudes than students with the 3 session ADP programme

			years ago= 46, same age group without training=146					participated in the programme 8 years before	- Long term benefits of ADP programme
Roberts et al., 2007	U.K.	Psychosis	1367 students baseline measures 1034 post questionnaires 14-22 years old from 29 schools	Interactive drama-based education “On the Edge”. Four hours overlapping weeks	Within group design (pre-test, post-test). Mixed-method design.	Increase knowledge, reduce stigma, increase help seeking behaviour	Knowledge questionnaire, stigma and attitude questionnaire Semi-structured interviews (85 students) to evaluate the programme	6 months follow up 313 students from 11 schools and 42 interviews.	Increased knowledge Reduced stigma Improved awareness for help seeking behaviour
Ruble et al., 2013	U.S. A	Depression	710 students 14-15 years old from 6 schools, 447 intervention group, 263 control group	Adolescent Depression Awareness Program (ADAP) curriculum based on the medical model implemented by psychiatrists or psychiatric nurses	Non-randomised cluster control trial	Evaluate the impact of the intervention on knowledge and attitudes towards depression and help-seeking behaviour.	Adolescent Depression Knowledge Questionnaire (ADKQ)	n/a	Positive change in knowledge attitude and help-seeking behaviour.
Saeker et al., 2010	U.S. A	ADHD	64 high school students (15-18 years old). 32 students experimental	Intervention Group :7 minutes and 33 second video of	Experimental design	Evaluate the effectiveness of different interventions.	Modified KADDS (Knowledge of Attention Deficit Disorder)	n/a	The experimental group did not differ in behavioural

			group, 32 intervention group	an individual with ADHD describing his personal experiences (PDEs) with ADHD and dispelling the 12 most common myths about ADHD Control Group: Providing only factual info			Modified BIS (Behavioural Attention Scale)		intentions. PDEs did not enhance learning of non-DPE facts.
Sakellari et al., 2011	Greece	Different types of mental health illnesses (bipolar mood disorder, depression, schizophrenia)	59 students (13-16 years old) Intervention group (n= 28), control group (n=31)	2-hour intervention implemented by the main researcher. Lecture and discussion (30 min)	Randomised cluster control trial with a waiting list control group. Qualitative	The perception of mental illness among adolescents. To evaluate the effectiveness of the programme	Individual interviews with open-ended questions before and after the intervention	n/a	Small changes occurred as a result of the intervention. attitudes towards the individuals with mental illness as far as therapy and dangerousness
Schulze et al., 2003	Germany	Schizophrenia	150 students (14-18 years old) 90 students experimental group, 60 students control group	“Crazy? So, what!”- 5-day programme – education and direct contact-artwork, games, discussions.	Quasi experimental design. Evaluation one month after the intervention	To evaluate the effectiveness of the intervention in decreasing preferred social distance and negative stereotypes about schizophrenia.	Specially designed questionnaire assessing stereotypes and preferred social distance	n/a	- Ceiling effects. - Attitude improvement, reduction of stereotypes and preferred social distance decreased after the intervention

						To evaluate if educational levels and social economic class would be variables that impact attitudes.			(not statistically significant)
Silton & Fogel, 2012	U.S.A	Autism	158 students 9-13 years old (Video I=44 Video II=39 Video III=31 Video IV=43)	Video I) Descriptive and explanatory information Video (4min. 42 sec) II) Descriptive explanatory information and peer strategies (22 min 42 sec) Video III) Descriptive information explanatory information and strengths information (22 min 42 sec) Video IV) Descriptive information, explanatory peer strategies and strengths information (22 min 42 sec)	Experimental design	To evaluate if peer strategies and strengths information can enhance an intervention aimed at improving behavioural attitudes towards individuals with autism	Shared Activities Questionnaire (Morgan, 1998), Adjective Checklist (Siperstein & Bak, 1980)	n/a	- Students had greater behavioural intentions after the intervention but no attitude differences were observed - Participants who viewed the peer strategies video had significantly better behavioural intentions than those who did not

Skre et al., 2013	Norway	Depression, anxiety, eating disorders and schizophrenia	1070 students, 12-17 years old n=520, experimental group. n= 550 control group) from 3 schools in Norway.	3-day intervention "Mental health for everyone" Delivered by the teacher. Individual tasks, group tasks, plenary sessions, illustrating video material	Non-randomised cluster control trial. Post-test measures were collected 2 months after the intervention.	Evaluate the effects of the intervention on improving adolescents' knowledge about mental health problems, eliminating stigma and identifying where one could seek mental health support. Evaluate if variables like age and gender could be correlated to attitudes.	Knowledge about mental disorders questionnaire, Prejudice beliefs Knowledge about where to seek help for mental health problems	n/a	- Knowledge increased substantially for anxiety, depression and schizophrenia - Moderately for anorexia - Younger students and boys held more prejudice beliefs than older students and girls. - Individuals with more prejudice beliefs did not mention any mental health services in which they could seek help
Spagnolo et al., 2008	U.S. A	Different types of mental disorders	426 students (pre-and post-intervention = 277 students, only post intervention= 149 to rule out testing effect	Recovery from a Serious Mental illness is Possible- 60-90-minute presentation (Education and contact through the narrative of the consumers)	Within group design (pre-test, post-test)	To evaluate the programme effectiveness in reducing blameworthy and stigmatising attitudes towards individuals with mental disorders	Attribution Questionnaire – Short form for Children (Watson et al., 2004)	n/a	Floor effects in blameworthy attitudes. Less stigmatising attitudes after the intervention.

Staniland & Byrne, 2013	Australia	Males with high functioning Autism	395 boys (48= intervention condition; 56=no intervention peer condition; 169= no intervention no peer condition)	“Understanding our Peers” 6 weeks – 50 minutes sessions Contact Education Online activities	Randomised Control Trial- two control groups	To evaluate the effects of the intervention in improving knowledge, attitudes and behavioural intentions towards boys with High Functioning Autism	AKQ, ACL, SAQ, SRF-R, PRQ-R, SIQ	Follow up after 3 months	- Better knowledge, better attitudes, no improvement in behavioural intentions in the experimental group - No spill over effects - The results sustained until the end of the following school term - More knowledge in students who engaged in online activities
Stuart, 2006	Canada	Schizophrenia	571 students (13 or younger-18 or older) from 8 high schools	“Reaching out” 2 lessons - indirect contact through video depictions of 5 individuals who have schizophrenia. Role playing.	Within group design (pre-test, post-test)	To evaluate the impact of the intervention on adolescents’ knowledge, social distance and to identify variables that could be correlated to more stigmatising views	World Psychiatric Association’s global anti- stigma program (knowledge about schizophrenia, social distance, demographic information)	n/a	- Ceiling effects in the knowledge questionnaire - Lack of correlation between levels of knowledge and preferred social distance - Small attitude change - The impact of the program differed depending on the

									age group and the gender
Swaim & Morgan, 2001	U.S. A	Autism	233 students 8-9years old and 11-12 years old (condition 1=87, condition 2 =77, condition 3= 78).	3 conditions 1) No autism video (1 min 15 sec) 2) Autism/no information video (1 minute 15 sec) 3) Autism/ descriptive and explanatory information (2 minute 15 sec)	Experimental design	To evaluate if providing information about autism would affect attitudes and behavioural intentions towards a peer with autism	Shared Activities Questionnaire Adjective Checklist	n/a	- The information provided had no effect on children's attitude towards the child with autistic symptoms - No differences were observed between the ratings of the participants in their behavioural intentions towards the child with autism and the one without.
Swartz et al. 2010	U.S. A	Depression	3538 students (14-15 years old) from 6 high schools	Adolescent Depression Awareness Program (ADAP) 3-hour curriculum based on the medical model implemented by psychiatrists or psychiatric nurses	Within group design (pre-test, post-test) post-test 6 weeks after the intervention	Increase students' knowledge about depression Attitudes towards depression treatment	Adolescent Depression ADKQ specifically designed by the author	n/a	- Increase in knowledge Improvement in attitudes towards depression

Ventieri, Clarke & Hay 2011	Australia	Different types of mental illnesses	195 students 9-12 years old. Intervention group n= 69. control group= 126)	2 sessions (90 min and 75 minutes). Didactic & interactive components playing, games and activities	Non-randomised cluster control trial	Test the effectiveness of the intervention in improving Knowledge social distance & attitudes towards individuals with mental illness	Attitude measures- specifically designed for the study Social distance Scale (Schulze 2003) Knowledge measure- specifically designed for the study	4 months follow up	Improvement in knowledge, attitude and reduction in preferred social distance towards people with mental disorders. Results maintained after four months above baseline
Watson et al. 2004	U.S. A	Different types of health disorders	1566 students 11-14 years old	Science of Mental Illness 5-6 sessions of 45 minutes- Interactive components- web based activities. Mainly based on the medical model. Implemented by the teacher	Within group design (pre-test, post-test design)	To evaluate the effectiveness of the programme in changing middle school children's attitude and knowledge about mental disorders	The knowledge Test (curriculum evaluation) Attribution Questionnaire adapted for children	n/a	Significant improvement in both attitude and knowledge Most effective in improving attitudes of individuals that initially had more negative baseline attitudes.

Table 5 - MMAT Scores for studies included in the review.

Articles	**MMAT scores**
(Campbell, 2007)	50%
(Campbell, Ferguson, Herzinger, Jackson, & Marino, 2004)	50%
(Chan et al., 2009)	25%
(Del Casale et al., 2013)	50%
(DeSocio et al., 2006)	50%
(Economou et al., 2012)	75%
(Economou et al., 2014)	75%
(Essler et al., 2006)	50%
(Friedrich et al., 1996b)	50%
(Holtz & Tessman, 2007c)	50%
(Kohlbauer et al., 2010)	75%
(Martínez-Zambrano et al., 2013b)	75%
(Meise et al., 2000)	75%
Sulzenbacher et al., 2002)	75%
(Morton & Campbell, 2008)	50%
(Naylor et al., 2009)	75%
(Nussey, Pistrang, & Murphy, 2013)	50%
(Paulus et al., 2009c)	75%
(Pinfold et al., 2003b)	25%
(Pinto-Foltz et al., 2011)	50%
(Pitre et al., 2007)	25%
(Ranson & Byrne, 2014a)	50%
(Rickwood et al., 2004)	75%

(Rillotta & Nettelbeck, 2007)	50%
(Roberts et al., 2007)	25%
(Ruble et al., 2013)	75%
Saeker et al. 2010	75%
(Sakellari et al., 2011)	75%
(Schulze et al., 2003)	100%
(Silton & Fogel, 2012)	75%
(Skre et al., 2013b)	100%
(Spagnolo et al., 2008)	50%
(Staniland & Byrne, 2013)	50%
(Stuart, 2006)	75%
(Swaim & Morgan, 2001)	50%
Swartz et al. (2010)	50%
Ventieri, Clarke & Hay (2011)	50%
Watson et al. (2004)	75%

4

Methodology

The overall methodology of the thesis was dictated by the research questions which are how typically developing adolescents perceive individuals with TS and whether a tailor made school-based intervention can ameliorate stigmatisation. In this short chapter, the overall methodology of the thesis with be discussed. In subsequent chapters (Chapter 5 and Chapter 7), each study's methodology and methods will be discussed in detail.

4.1. Exploratory sequential mixed-method design

In this short chapter, the overall methodology of the thesis with be discussed. In subsequent chapters (Chapter 5 and Chapter 7), each study's methodology will be discussed in detail.

The overall aims of the thesis were to understand how tic-free adolescents conceptualise Tourette's syndrome; to design an intervention to ameliorate stigmatisation for the condition based on the aforementioned data; to develop new questionnaires that assess knowledge, attitudes and behavioural intentions based on user-defined criteria and finally, to evaluate the effectiveness of the intervention. The objective of the thesis dictated that a two-phase sequential mixed model research design be implemented in which the qualitative component precedes the quantitative element. Phase One was essentially an exploratory qualitative study the findings of which provided the basis for the development of an anti-stigma intervention and user-defined questionnaires. The effectiveness of the intervention was tested in the second phase of the study by utilising a convergent mixed method design in which the qualitative and quantitative results were collected

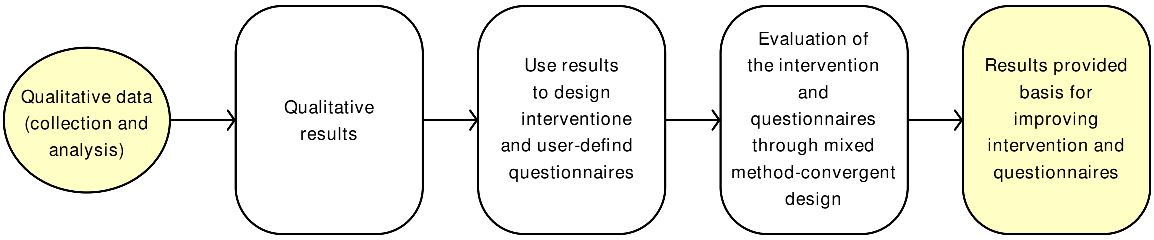

Figure 2 - The research design of the thesis.

simultaneously. The results of the second part of the study provided the basis for improving the intervention (Figure 3, p.111).

As is evident from Figure 3 data collection did not occur in a parallel manner. Rather, one methodological strand of the research was completed and analysed at a time. Thus, qualitative data provided a basis for collection of the quantitative data. Qualitative data were collected in March of 2015 for developmental purposes, in order to capture typically developing adolescents' perceptions of their peers with Tourette's syndrome. These findings then guided the development of the intervention, and subsequent scales for a quantitative survey instrument. This enabled a tailor-made intervention as well as user-defined measures for the aforementioned population. This process may be described as a sequential mixed-method design according to Greene (1989).

Exploratory sequential mixed method design is a common practice in intervention research, in order to gain qualitative information before the intervention, to shape the intervention, and to develop appropriate instruments. Qualitative data allows for a phenomenon to be explored, and generate a second, quantitative phase that tests the interventions' effectiveness and the questionnaires' validity and reliability. This two-step approach (qualitative followed by quantitative) is especially advantageous when the phenomenon under study is under- researched, and when one of the aims is to develop a new measure. In this specific project, grounded theory was utilised in order to develop a novel theory about a poorly understood phenomenon such as attitudes and opinions about TS. The theory was subsequently used to develop an intervention which was followed up by a mixed-method study that tests the interventions effectiveness. The later study was conducted between April and July of 2016.

Mixed-method research has emerged as a viable third methodological movement of research and it aims to integrate qualitative and quantitative research into one study, in order to develop a comprehensive understanding of a complex human phenomenon (Creswell, 2013). This approach argues that the epistemological and ontological positions that qualitative and quantitative paradigms are based on (positivism and constructivism respectively) are not irreconcilable and incompatible. The positivist worldview posits that there is an independent reality, and the researcher can obtain an objective stance towards the phenomenon being studied. It is also based on deductive reasoning (top-down logic), by which the researcher deduces a hypothesis that must be empirically tested. On the contrary, from a constructivist point of view no tangible reality exists. The qualitative researcher also acknowledges that because of the researchers pre-existing ideas and bias objectivity cannot be achieved. Theoretical ideas are also formed through an inductive process “bottom up" by which specific observations help formulate and develop some general conclusions or theories.

It has been vehemently argued that combining these two different ontological and epistemological paradigms is not possible (Guba, Lincoln, Denzin, & Lincoln, 1998). However, mixed-method research maintains that the distinction between “complete objectivity’’ and ‘‘complete subjectivity’’ this is a pseudo-dichotomy when faced with human-beings and real world-research (Morgan, 2007). The supremacy of positivist over constructivist is refuted, and qualitative and quantitate research are viewed as an “interactive continuum” (Newman & Benz, 1998). A mixed-method approach is founded on the paradigm of pragmatism. Pragmatism places the onus on the research question rather than conforming to a methodological orthodoxy and is based on the principle that the investigators are able to maintain subjectivity in their own reflections on research, as well as objectivity in data collection and analysis (Teddlie & Tashakkori, 2003). It is therefore based on methodological eclecticism and on the notion that the outcomes are more important than the process (Creswell & Plano Clark, 2007). Methodological eclecticism asserts that there is no clear link between a specific paradigm and a specific methodology. It is also based on abductive reasoning which is what Morgan (2007) refers to as the complementary and constant waving between inductive and deductive theoretical development rather than a reliance on one or the other. It is also founded on intersubjectivity, the acceptance that a single "realworld" exists but simultaneously all humans have their own unique interpretations of that world. Finally, pragmatism is grounded on the assumption that all data can potentially be transferrable. Thus, not all quantitative data can be generalised to every possible historical or cultural setting nor can all qualitative data only be relevant to specific setting and not applicable beyond that setting.

Mixed-methodology was utilized in the current research to build upon the strengths of both quantitative and qualitative paradigms and to provide the most complete and comprehensive answers to the research question. Sequential mixed method design may make it easier to retain the integrity of each method and to evaluate each method according to method-specific quality criteria, because one study is completed before the other begins. Despite the aforementioned advantages, Creswell and Plano Clark (2007) have emphasised that, mixed methods research can be more strenuous and a substantial length of time to complete both data collection phases may be required. In addition, mixed methods research also requires researchers to develop a broader set of skills that span both the quantitative and qualitative aspects. Furthermore, it may be time consuming, since the first data of the study would have to be analysed before proceeding to the second phase of the study.

In this short chapter it is explained why a sequential mixed- methods approach was chosen for this research. The method of each of the two studies is discussed separately in the subsequent chapters. In the methodology section of Chapter 5 the reasons as to why qualitative research was chosen are discussed as well as why grounded theory was considered appropriate. Similarly, in the study design section of the second study (Chapter 7) of the thesis it is explained why concurrent mixed method design was appropriate in order to evaluate the effectiveness of the intervention.

5

Study One: Adolescents' Perception of Their Peers with Tourette's Syndrome: An Exploratory Study[*]

5.1. Introduction

In this chapter the empirical testing of the knowledge adolescents had about TS and the feelings they developed towards their peers with TS is reported. This qualitative study was deemed necessary since as shown in previous chapters insufficient data existed that clarified why individuals with TS might be excluded from their peer group. This data was imperative in order to develop a pertinent intervention that would address adolescents' concerns about their peers with Tourette's syndrome.

In the previous chapters of the thesis it was demonstrated that youths with TS experience impaired peer relations and are at a significantly higher risk of stigmatisation and victimisation compared to their typically developing peers. Social maladjustment may be due to the low social acceptability of the tics and children and adolescents with TS may be confronted with negative attitudes from their peers in school.

Previous literature has given considerably less attention to understanding adolescents' awareness and attitudes towards their peers with TS and research has been primarily directed towards young children and higher education students (aged 18 years old) (Woods, Fuqua, & Outman, 1999; Woods, 2002; Woods & Marcks, 2005). Although the

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[*] A slightly altered version of this chapter has been published as Malli, M. A., & Forrester-Jones, R. (2017). "I'm not being rude, I'd want somebody normal": Adolescents' Perception of their Peers with Tourette's syndrome: an Exploratory Study. Journal of developmental and physical disabilities, 29(2), 279-305.

intensity and frequency of the tics usually decline during puberty, according to the clinical perspective, adolescence is the period in which individuals with TS experience their symptoms as most challenging and when exclusionary practices become more prominent (Sukhodolsky, Eicher, & Leckman, 2013). Due to the negative evaluation and stigma associated with TS, individuals with the condition may have limited chances to develop social adaptive skills and supportive social relationships, both poignant developmental challenges for adolescence (Sullivan, 2013). Therefore, the focus of research should be placed on this specific age-group.

Another limitation relating to previous studies of attitudes towards Tourette's syndrome is that they have been predominantly based on narrow questionnaires that solely investigate social acceptability and overt prejudice and not social participation and friendships. Furthermore, possible explanations and motives about the causation of social exclusion have not been provided. The aforementioned measures have also failed to uncover underlying biases that are more likely to occur in today's society where overt discrimination is considered morally reprehensible and the tendency to suppress inappropriate forms of prejudice is more pronounced (Bergsieker, Leslie, Constantine, & Fiske, 2012). Finally, only a limited number of studies have examined the knowledge base typically developing peers have about TS and the possible impact that this knowledge might have on their subsequent behaviour. Therefore, a nuanced and substantive understanding of how typically developing adolescents perceive their peers with TS that discerns their views on forming relationships with them is lacking in the literature.

Interestingly, studies to date suggest that stigma towards individuals with TS is robust and resilient to change and there have been a number of unsuccessful interventions (Friedrich, Morgan, & Devine, 1996; Woods & Marcks, 2005). Therefore, more interventions need to be developed that are not solely based on theoretical principles but explicitly address the concerns of specific subgroups. A theory about typically developing adolescents' perception towards peers with Tourette's syndrome could help identify modifiable causal factors associated with attitudes and support the development of more tailor- made interventions in order to positively change the referent age-groups' knowledge, attitudes and behavioural intentions.

In an attempt to address the gaps in the literature the current study aimed to explore how adolescents with TS are perceived by their peers from a qualitative stance in order to provide a richer understanding of the phenomenon. More specifically the study aims were as follows:

1. To investigate typically developing adolescents' knowledge about TS, expressed in their own words;
2. To explore the perception and feelings of tic-free adolescents towards individuals who do have TS utilizing a range of qualitative methods;
3. To assess typical students' intent to interact with individuals with TS and to provide a comprehensive understanding of their behavioural intentions.

The overall objective of the study was to gain insight into how the condition is conceptualised by adolescents who do not have Tourette's syndrome, and to provide further data on this under-studied subject of how adolescents with TS are perceived by their typically developing peers. Considering all the aforementioned points, the current study would be the first that aims to provide an understanding of this social phenomenon.

It was also exemplified in Chapter 3 that currently, no validated self-reported measures that specifically pertain to neurotypical adolescents' knowledge and attitudes towards individuals with TS exist. Although there are a variety of measures that assess societal attitudes towards individuals with behaviourally defined conditions, these do not take into consideration the unique developmental period of adolescence, and the particular context and culture in which they interact. Therefore, a secondary objective of the study was to utilise the qualitative data as a recourse for the design of quantitative questionnaires that assess adolescents' knowledge, beliefs and behavioural intentions towards peers with TS. Measures were therefore developed utilizing user-defined criteria in order to produce culturally appropriate questionnaires (See Study Two chapter 7, Methodology Section).

5.2. Methodology

The research question dictated the usage of qualitative methodology. This approach which is rooted in social constructivism asserts that there is no single unitary reality that 'is out there'. Rather people construct their own meaning and truth through interaction with the world around them. Furthermore, the emphasis is not placed in determining cause and effect of a social phenomenon but in meaning making and the participants' interpretation of their own reality and the world around them (e.g. how would having an individual with TS in their classroom affect its dynamic) (Merriam, 2009). Through an inductive approach qualitative researchers aim to develop and generate a theory from the participants' subjective viewpoint.

Qualitative research has been criticised because it is time consuming, and its conclusions are difficult to generalise beyond the research setting and participants. However, it should be argued that qualitative methods enable us to better understand social realities and provide rich explanations for complex phenomena, as is this one.

5.2.1. Setting and participants

Difficulties gaining access to schools and recruiting adolescents as research participants were initially encountered. More than 30 secondary schools and head teachers were contacted by phone and emails by the investigator. The majority did not respond and some that initially expressed an interest in the project declined due to other research commitments.

The researcher's supervisor and her professional connection to a head teacher were instrumental in gaining access to a comprehensive secondary school in South East England. This enabled the researcher and her supervisor to arrange a meeting with the key individual for obtaining approval. During this appointment, a synopsis of the study was presented and the objectives, the aims, the significance, the timeframe, and the inclusion criteria of the project were highlighted. Approval was granted.

Pupils from the school were of low attainment and more than one quarter were recorded as having English as an Additional Language (EAL). There were 33 languages spoken altogether including Polish, Czeska, and Lithuanian with Slovak and Latvian being the most common first languages. The town was socially and economically deprived and characterised by high unemployment rates thus, above the regional and national averages.

The study's inclusion criteria stipulated that the participants had to be English speaking pupils between the ages of fourteen to seventeen. Although English did not have to be their first language it was imperative for the study that they could speak English fluently, in order to provide meaningful qualitative data. Due to the fact that the school had so many pupils with limited English skills, the participants for the study were handpicked from various classes in Year Ten (14-15 years old) by the head teacher.

Participants included twenty-two year-Ten pupils (17 females and 5 males). Regarding the participants' first language 81.8% reported it being English, 4.5% Latvian, 4.5% Slovak, 4.5% Turkish and 4.5% Russian. Two participants (9.1%) reported having a relative with TS, half of the sample had previously watched a show on TV that included a person with Tourette's syndrome, and 9 (40.9%) had never observed a person with Tourette's syndrome.

5.2.2. Ethical considerations

The study was reviewed and given an ethically favourable opinion by the University of Kent Ethics Committee (February 2015, see appendix V). Initially, an active consent was considered appropriate for obtaining parental approval to conduct research with minors. It has been suggested that this approach, in which the parent or guardian is required to return a signed consent form in order for the child to take part in the study, is the gold standard rule in enrolling participants (Esbensen et al., 1996). It has been characterised as the favourable ethical option because it entails the participants' autonomous decision.

The head of the school however pointed out that an opt-out approach would be the most appropriate way to gain the sample required within the context of a very diverse and socio-economically deprived catchment area. Indeed, it has been argued that the opt-in recruitment method inclines to elicit high percentages of participants from families with high levels of parental education and lower rates of respondents from ethnic minorities and therefore produce sample bias (Dent et al., 1993; Esbensen, Melde, Taylor, & Peterson, 2008). In the 'opt-out' system all the parents of the potential participants are informed about the study through the distribution of information sheets and consent is assumed for every child by default unless the parents indicate their refusal (Junghans, Feder, Hemingway, Timmis, & Jones, 2005). Nevertheless, the researcher was initially hesitant to use the 'opt-out' system which uses passive consent instead of active consent since it is acknowledged as a controversial approach to recruiting research participants in school settings. Indeed due to the reduced stringency of the consent requirements it cannot be established with any confidence that parents have made a well-informed decision not to raise objections if no opt-out form is submitted. However, through her subsequent research of the relevant literature the researcher discovered that the opt-out system is more commonly used within the context of a school. In favour of this approach, it might be argued that in school contexts parental consent is at least supplemented by the consent of the head teacher who acts in ‘loco parentis’. That extra ethical safety net might justify weakening the requirements for parental consent at least in those cases that might otherwise be considered appropriate for standing consent. Ellickson & Hawes (1989) also demonstrated that 87% of non-respondents to a passive consent process had received all the necessary information and had consciously chosen for their children to take part. This form of recruiting participants is also justifiable when the potential risk of participation is low and there is no likelihood of actual harm. Furthermore, it helps recruit a representative population without violating the participants’ right of choice (Lacy et al., 2012) (Appendix A-E).

Whilst it was imperative for the study to adhere to school policy, the researcher also needed to ensure that the new recruitment strategy was approved by the ethics committee before the research could take place. Therefore the researcher and her first supervisor requested for Chair’s Action in order to approve the amendments pertaining to the methodology used to recruit participants. A request for a quick decision was made to the Chair since since the researcher was informed about the schools opt-out policy only a week before the pilot study was due to take place. This chain of events highlighted that schools are not under the exclusive control of the researcher.

Following the ethics amendments, an ‘opt-out’ recruitment approach was utilised in order to gain parental/ guardian consent and written assent was sought from the participants (see appendix W).

5.3. Data collection

Focus groups and free text writing were combined to achieve a nuanced and enhanced understanding of the social phenomenon under investigation. Free text writing provided a concrete individual-level data set that was not skewed from or influenced by the norm of the peer group (Murphy & Dingwall, 2003). On the other hand, focus groups provided the chance to elicit the shared understanding and meaning of the social phenomenon (Kitzinger, 1994). Thus, by combining attitudes expressed privately, with those articulated within group discussions a more comprehensive understanding of the structure of the phenomenon was more likely to emerge (Lambert & Loiselle, 2008). One-to-one interviews with the participants were considered for this study in order to allow a greater rapport with the interviewees. However, they were limited in terms of gathering data about how the condition is conceptualised within a peer group setting. They could not provide the privacy of free-text writing which was chosen as a method to gather data which could minimise social desirability bias. The combination of these two techniques was chosen to provide an idea of how an individual with the condition would be conceptualised and treated in the context of his peer group but also by each of his peers individually.

5.3.1. Free text writing

All participants were given ten minutes to express their thoughts and feelings in relation to individuals with TS through free text writing. Instructions were written as follows:

> *"We want to know what you think about people with Tourette's syndrome.* Please remember that no one else apart from the researchers, will see what you have written about, so feel free to express your thoughts and feelings.
>
> *What do you think about people with Tourette's syndrome? How do they make you feel?"*

Free text responses were deemed appropriate for the current research since this method can elicit rich data by providing the respondents with the opportunity to express their thoughts in their own words, using their own vocabulary. Participants were encouraged to write as much or as little as they wished; express themselves in their own words; and were also discouraged from evaluating the quality of their text in terms of writing skills such as spelling, punctuation and neatness so as not to detract from the focus of thinking about TS. Similar to questionnaires free text writing can also elicit thoughts that participants might not be willing to disclose in a face-to face interview. Responses may also identify issues and perspectives that might not have been covered in more forced choice questionnaires (Garcia, Evans, & Reshaw, 2004).

5.3.2. Focus groups

Each focus group consisted of four to six participants. These clusters were arranged by the teacher based on close friendship networks between the participants. It has been argued that discussions conducted between members of a friendship group hold very close resemblance to naturally occurring dialogues (Kitzinger, 1994). These settings might also give some “hesitant” interviewees the impetus to participate (Puchta & Potter, 2004). In contrast to individual interviews youths are more likely to disclose personal thoughts and attitudes within a group of peers than they are to an adult interviewer to whom they most probably will express opinions and views that comply with the social norm (Krumpal, 2013).

The interview guide was developed from gaps about attitudes towards individuals with TS identified through the literature review. Data from the free text writing also determined pertinent questions that were further explored in the group context (See appendix F).

During the focus groups participants were also introduced to two vignettes in which the protagonist, a typically developing adolescent, was faced with a moral dilemma about the social inclusion or exclusion of a peer with TS in two different social contexts (See appendix F). The use of the vignettes was twofold: i) to elicit the participants’ judgement regarding the protagonists' decision and ii) to facilitate discussion about the participants' moral reasoning when making their decisions about peer group exclusion and inclusion (Horn, Killen, & Stangor, 1999). The intention of the first story was to understand if rejection is considered permissible because an individual with TS does not assimilate to the norms of a group. The second vignette was utilised in order to access if exclusion was justified as a result of personal choice.

5.4. Procedure

A pilot with four students outside of the study sample was conducted after a favourable ethical opinion had been granted and one week before the main study. Since no significant changes were made to the research instruments the pilot data was included in the study's main results. This is common practice in qualitative studies since consecutive interviews help the researcher progressively gain more insight and focus in the interview schedule (Cutler, Murphy, Gilmour, & Heyman, 2009; Holloway, 1997).

The study was undertaken during the students' regular school timetable. Participants were gathered into a large school room during their regular 40-minute classroom period. First a four-minute video was used to introduce the participants to the symptomology of Tourette's syndrome. The video which was obtained from the public domain depicted a Caucasian adolescent girl, of approximately the same age as the participants, who exhibited TS symptomology, including simple and complex tics both motor and phonic in the form of repetitive eye-blinking throat-clearing sounds, grunting, sniffing, coughing, head jerking, neck stretching and facial grimacing. The individual was viewed from the waist up while sitting in a chair in front of a desk that was against a neutral background. The girl briefly introduced herself and mentioned that the purpose of creating the video was to demonstrate the transient exacerbation of tics while she was engaged with homework (see https://www.youtube.com/watch?v=O6j8YM1CcPU). The accurateness of the symptomology displayed was assessed by four parents of children with TS who belonged to a TS support association outside the main study and its validity was also academically peer reviewed for the purpose of this study. Each of the groups concurred that it accurately reflected TS behaviour. This video overcame the limitations of the videos used in previous studies (Boudjouk, Woods, Miltenberger, & Long, 2000; Friedrich et al., 1996) in which only motor tics were exhibited as the basic component of TS. Furthermore, by using a video of an actual individual with TS as opposed to an actor simulating tics, the external validity of the study was enhanced. Ethically, the use of a video instead of an in vivo adolescent with TS ensured a child with TS was not identified or singled out during the research.

After viewing the short stimulus video participants were asked to complete a demographic and free text writing measure. The pupils were closely monitored by the researcher and a school teacher in order to prevent any sharing of information in regards to their responses and to ensure that all of the measures had been filled in. During the following week, the focus groups were facilitated by the researcher during three consecutive 40 minute classroom periods within the school premises. A school staff member was sitting at the back of the room in each of the sessions.

In order to establish rapport with the participants the researcher had an extensive discussion with them during a 40 minutes classroom period immediately after the free-text writing measure was completed. The researcher talked about herself and the reasons for doing her study. The interviewees also asked about her background and why she had chosen to do the study in the UK and not her homeland, Greece. Individuals that were non-UK and had recently moved to the country disclosed their experiences of moving to a new country. Also, interviewees enthusiastically talked about their future dreams and goals in life. This extended discussion in which most of the interviewees actively participated appeared to enable them to feel more comfortable in the presence of the researcher, foster trust as well as feel less judged and believe interviewer is genuinely interested in what they had to say.

5.5. Data analysis

The data obtained from the free-text writing and focus group were collated in order to identify a comprehensive model for the phenomenon under study. Grounded theory (Corbin & Strauss, 2008; Glaser & Strauss, 1967) guided the data analysis which was deemed appropriate due to its suitability to explore subjects that have been relatively under studied (Goulding, 2002). Furthermore, it was the intention of the study to move beyond a thick description of the social phenomenon to the development of an explanatory theoretical model grounded in the data. It should be highlighted that although grounded theory analysis was used, the study is not a "pure" grounded theory study. Thus, all the canons and procedures that pure grounded theory dictates were not carried out. Indeed, all of the focus groups were carried out on the same day due to the schools restricted timetable. Inevitably, it was not possible to analyse the data and use the concepts derived from the analysis to form the basis for the subsequent data collection. Thus, data collection and analysis did not continue an ongoing cycle throughout the research process. Furthermore, it was not possible to use theoretical sampling, as the number of participants had to be predetermined (Corbin & Strauss, 2008).

The transcripts and free text writing were initially rigorously read multiple times by both the researcher and her first supervisor who independently coded and categorised the data. Patterns of categories were subjected to a process of 'constant comparison' between the codes and the categories in order to adjust and refine them. Categories incorporated several codes under a more conceptual heading. During the next stage, the categories were explored further and higher order categories (or sub-themes) were identified that moved beyond description to interpretation. Elongated discussions with the first supervisor comparing categories and sub-themes followed until saturation and also to ensure thematic-reliability. This process yielded one overarching theme, three sub-themes and seven categories.

Causal conditions, contexts, and behavioural outcomes relating to the studied phenomenon were uncovered in order to identify a model of how individuals with TS are perceived by their typically developing peers. Memos pertaining to hypotheses about the relationship between the categories and sub-themes were used to facilitate a more theoretical understanding of the data. This was aided by diagrammatic representations of possible relationships between the categories and sub-themes constructed in a systematic and rigorous way (Figure 4, p.129). NVivo 10, a software tool for managing qualitative research data was used to facilitate the grounded theory analysis (Hutchison, Johnston, & Breckon, 2010).

Throughout the research process and the analysis, the researcher undertook reflexive research practice. It has been well established that reflexivity in qualitative research enhances the reliability of the study, and the subjective nature of qualitative research is no longer perceived as a barrier but an opportunity (Finlay, 2002). Personal reflexivity refers to the process by which the researchers self-reflect on the ways their personal views, attitudes, experiences, and values may have shaped and affected the research process and subsequently the creation of knowledge (Willig, 2013). Experience in the field of education as a special educational teacher and personally witnessing the marginalisation and social exclusion of individuals with TS by their typically developing peers may have affected the interview schedule in the focus groups and the analysis. Thus, although the researcher tried to remain as objective as possible the lens of her experiences may have been used in making meaning of the participants narratives. The questions in the focus groups however, were as open-ended as possible in order for the research to remain exploratory in nature and allow the participants to introduce new perspectives on the subject. Furthermore, the researcher was aware that the participants who were adolescents may have been less willing to share their views and experiences with the researcher who might have been perceived as an authority figure. Efforts to establish an anti-authoritarian relationship between researcher and researched was made through gaining their trust by spending time with them before the interviews and talking about herself.

5.6. Results

The overarching theme to emerge from the data was “Straying from the boundaries of normalcy” as illustrated in Figure 4. The below figure demonstrates a conceptual framework for understanding the process by which the participants cognitively conceptualise Tourette’s syndrome, their subsequent affective responses and their behavioural intentions towards their peers with TS. Three key sub-themes were identified as being involved in the process: ‘Assembling notions of Tourette’s syndrome’; ‘Socially constructing TS; and ‘Establishing boundaries’. Participants appeared to construct their notion about TS by processing popular

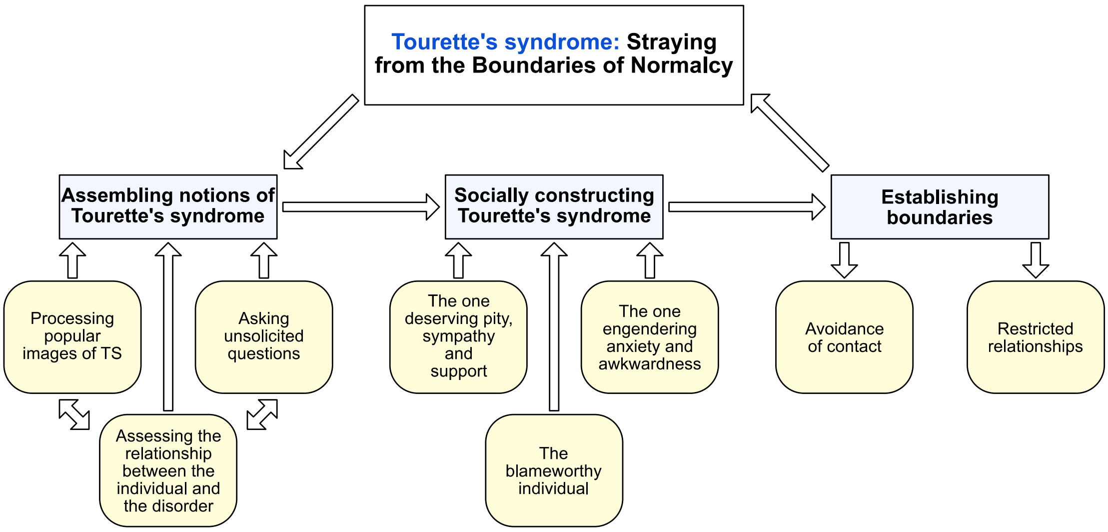

Figure 3 - Process by which typically developing adolescents perceived their peers with Tourette's syndrome.

cultural images of TS, assessing the responsibility the individual had over the condition and asking questions about the syndrome. This social construction of TS led to a perceived social identity which the adolescents projected onto a person with TS. Most participants identified individuals with TS as deserving pity and sympathy whilst others viewed them as responsible for their condition. Individuals with TS were also perceived as people that would make typically developing adolescents feel anxious and awkward. This socially constructed social identity of people with TS which was bound up in pejorative ideas about the condition defined the subsequent behaviour of the participants towards their peers with TS; establishing boundaries by either avoiding contact or creating restricted relationships with their peers with TS.

The themes and subthemes are described below accompanied with substantive anonymous quotes from participants which have been unaltered by the authors to maintain participant authenticity.

5.6.1. Assembling notions of Tourette’s syndrome

The participants appeared to have largely constructed their own perceptions about the nature of TS through processing images that had been presented in popular culture. In turn, the adolescents made assumptions about the controllability and responsibility individuals had over their symptomology. Adolescents also expressed a keen interest in understanding the condition and familiarising themselves with individuals with TS, indicating that the amount of information they had previously received had been insufficient.

5.6.1.1. Processing popular images of Tourette’s syndrome

Participants reported that they had experienced relatively little contact with individuals with TS although the majority were aware of the syndrome through images on the TV and the internet. Erroneous beliefs prevailed among them pertaining to the core feature of the condition. Both documentaries and movies that the participants recalled focused on coprolalia, the uncontrollable utterance of obscenities and profanities (American Psychiatric Association, 2013). In the movies they mentioned such as “Not Another Teen Movie” and documentaries series such as the “Undatables” individuals with TS were reduced to stereotypes for the sake of comedy or, according to the adolescents ‘losers’ who needed help. Conversely, other relevant aspects of the syndrome were omitted. This is exemplified in the following quotes:

> *PL3: I didn’t know that Tourette’s also moved your body, I thought it was only* swearing but now I take it as a serious syndrome. (Free text writing)

> *TP3: [Talking about Not Another Teen movie] Can’t remember what it was* called but there was like this cheerleader and she'd get mad and shout out these swear words (Focus group 3).

The above quotes show how participants were exposed to a media image of TS in which individuals with the condition were rendered as more different than similar to typically developing individuals. By viewing the most extreme cases, TS was conceptualised as a cursing disease that perpetuated the notion of social misfit and “otherness”. Indeed, uncontrollable swearing was identified as the main barrier that segregated individuals with TS, impeded social interaction and ultimately distinguished them from their peers to the extent of ghettoizing individuals as indicated below during a discussion about dating:

> *TP4:…So it might be easier for them [people with TS] to date somebody with TS* (Focus group 3)

5.6.1.2. Assessing the relationship between the individual and the condition

The vast majority of the participants reported that individuals with TS had no control over their symptomology and were therefore not responsible for their condition. This highlights the importance of inferences of personal responsibility to a condition in determining the observer’s understanding and emotional response to it (Weiner, 1986). The tics were perceived to stem from within the body and people with TS were unable to exert control over their physical movements and discourse:

> TP12: They can't control it, because this comes from inside the body (Focus Group 2).

> *TP7: They're not really to blame. It’s just like something happens to them and* they can't get rid of it (Focus Group 4).

Simultaneously, it was implied that Tourette’s syndrome was something external to the autonomous self that violated the person’s agency. Thus, the person’s involuntary actions were imposed on them by the omnipresent syndrome and the real individual was trapped in an uncontrollable body. The profoundly disempowered real self was dominated by the "illness" that prevailed over the passive physical and social agency of the individual:

> TP6: They can't go anywhere without having tics, they can't control it like that (Focus group 4).
>
> *TP14: It's like it forces or something, forces her to react like that (Free* text writing).

A deprivation of agency implies weakness, immaturity or even danger. Indeed, some participants maintained that the uncontrollability and randomness of the tics may inevitably create danger. This led to a feeling that physical distance had to be maintained in the interest of self-preservation.

> *TP2: And it would be like scary 'cause you don't know what gonna happen. I* will be wondering what they are going to be doing next (Focus group 3).

Only a few of the respondents were sceptical about the uncontrollability of the symptomology; insinuating that the tics were exaggerated. They implied that the symptoms of TS could be alleviated if the actors put enough effort and concentration into the specific task:

> PL3: I feel like they're putting it on (Focus group 1).
>
> TP14: it looks like you can control it, if like you put like effort, but I don't know (Focus group 4)

5.6.1.3. Asking unsolicited questions

Participants expressed a desire to understand the complex nature of Tourette's syndrome and the subsequent effects it may have on the inflicted individual. Although the participants' interest in Tourette's may have merely been sparked by the study itself and therefore have reflected the Hawthorne effect (McCambridge, Witton, & Elbourne, 2014), these queries were initiated by the young people who took part in the study unprompted by the researcher or their teacher. Questions were posed pertaining to the aetiological causes of TS, the genetic predisposition associated to the condition, the environmental factors that might influence the tics, the course of syndrome and more specifically if the symptomology of the condition diminishes with age. Adolescents were also curious about the supressibility of the tics and the level of the individual's control over them. They also expressed an interest in finding out the coping mechanisms of individuals with TS. This set of questions might suggest that despite the participants' exposure to the exaggerated media portrayals of TS they still have unanswered questions and a desire to familiarise themselves with pertinent information and individuals with TS as indicated below:

> PL3: Is Tourette's genetic? (Focus group 1)
>
> TP15: I would like to know if people are born with it or can it be genetic and they get it later on in life? (Free text writing)
>
> TP3: I would like to know how she copes with it and keeps herself calm through it? (Free text writing)
>
> TP6: Can you control it to a certain extent? Was you born with it? Will it go? (Free text writing)

5.6.2. Socially constructing Tourette's syndrome

Based on the limited information about TS available to the pupils and the affective reaction towards the TS population, the adolescents in the study attempted to socially categorise individuals with TS (Turner, Hogg, Oakes, Reicher, & Wetherell, 1987). The social identity of individuals with TS was socially constructed through a process of social comparison to what the participants understood as "normal". The perceived differentiating characteristics between the participants and their peers with TS were accentuated and professed as important and salient. Through this procedure people with TS were viewed as “others” and perceived as ‘out-group’ members that possessed characteristics that the adolescents in the study acknowledged as derogatory. Most participants were conflicted between viewing people with TS as a personification of their deficits and subsequently as deserving sympathy and pity and as a source of discomfort. Others perceived them as responsible for their derogation whilst some viewed them as engendering anxiety and awkwardness.

5.6.2.1. The one disserving pity, compassion and support

Adolescents in the study compared their perceived own abilities and future opportunities with those of their peers with TS. The latter were viewed as abject and disadvantaged in relation to an ideal of normalcy held by the pupils. This process which can be interpreted within the downward social comparison framework (Finlay & Lyons, 2000; Wills, 1981) elicited feelings of pity towards individuals with TS and simultaneously enhanced the participants’ own self-esteem. Indeed, participants thoroughly described what they imagined having TS would entail for one’s social standing. A compromised life was delineated in which individuals with TS would endure social isolation, low levels of social connectedness and limited career prospects. Amongst the social difficulties their peers would encounter, teasing and social isolation were the most prominent:

> Moderator: How would you imagine him/her during break or lunch?
>
> TP8: Probably alone. Probably one or maybe two friends.
>
> *TP3: .....Maybe a few people would make a few jokes [within the classroom]. But* then outside probably they might not sit with her and she'll be quite lonely (Focus Group 3).
>
> *TP14: I felt like kind of sorry ‘cause it must be hard for them to do certain things* and have friends and stuff. And it must take a lot more effort than everybody else.

> *’Cause you have to try and* build a relationship with people and it might be a lot harder because of the tics and stuff. (Focus group 4)
>
> *TP12: In this situation for them it’s much harder to find a friend some people don’t want to be their friend because they has that illness [sic] (*Free text writing).

Some participants maintained that the tics and the subsequent burden they entail could impede individuals with TS from achieving high academic performances:

> TP3: I felt a little sorry for her cause she was trying to do her homework, so the tics might have been distracting her, so she might not have been able to do her work the best that she could have done. So, it might affect her grades (Focus group 3)
>
> *PL5: I'd feel sorry for him. Not really sure if they’d drop out of school.*
>
> Moderator: Why would they drop out of school?
>
> PL5: Am just thinking, yeah, could they like concentrate with all the twitches? (Focus group 1)

A considerable number of participants expressed a concern about the potential barriers individuals with Tourette’s may encounter in the future while pursuing and acquiring a job. Conversations were centred on employment discrimination. The tics and ‘socially inappropriate behaviour’ could restrict and limit their career choices and many participants posited that their ‘otherness’ would prohibit them from pursuing positions that would involve public speaking and face-to-face interaction. It was therefore insinuated that jobs that require less public contact would be more appropriate for them:

> *TP6: …… like feels like if he wante*d a job [individual with TS] to speak in front of lots of people, he wouldn't be able to do it, because he wouldn't be able to control it.

> *TP 5: Yeah. In certain jobs, they'll not be able to pursuit their dream ‘cause the* condition would hold you back...or from certain parts of it. Like if you're talking in front of groups. (Focus group 4)

By conceptualising individuals with TS as inferior to themselves, disadvantaged and unaccountable of their misfortune, feelings of pity and sympathy were elicited. Indeed, the phrase “I feel sorry for” or “I feel sad for” was used by eleven of the respondents to describe their emotional response towards an individual with TS.

> *TP10: Also it makes me feel sad as they can’t have a normal life as they want,* they will always have people looking down at them which could lead them to feel bad about themselves (Free text writing)

> TP11: This girl makes me feel sorry for her as she is different from other people (Free text writing)

> *TP3: I feel bad for anyone who has Tourette’s. Perso*nally, I would hang around *them because it’s not their own liking to have such a syndrome (Free text* writing).

Through social downward comparison (Wills, 1981) participants re-evaluated their own social status as well as their groups. They elaborated on positive information about their identity in comparison to the less fortunate group of people with TS and enhanced their self-esteem as seen below:

> TP17: We are lucky that we ain't got Tourette (Free text writing)

> TP 7: I felt sorry for her, but I also felt glad because I didn't have it (Focus group 4)

Pity in some cases manifested itself in the form egocentric sadness that did not entail benevolence. Thus, although the participants appeared to feel compassion for the targeted, individual they were not willing to act in a friendly and approachable manner towards their peers with TS.

> *TP12: I think it's very hard to [sic] that person to live like that. I feel a bit sorry, if somebody repeating the same thing..I wouldn't [want] that person to be in my* class (Free text writing).

However, a number of participants maintained that they would act in an altruistic manner and help their peers in need. Pity implies that the targeted individual deserves protection, caring and providing for (Thompson, Cowan, & Rosenhan, 1980). Some of the participants suggested that they would undertake a custodial role even if it was not being asked of them. This emphasises typically developing adolescents' effort to express socially appropriate and positive attitudes towards peers with TS but simultaneously indicated their conviction that individuals with TS may need to rely on tic-free components for support.

> Moderator: Do you think that he or she is capable of sorting things out by himself or does he need help?
>
> TP8: Cause it kind of like *depends on how bad the tics are. 'Cause if actually [he] gets tics and he's with the group, then yeah he can't really do stuff for* himself (Focus group 3).
>
> TP15: You know, people will be like taking the mick out of them. They are not like normal people; they need more help than us (Focus group 2).

The participants' self-esteem was also elevated by viewing their actions towards their peers with TS as fair and moral. By providing unsolicited help to their peers and not directly discriminating against them, an action which is perceived as unjust, the participants in turn appeared to obtain a positive self-identity for being helpful. PL5 characteristically mentioned:

> *“.... I wouldn't stay out of it, even though he (the individual that’s being bullied)* might get angry at me, I wouldn't. Cause at the end of the day I'd know I've done *something that would change people’s lives. Yeah!”* (Focus Group 1)

5.6.2.2. The blameworthy individual

A limited number of participants considered the target individuals responsible for their tics. Inferences of responsibility elicited negative emotions and diminished adolescents’ willingness to provide social support to the hypothetical peer. In fact, angry responses were more common in the adolescents who insinuated that the symptomology is high on personal control:

> TP9:... I feel like they can't control it but then it seems a little strange that they *can't. I don’t know. They should at least try. The other people try, so why shouldn’t they? Otherwise, just let them be on their own (Focus Group 3*)

This concept could be linked to Weiner’s attribution theory (1986; 1995) that posits that people search for the causation of the behaviours of others. An individual that is perceived to have control over their deviant behaviour and is deemed responsible for their condition may elicit negative responses such as anger from the person they are interacting with. Anger may also influence the observer’s behaviour and higher inferences of responsibility are associated with lower acceptance rates and greater exclusion (Juvonen, 1991).

5.6.2.3. The one engendering anxiety and awkwardness

Throughout the study the participants anticipated that discomfort and awkwardness would stem from a social interaction with an individual with TS. It was suggested that anxiety may originate from the inherently novel situation and the lack of clear rules of conduct as well as the lack of affective readiness on the part of the participants. The unsettling situation in which the normal are uncertain about how to behave around the out-group member has been labelled as "interaction pathology" (Davis, 1961) can be exemplified by the three participants below:

> *TP3: I may be a bit nervous around her because if she does something I wouldn't* know what to do (Free text writing)

> TP16: Honestly, whilst watching the video clip I feel quite anxious because I *wouldn't know what to do when I am around someone with Tourette's syndrome* (Free text writing)

> TP5: It made me feel anxious. Yeah, uneasy (Focus group 4)

The belief that TS may involve unpredictable behaviour is also a factor that may create an anxiety provoking situation (Corrigan et al., 2005). Some participants maintained that the uncontrollability and randomness of the tics might ostensibly create danger. The tics and the unpredictable way they manifest could appear threatening. The perceived lack of control that people with TS had over their tics appeared anxiety provoking for some of the participants:

> *TP12: When I'm seen [sic] person that has Tourette's syndrome I feel very* scared. I know they will not going to do anything but how they acting is scares me [sic] (Free text writing)

Anxiety may also be invoked from conflicting and incompatible feelings that stem from interacting with an individual that is perceived as an outgroup member (Stephan, 2014). Thus, some participants' evaluations contained both positive and negative dimensions. Well intentioned adolescents were motivated by egalitarian values and were concerned about behaving in a non-stigmatising manner towards an individual who they viewed as disadvantaged. Simultaneously, they acknowledged the potential social cost of affiliating with an individual with TS and voiced their negative attitudes in a concealed manner. In other words, the participants tried to maintain a non-prejudiced self-image while expressing negative attitudes and feelings towards the specific outgroup members. Therefore, attitudinal ambivalence was evoked by inconstant and contradictory reactions to individuals with TS which may have resulted in inter-emotion tension (Katz, 1981; Thompson & Zanna, 1995). Thus, two dissonant emotions or cognitions towards a social target may evoke anxiety towards the specific individual.

> TP18: They are still people and they still have feelings and the right to have *friends, even though I reckon sometimes it would get annoying… it would be* extremely distracting and sometimes I may feel uncomfortable. (Free text writing)

This form of cognitive dissonance may create a discomforting experience which in turn might lead to quite extreme and amplified responses (Bell & Esses, 2002). Characteristically, in this study a number of adolescents expressed overly positive intensions towards their peers with TS.

> TP13: I think that it would stay with them forever and really sorry for her. (Free text writing)
>
> PL5: I'd try to make them like best friends (Focus Group 1)

Participants also expressed a concern about the negative social consequences of being closely linked to an individual with TS. More specifically they voiced a fear of "social contamination" (Thornberg, 2015), and were apprehensive about other in-group members' reaction. Some of the participants stated that the social encounters with individuals with TS would preferably take place in settings where the tics were less likely to be conspicuous and where less people would be present. Thus, adolescents wanted to maintain a distance from the hypervisibility imposed on the individual with TS. Public exposure would elicit feelings of discomfort, unwanted attention and embarrassment for all parties involved. More specifically, being directly associated with the person with TS could threaten their social image:

> TP11: I don't know yeah, I wouldn't go to the cinema or the malls...if I had the *choice I would like parks. I think it's the best place, 'cause they would much better if... If you go to the cinema or somewhere the mall yo*u would feel uncomfortable other people around would feel uncomfortable, this person feels. You know what I mean right? In the park, it's much better. And not only in the park but somewhere like where less people (Focus group 2)

Participants anticipated that the devaluation that stems from the “mark” of TS would spread to the people associated with the individual with TS. As a result, they feared that they would also be the subject of curiosity and misunderstanding and subsequently devalued and denigrated by their peers. This phenomenon which Goffman (1963) termed as courtesy-stigma suggests that stigmatisation does not only affect the ‘discredited’ but also impacts those who are associated with the stigmatized individuals. The adolescents in this study acknowledged the social risk of bearing courtesy-stigma and explained the mechanism by which devaluation spreads to other people associated with the targeted individual. They also expressed scepticism towards the likelihood of the stigmatised individual being benefited or even destigmatised as a function of association with a “normal” individual.

> TP14: Like you stand up for them and they're gonna judge you because you're standing up for them.
>
> TP5: Other people don't wanna. So you feel embarrassed to. In fact, you get even more judged than who their actually judging. (Focus group 4)

Some of the participants also feared that by being closely associated to a devalued individual their peers may perceive them as bearing similar derogatory attributes as the stigmatised. In the case of a chosen affiliation, such as a romantic partner, adolescents were concerned that their peers might infer that they too posed an invisible “mark” that would make affiliations with higher status individuals impossible. For instance, participant TP12 stated:

> If I had that boyfriend (with TS) people gonna judge me. Not me, but they're gonna say she's beautiful but he, he is like, has this syndrome. He's not like *(pause). What's wrong with her? You know?" (Focus group 2)*

5.6.3. Establishing boundaries

By perceiving adolescents with TS as outgroup members the participants of the study expected that they would prefer to avoid contact with them or they would only be willing to form restricted relationships. Interestingly, social distance would be dependent on the severity and the disruptiveness of the tics. Thus, the concealability of the tics were conceived as the prime means of retaining social acceptability since less visible symptoms of the condition suggest more ambiguous boundaries between "them" and "us". In other words, by approaching normalcy and eradicating personal differences marginalisation could be attenuated. These findings confirm that the frequency of the tics have effects on how comfortable the observer might feel while interacting with an individual with TS (Woods et al., 1999). The same concept was highlighted when the participants viewed rehabilitation and cure as a means to overcoming the limitations related to the condition:

> Moderator: Could you potentially date somebody with TS?
>
> TP6: It depends.. like you can be with them and after a few years it can get better and they could really stop it. Like stop having Tourette's and just be a normal person (Focus group 4)

5.6.3.1. Avoidance of contact

A limited number of participants asserted that they would preferably maintain a social and physical distance from their peers with Tourette's syndrome. Irritated responses and a propensity to segregate individuals with TS were linked to the adolescents that were sceptical about their peers' inability to exert control over their symptomology. In other words, the hypothetical peer with TS was viewed as an individual that consciously and deliberately refused to assimilate to the norm. However, anger and the intention to avoid individuals with TS was not limited to participants who viewed having TS as a form of derogation. Participants' understanding about the uncontrollability of the syndrome and their sympathy towards their peers did not necessarily positively influence their acceptance or their desire to minimise the social distance between them. Possibly fear, discomfort guilt, and pity may have inhibited them from interacting with individuals with TS:

> *TP7: I think she can't help it and to be honest I wouldn't hang round [sic] with* her when it gets bad I would feel awkward but I would feel sorry (Free text writing)
>
> PL5: Having a person like that in my class would be very annoying and I would not like to sit next to her because she looks very stupid and childish the things she does and the noises she makes are very annoying and it would be very uncomfortable to sit with her or hang around with her (Free text writing).
>
> TP1: So if it's really bad then yeah maybe like he should go like to another school. But umm but if it's not like that bad, that's fine. Then maybe he should be with everyone else (Focus group 4)

5.6.3.2. Restricted relationships

Exclusion, in the form of overt rejection was perceived negatively by most of the participants across the four groups. Consistent with Killen's research (Killen & Stangor, 2001) adolescents evaluated straightforward rejection as morally reprehensible.

> TP 8: Yeah, it's just rude if they tease someone because they have Tourette's (Focus group 3)

However, the majority of the participants differentiated their reciprocated relationships with other typically developing peers from the anticipated affiliations that would be formed with individuals with TS. A form of relationship was delineated which would be lacking in intimacy, companionship and closeness; three of the most important elements that define the quality of a friendship (Sullivan, 2013):

> *PL3: No, I wouldn't mind to hang out with a person with Tourette's, but I would want my space as well. ...I would want not every day hang out, no*t every day be *together, not every day talk. Don't want him to cling on to me. Yeah*
>
> Moderator: So, do you do that with your other friends?
>
> PL3: Not really
>
> PL1: So, he/she would be like a secondary friend?
>
> *PL3: He wouldn't be at the same level, the same le*vel as all my other friends. (Focus group 1)

Participants would provide support for their peer with TS, assist them if necessary, advocate on their behalf if their rights were being threatened and defend them if they were disrespected. However, they would not actively socialise with them, thus describing a friendship that is based on benevolence and not on reciprocity. Their feelings therefore appeared to stem from a sense of duty and morality.

> TP2: I would hang around with them but not like everywhere. I would need my space to hang around with my other friends as well that are more like me. They *wouldn't be like my best friends, the one I call when things get bad like to help* me. How could they? Right? (Focus group 3)

Greater hesitancy towards engaging in relationships with individuals with TS became more apparent as the relationships deepened. As it was also acknowledged, in order for a romantic relationship to be formed, intimacy must be created between the two partners. However, in the case of individuals with TS participants claimed that closeness might be harder to achieve and pity, sympathy and a tendency towards over-solicitousness could be perceived as a potential barrier to genuine attraction.

> TP 14: (on dating) It depends on how comfortable I am around him, cause If I feel like guilty and sorry for him, want to help him then I don't think I can be in a relationship with him (Focus group 4)
>
> PL3: I'm being honest yeah. I'm not being rude, I'd want to have somebody *normal. I'm not saying they're not no*rmal (individuals with TS). But you know what I mean. I would mostly feel so sorry [for] him (Focus group 1)

Adolescents perpetuated superficial relationships that were fuelled by a parenting attitude and the participants' need to provide protection and social comfort to the disadvantaged "other". Affiliations were perceived as a charitable and altruistic act that made the actor feel superior.

5.7. Discussion

Previous studies which have examined typically developing youths' attitudes towards individuals with TS have failed to explore the context of individual's perceptions and the reasons why these views may develop. The current study compliments and expands upon the relevant literature by broadening our understanding about adolescents' beliefs and perceptions about TS and their subsequent affective and behavioural reactions.

The grounded theory analysis provided an overarching theoretical framework for understanding the process by which individuals with TS are perceived by their typically developing peers. Misconceptions about TS which were mostly perpetuated by stereotypical media images prevailed among adolescents' understanding about the condition. Participants who constructed TS as a condition beyond the individuals control perceived their peers as being deprived of agency and strength and as straying from the boundaries of normalcy. The wide gap that was formed between "them" and "us" was accompanied by downwards social comparison (Finlay & Lyons, 2000; Wills, 1981). Individuals with TS were viewed as in need of assistance and a form of patronising help was offered. This kind of 'benevolence stigma' (Cohen & Struening, 1962; Corrigan, Edwards, Green, Diwan, & Penn, 2001) in combination with the fear of being 'socially contaminated' inhibited the initiation of meaningful social relationships with individuals with TS. A close degree of contact was also impeded due to intergroup anxiety and a form of cognitive dissonance. On the other hand, the limited number of participants that held individuals with TS responsible for their symptomology expressed a plenary desire for social distance. However, decidedly negative behavioural intentions were not solely limited to adolescents who elicited inferences of responsibility to people with TS indicating that attributional models of stigmatisation may be of secondary importance in the case of TS.

This theory suggests that it is likely that youths with TS would be peripheral to social networks in comparison to typically developing adolescents. Although the latter reported that peers with TS would mostly be socially accepted, the quality of the friendships that they would be willing to engage in would be different from the ones they would establish with their neurotypical peers. Previous research suggests that it is the quality of reciprocated friendships, rather than the number of relationships or the presence or absence of certain types of affiliations that can be associated with social adjustment and social skills, internalised distress and, ultimately, the quality of life of individuals with TS (Sullivan, 2013). This study also highlights that stigma and discrimination do not necessarily manifest themselves in the form of overt antipathy and that prejudiced attitudes revealed can depend on the level of intimacy in question. As opposed to active rejection an individual with TS might endure a form of prejudice which can appear in the form of patronisation or neglect (Cuddy, Fiske, & Glick, 2007).

The imperative need for an educational component in an intervention for typically developing adolescents was exemplified by the participants' skewed and inadequate knowledge about the condition. Adolescents' curiosity about the condition, as indicated in the subtheme of 'unsolicited questions', further highlights their willingness to understand the core nature of TS. Most of the participants based their impressions of individuals with TS on various mediums of popular culture, verifying the notion that the media is a potent force that has a detrimental impact on the creation of perceptions (Mutz & Goldman, 2010). Adolescents who reported having relatives with Tourette's syndrome also endorsed commonly held stereotypes pertaining to the syndrome and acquired their knowledge from TV. In popular cultural portrayals, the accuracy of symptomology has been wilfully abandoned and extreme cases of TS have prevailed over the typical ones. The participants' stereotypical recollections of images of TS in the media are consistent with the findings of a recent study (Calder-Sprackman, Sutherland, & Doja, 2014) which demonstrated that Tourette's is used in films and television to exonerate the use of profanities and entertain the audience. Thus, for comic effect, Tourette's syndrome has been presented as the 'swearing disorder' where as in fact although coprolalia can be encountered in individuals with TS, only a minority is afflicted. Indeed, the prevalence rate of coprolalia ranges from 10 to 30 percent of the TS population and can be associated with severe symptomology (Eddy & Cavanna, 2013; Freeman et al., 2009)

Participants placed the onus on the abnormalities that underpin the condition and the pronounced differences between "them" and "us". In anticipation of intergroup interaction participants alluded to the notion of intergroup anxiety. Consistent with previous research anxiety would be evoked by affective components, leading to discomfort (Stephan, 2014).

Uncertainty and unfamiliarity may accentuate negative emotions which in turn could lead individuals without TS to quickly disengage from intergroup interaction. Indeed, the current study suggests that uncertainty about how to react towards individuals that were perceived as different, individual's self-doubt about their competence in facilitating interaction, and the expectancy that contact might be extremely difficult, exacerbated anxiety. The results of the study also suggest that psychological discomfort may stem from a form of cognitive dissonance. Individuals' egalitarian values and positive evaluations towards their peers with TS conflicted with the identification of their peers with TS as outgroup members (Festinger, 1962).

The adolescents in the study were also concerned about the negative social consequences interaction with an out-group member might entail and apprehensive about disapproval from other intragroup members. This would lead some to avoid and/or seek to create a distance between themselves and outgroup individuals (Stephan, 2014). Indeed, adolescents feared that a social relationship with a "blemished" individual (Goffman, 1963) would erode their social standing. In line with the mechanism of evaluative conditioning, their own devaluation by their peers would merely be the result of being paired with a disliked stimulus; in this instance a peer with TS (Walther, 2002). The phenomenon of the social contagion and spreading of stigma to individuals that do not possess the undesirable characteristic(s) is supported by much empirical evidence and acknowledged as a robust social phenomenon (Goffman, 1963; Pryor, Reeder, & Monroe, 2012). Studies suggest that the bearer of courtesy stigma does not necessarily have to have close and meaningful ties with a stigmatised person but the spill over effects could be a result of merely being observed in their presence (Hebl & Mannix, 2003).

In an attempt to understand TS participants also evaluated the actor's perceived responsibility in the manifestation of the condition. The vast majority of adolescents in the study attributed the causation of TS solely to factors that were beyond the individual's control and therefore inferred reduced personal responsibility and decreased moral accountability. Simultaneously however the actor was viewed as a passive victim and TS as a deterministic and stable condition that induced feelings of pity and sympathy towards the "sufferer". The limited participants who perceived the cause of TS as high on personal control exhibited negative emotions towards their peers with TS. These findings are especially interesting when studied in conjunction with the attribution theory framework (Weiner 1986, 1995). According to Weiner's attribution-emotion- action model the perceived level of responsibility and control an individual has over their actions may influence the observers' emotional response towards them and their subsequent behaviour. More specifically low controllability may engender high levels of sympathy and pity and elicit positive behavioural intentions. Conversely, perceived controllability may evoke more negative emotions, high levels of anger and subsequently antisocial behavioural intentions. The current study suggests that in the context of TS the theory is only partially supported. Perceived uncontrollability over the tics determined the participants' emotional reactions and the hypothetical peer elicited sympathy and pity. However, these emotions towards individuals with TS did not always increase the participants' willingness to decrease social distance and did not necessarily elicit benign reactions. Thus, adolescents who attributed the causation of the condition to factors beyond the afflicted persons control still avoided a close degree of contact with the target individual, and pity did not translate to prosocial behaviour and a desire to help. Our findings highlight that attitudes and behavioural intentions towards adolescents with TS is a complicated matter associated with a variety of issues that do not solely relate to attributional models and blame. Therefore, attributing the causation of TS to factors beyond the inflicted person's control cannot be perceived as a panacea for stigma reduction.

The findings also suggest that pity resulted from adolescents seeking out comparison with a group they viewed to be socially disadvantaged and wrongfully discriminated against. The theory of social comparison-based emotions maintains that affective reactions can be elicited by comparing oneself to superior others (upwards comparison) or inferior others (downwards comparison) (Smith, 2000). In this instance, a downwards comparison was activated that induced feelings of pity since the individuals were being perceived as a member of a lower status with limited prospects and life chances. Pity in the context of the study cultivated a form of benevolence stigma (Cohen & Struening, 1962; Corrigan et al., 2001). Thus, people with TS were perceived as powerless, fragile, vulnerable; the condition as a socially disabling condition to which people react with kindness but simultaneously akin to how parents treat their children. Consistent with the stereotypical content model hypotheses the findings of this study suggest that pity results in a paternalistic form of prejudice (Cuddy et al., 2007). In contrast to direct and aggressive forms of discrimination patronisation is masked by a seemingly positive behaviour towards the recipient. However, an unequal power relation between the "sufferer" and the one who pities them was implied. This entails a subordination and an unseemly dependency. The benevolent and ablest helper provides assistance to the less fortunate and expresses affection but not respect. The latter are viewed as in need of help and are construed as unable to manage on their own without outside intervention. The asymmetric power relationship that has been formed implies the superiority of the patron and the inferiority of the receiver. Indeed, as Nietzsche has pointed out actions taken because of pity are not necessarily altruistic. Rather they mostly stem from a selfish need to eliminate the internal pain caused by viewing the suffering of another. This form of paternalistic prejudice or benevolence stigma is subtler since it manifests in the form of protective tendencies although concealing condescension. It is also considered as positive behaviour from the point of the perceiver and therefore more likely to remain unchallenged.

Indeed, in contrast to overt discrimination which was perceived by the study participants as moral transgression, individuals maintained that they would behave in an ostensibly unprejudiced manner towards the out-group members. Thus, providing patronising help was conceived as motivated by moral principles and values and therefore justified as benevolent. Therefore, in keeping with previous research the findings of this study suggest that people try to behave in a manner that appears to be moral and maintain a positive social identity through exhibiting what is believed to be positive behaviour towards out-groups (Ellemers, Pagliaro, & Barreto, 2013). Nevertheless, Fominaya, Corrigan, & Rüsch (2016) suggest that being viewed as an object of pity can have detrimental impacts on a person's sense of worth, self-esteem and sense of empowerment.

Pity and benevolent attitudes also aroused a strong desire for social distance among adolescents and in this sense, our findings are consistent with those of Fominaya et al. (2016) who demonstrate that pity regarding mental illness is correlated to decreased anger but also increased social avoidance. It appears therefore that one's desire to provide help may be impeded by the desire to maintain a social distance. Indeed, in line with the Behaviours from intergroup Affect and Stereotype (BIAS) map (Cuddy et al., 2007) pity may engender an active facilitation, in this instance patronisation, but also a passive form of harm which is presented in the form of neglect and avoidance. Participants wanted to avoid the negative feelings that pity evokes and maintain distance from the unfortunate other or help him without wanting to establish a reciprocal relationship.

5.8. Limitations of this study

While this study makes important strides in understanding how individuals with TS are perceived by their peers, the findings should be interpreted within the context of the current study design. First, the study was limited by a small and homogeneous sample size, and the use of one data collection site. The participants were recruited through convenience sampling making the findings of the study specific to a particular school and target group and therefore difficult to generalise to the larger population of adolescents in the United Kingdom. Furthermore, although the school in question is characterised by ethnic diversity, due to the stringent participants' selection criteria, the sample of individuals that were from an ethnic background other than White-English were underrepresented. Convenience sampling also led to unequal representation of males (only 23 % of the sample) which suggests that the results may be limited when extrapolating both genders. A variety of studies suggest that females exhibit more positive attitudes towards individuals with disabilities and therefore our findings may be positively skewed (Goreczny, Bender, Caruso, & Feinstein, 2011).

Other limitations relate to the stimulus presented to the participants. In the video, the young woman with TS exhibited tics that were both high in frequency, intensity and severity which may have negatively influenced the participants' willingness to interact with individuals with Tourette's syndrome (Woods et al., 1999). Furthermore, Boudjouk's study (2000) demonstrates that adolescent females with tics are evaluated less positively than males with tics. Finally, by viewing segments of a video as opposed to an adolescent with TS in vivo the external validity of the study is limited. Therefore, hesitancy voiced by participants may have been due to a lack of familiarity rather than TS itself.

Further methodological limitations emerged as a result of using focus groups. In at least two focus groups a "collective" voice emerged that mostly represented the most influential participants of the group (Kitzinger, 1994). Consensus within the group and adolescents' need for peer approval may have limited some participants' willingness to express diverse views or opinions that could have been considered less socially desirable

(Sullivan, 2013). More specifically, a number of the study females exhibited a tendency to conform to the opinions of more confident and dominant peers and to rephrase and adjust their answers in order for them to comply with social desirable responses. Potential response bias could have also been due to the presence of a staff member in the room where the focus groups were taking place. Although the ‘authority figure’ did not participate in any of the discussions and was sitting silently in the back of the room, her mere presence may have inhibited the participants from expressing less socially desirable responses. Furthermore, despite the researcher’s effort to get all the participants involved and to encourage group building, not all adolescents participated equally. Thus, some responses were no longer than a few words and the participants were not able to fully engage in the conversations and the structure of the focus groups were more akin to an interview.

Finally, social desirability bias may provide a plausible explanation as to why the participants attributed the act of social exclusion solely to other peers and not to themselves. Driven by self-presentation and prejudice concerns participants may have underrepresented behaviours that they considered to be socially inappropriate. Overrepresentation of one’s own kindness and generosity may also be due to the “holier than thou” effect; people tend to perceive themselves as uniquely and unrealistically moral, ethical and altruistic in comparison to their peers (Epley & Dunning, 2000).

Researching in a school environment highlighted that unlike a laboratory setting, the social conditions of schools are not under the exclusive control of the researcher. One of the main challenges of doing school-based research is recruiting and gaining access to schools. Schools need to prioritise their teaching commitments and the immense pressure to constantly improve educational attainments make them unwilling to make space for research. Not being able to access schools is especially relevant to early career researchers.

Research demands may also be at odds with the purposes and policies of the schools. The head teacher chose the pool of participants that were invited to take part in the study, which may have created a biased sample. Furthermore, an authority figure had to be present during each of the focus groups which may have made the participants less willing to disclose negative attitudes. Thus, the school monitored and controlled the sampling process but also the encounters with the participants. This might have limited the validity of the study. There was a power imbalance between the researcher and the gatekeeper that was not negotiated. Thus, the researcher had to be flexible in what was actually possible in relation to research and adhere to the school's policy.

5.9. Implications for practice and future directions

Despite the identified limitations the study builds on scarce literature on adolescents and TS and advances knowledge about the barriers to social inclusion for individuals with TS. Targeting and addressing specific misconceptions in an educational intervention may be important because as studies suggest erroneous beliefs may be harder to change and may adversely affect the understanding of new information (Taylor & Kowalski, 2004). The findings of this study will help enrich educational/ anti-stigma interventions on the transmission of knowledge. It highlights that the media is the primary source of information for adolescents that is used to inform about TS and therefore can potentially be used for educational purposes. Indeed, future interventions addressed towards adolescents should utilise and exploit its dynamic role.

The findings also highlight that the stereotype of a distressed and disempowered individual with TS should be demythologised. Within this context emphasis should be placed on the contextual factors that impact the biological processes rather than solely attributing tics to defective genes. By presenting the antecedents that facilitate TS expression people with TS can be viewed as less distinct (Conelea & Woods, 2008). The nature of control over one's tics should also be addressed. Thus, the ability for one to suppress the tics even for a prolonged period of time may attenuate unpredictability and ultimately make individuals with TS appear less dangerous and disempowered by the condition. Conversely, emphasis on a biogenetic model and lack of any control over the symptomology can encourage the notion of socially distinct and even 'dangerous' and subsequently widen the gap between "them" and "us".

Future interventions also need to augment adolescents' willingness to help individuals with TS but without perpetuating the image of helplessness. Recent studies suggest that helping behaviour is not necessarily contingent to stereotypical depictions that elicit pity (Kamenetsky, Dimakos, Aslemand, Saleh, & Ali-Mohammed, 2016). Social inclusion can be achieved by communicating images of individuals with TS that are more congruent with typically developing peers' self-image. Empathy can be cultivated by addressing and focusing on the similarities between the stigmatised and the norms and not ruminating on deficits and differences (Campbell, Ferguson, Herzinger, Jackson, & Marino, 2004).

5.10. Summary

The aim of the study was to understand how TS is conceptualised by adolescents and explore how individuals with TS are perceived by their typically developing peers. Free text writing and focus groups were used to elicit the views of twenty-two Year Ten students from a secondary school in South East England. Grounded theory analysis was used to develop an analytical framework. Participants' understanding about the condition was construed from misconceptions, unfamiliarity and unanswered questions. Adolescents who conceived TS as a condition beyond the individual's control perceived their peers as being deprived of agency and strength and as straying from the boundaries of normalcy. People with TS were viewed as individuals deserving pity, and in need of support. Although participants maintained they had feelings of social politeness towards those with TS, they would avoid initiating meaningful social relationships with them due to fear of 'social contamination'. Intergroup anxiety would also inhibit a close degree of social contact. Participants that viewed those with TS as responsible for their condition expressed a plenary desire for social distance. However, these behavioural intentions were not limited to adolescents that elicited inferences of responsibility to people with TS, indicating that attributional models of stigmatisation may be of secondary importance in the case of TS.

The data generated from this study informed the intervention. More specifically the study indicated that the participants had misconceptions about the condition which stemmed from inaccurate and sensualised representations of TS through social media. These misconceptions were addressed in the educational component of the intervention. Furthermore, it aimed to distort the pity and blameworthy discourse about the condition which inevitably led to marginalisation. The intervention will be discussed in the following chapter.

6

The Intervention

The previous chapters of the thesis have indicated the form the intervention needed to take in order to ameliorate stigmatisation for individuals with TS. More specifically, chapter 5 suggested which misconceptions needed to be addressed and the importance of addressing the discourse of pity and blameworthiness. In this chapter the justification for the design on the intervention will be discussed in detail.

6.1. Introduction

Previous empirical research related to improving attitudes towards Tourette's (e.g. Friedrich et al., 1996; Woods & Marcks, 2005) has tended to entail very short educational interventions that simply provide knowledge about Tourette's to the participants, without taking into consideration the characteristics of the targeted population (e.g. demographics, previous contact with people with Tourette's syndrome, previous knowledge about TS, attitudes and behavioural intentions towards individuals with TS).

In this study, an intervention workshop was developed using data from Study One, thereby enabling it to be tailored to the populations' understanding and attitudes towards Tourette's syndrome. In this way, Study One functioned as a tool to guide the development of the subsequent workshop; helping to identify factors which were associated with adolescents' attitudes and behavioural intentions; and providing specific directions as to the areas that needed to be targeted in the intervention.

Study One helped us identify modifiable variables associated with stigmatising attitudes and pinpoint the aims of the intervention; dissipate misconceptions and stereotypes about TS and reduce preference for social distance by challenging the collective pejorative understanding of the condition. The biological causes of Tourette's had to be presented in order to avoid typically developing adolescents believing that individuals with TS are responsible for their condition. The intervention however did not rely on the medical model. The workshop intended to modify the erroneous belief that individuals with TS are fundamentally different from their typically developing peers. It also aimed to shift impressions of people with Tourette's as a 'dependent and unfortunate others' who should be treated as a child. Therefore, whilst the workshop was designed to evoke empathy it also tried to present Tourette's syndrome in a positive rather than a 'pitiful' light.

6.2. Theoretical foundation for supporting the use of the intervention

The intervention was based on the findings of Study One in combination with the results of the previous systematic review (see chapter 3). The intervention workshop consisted of the following:

The viewing of a feature film "Front of the Class" as well as a PowerPoint presentation of factual data about the condition followed by a discussion with the participants; this format being based on Allport's (1979) principle that attitudinal and behavioural changes can be achieved through both education and contact. The combined activities aimed to challenge pre-existing stereotypes through contact with an individual with TS, replace myths with accurate information, and build upon the similarities rather than the differences between the participants and individuals with TS.

Contact is a commonly used effective approach that promotes intergroup contact in order to ameliorate stigmatisation (Corrigan & Penn, 1999). As mentioned in the previous chapter contact can be in the form of ‘in vivo’ communication or indirect, video-based contact. In vivo contact requires individuals with TS who would be willing to disclose their personal experiences, take time out of their everyday lives to deliver their presentations and comply with the schools’ timetable. Despite the obvious advantages of face-to-face interaction recent studies suggest that both forms of contact can lead to significant changes in attitudes towards stigmatised individuals (Janoušková et al., 2017; Reinke, Corrigan, Leonhard, Lundin, & Kubiak, 2004). In the context of the current research indirect contact was considered more appropriate due to the study’s intent to develop an intervention that can be easily disseminated within school settings, in a cost-efficient manner. Furthermore, having different individuals present their lived experience of TS can be viewed as a barrier to achieving consistency in the message that is being communicated. Conversely, an intervention that is based on an indirect-video-based contact is reusable; it can be more easily disseminated; it offers consistency and provides anonymity to the presenters and also ensures that no strenuous requests are put on them (Clement et al., 2012).

Adolescents’ unsolicited questions concerning TS belied their insufficient knowledge about the nature of the condition (see Chapter 5). This suggested that (non-physical) contact with an individual with TS should be accompanied by an educational component in order to improve TS literacy. Thus, factual information should be provided and commonly held stereotypes should be addressed and demythologised.

This sequence of the intervention elements (i.e. film, power point presentation, discussion) was chosen as per Wahl and Yonatan Lefkowits (1989)’s advice that educational material of a specific condition presented before a film may not be critically appraised by the audience; its value being overshadowed by the film. On the contrary in this study, the overall aim of the film was to sensitise the audience to TS in order for them to be emotionally and mentally ready to process new information.

The intervention was implemented during a 2hour single session. The duration of the intervention was designed taking into account the schools' timetable. Furthermore, our systematic review on school-based interventions (Chapter 3) exemplifies that a brief intervention can be just as effective as a lengthy one in ameliorating stigmatisation. Each part of the two components of the intervention are discussed in detail below:

1. *The film "In Front of the class"*

Indirect contact with an individual with TS was facilitated by the film "In Front of the Class". A number of recent studies indicate that films can be particularly effective in causing audiences to reflect on stereotypes and subsequently reduce stigmatising attitudes (Altindag, Yanik, Ucok, Alptekin, & Ozkan, 2006; Conrad et al., 2014; Quinn, Shulman, Knifton, & Byrne, 2011). The film was deemed appropriate due to the targeted groups' interests and understanding about TS. As opposed to previous interventions, the specific workshop aimed to elicit a strong emotional response from the audience which would possibly not be feasible in the context a traditional lecture. Furthermore, as Study One highlighted mainstream media was the basic source of information and misinformation about TS for adolescents. Through exaggerated, cliché and ridiculed portrayals, TS was conceptualised as a cursing disorder affecting individuals whom it disempowered and who were substantially different from typically developing adolescents. It was the aim of the study to contrast the stereotypical media portrayal of TS and allow the participants to identify with an individual with TS using the same medium that was used to misinform them.

The film is based on Brad Cohen's autobiographical book in which the protagonist manages to overcome the hurdles set by society and accomplish his lifelong dream of becoming a teacher (Cohen & Wysocky, 2008). Through first person narrative the film explores Brad’s life with Tourette’s syndrome as an adult while attempting to become a teacher as well as actively trying to engage in life and romance. Using numerous flashbacks the film repeatedly depicts how as a child Brad tries to ‘fit in’ with his peer group, endures bullying from them and performs poorly academically because of his teachers’ ignorance about Tourette’s syndrome, inflexibility, and unwillingness to make reasonable accommodations. Yet, the film also shows how the head teacher of his school inspires him to become a teacher in order to touch the lives of other children.

The film places a focus on the protagonist’s agency and free spirit while also pointing out the constraints society sets out for him and in doing so succeeds in eschewing the regressive medical model. However, other secondary themes are tackled such as a difficult father-son relationship, lower than normative expectations for people with TS, and employment discrimination.

The relationship between the protagonist and his father is delineated by the latter’s conflicting feelings of love, guilt, unwillingness to accept his son’s condition and impatience. Low expectations for individuals with TS is also highlighted through the support group Brad and his mother attend after he first received his diagnoses. In these meetings individuals with TS and their parents appeared to have adapted and accepted the fact that the hypervisibility of TS renders it socially disabling and that TS can be only be dealt with in the realm of the family. Finally, a blatant incident of employment discrimination is presented when the protagonist is categorically turned down from job despite being the most qualified candidate for it due TS-bias.

Besides the protagonist the film depicts secondary characters that either assist Brad in accomplishing his goals (e.g. his mother, his brother, the head teacher etc.) or create barriers for him. This occurred by either discriminating against him or pitying him (e.g. employers discriminating against him). These characters could work as prosocial models or negative role models for the audience that are likely to mimic desirable and avoid undesirable actions. Indeed, according to social cognitive theory (Bandura, 1986) people tend to learn vicariously by observing certain behaviours. In the case of “Front of the class” viewing similar others exhibit prosocial behaviour towards an individual with TS and observing the positive outcome of their action could reinforce positive behaviour. Respectively, negative behaviour towards individuals with TS is negatively reinforced.

The film can be embedded in the category of Entertainment-Educational films, due to its content. Entertainment- Education (EE) is a commonly used communication strategy that is based on the delivery of educational and prosocial messages through popular media, for instance radio, soap operas, films etc.(Moyer-Gusé, 2008; Singhal & Rogers, 2004). EE has been used widely since the 1940’s in the context of health educational campaigns but also in an attempt to change the social norms in relation to mental disorders, gay marriages etc. It’s effectiveness in promoting social change has been exemplified by a wide variety of evaluation studies (Pondexter, 2004). As any other form of psychoeducational intervention, it aims to influence awareness, attitude and subsequently the behaviour of the audience in both an individual and collective level. However, its uniqueness lies in the fact that it’s not solely based on the diffusion of information but its narrative structure allows the audience to immerse and engage into the world in which the drama is taking place and identify and feel sympathy towards the main characters. Thus, as in other forms of arts films enable the audience to engage in an emotional level and reduce the sense of otherness, allow the viewers to identify with the protagonist and break the barriers between “them” and “us”. Furthermore, it enables the audience to experience contact with the protagonist almost at a face-to-face interaction as if a reciprocal conversation and relationship (Horton & Richard Wohl, 1956). Thus, EE induces a form of cognitive and emotional response that allows the viewers to adopt the perspective of the protagonists.

Educational Entertainment is also based on the theoretical principles of the Extended Elaboration Likelihood model (E-ELM) that posits that subtle persuasive messages can be conveyed without the viewers being fully aware of the influential intent if they are engaged in the story plot (Petty & Cacioppo, 1986; Shrum, 2012). Thus, audiences' resistance, and defence mechanisms in accepting the persuasive message that is being communicated are more likely to be weakened.

The film does not only examine TS closely but also indirectly criticises the value of homogeneity and society's and school's failure to accommodate for different abilities. It also addresses some of adolescents' concerns regarding individuals with TS according to Study One; inevitably low attainment in school, restricted career choices, vulnerability to employment discrimination and inabilities to acquire and maintain a job that is directly associated to public speaking and social interaction. The film suggests that academic success may be dependent on the appropriate accommodations provided to individuals with special educational needs and that challenges in acquiring jobs may be partially based on stigma and not "within the individual". It also depicts the protagonist as an active rather than passive character that does not live within an isolated realm. In doing so the film manages to deconstruct the discourse of pity in relation to TS.

The film does not position Brad as a tragic victim and TS is not framed within a narrative of individual misfortune. It is not centred on the challenges that the protagonist must overcome in order to fit into society's stringent criteria of normalcy. The film diverts from the regressive medical model that views any form of disability as a functional impairment within the individual that needs to be treated, cured or rehabilitated (Oliver, 1996). On the contrary, the film placed the onus on the external and attitudinal barriers that the society poses on the individual and in the plot the main character does not adapt to fit society's notion of normalisation. In other words, the protagonist does not try to find a cure to TS or assimilate to norm by completely eliminating the symptomology of TS. Tourette's syndrome was not presented as a problem that the main protagonist must individually overcome but the emphasis was placed on the limitations and constraints which society posed on the protagonist and the need for social change. Indeed, the cultural construction of disability is delineated by pointing out various forms of discrimination that impede Brad's success.

The film also manages to overcome the stereotype of the "supercrip prototype", a prevalent image in the media in which the disabled individual is portrayed as a savant or a remarkable achiever that manages to overcome their disability through sufficient willpower, self-discipline, stamina and determination (Kama, 2004). This model which is intended to aspire and motivate non-disabled people may reinforce the idea that limitations imposed by disability can be overcome and normalisation can be achieved if the disabled people work hard and dedicate themselves to this goal. The obvious disadvantage of this is that it can fosters an essentialist framework that poses unrealistic expectations about what is attainable for individuals that stray from the norm and absolves society's responsibilities of providing reasonable accommodation for all. Thus, it provides a standard against which other people will be judged and may therefore disempower disabled individuals that have not achieved extraordinary deeds (Shakespeare, 1994).

In contrast to the supercrip narrative Brad is not portrayed as accomplishing superhuman feats nor does he appear to possess abilities beyond those of most people. Although the protagonist manages to become an inspirational teacher, the film resists indulging the stereotype of the compensatory abilities (Shakespeare, 1999) in which the disabled person is compensated with talents to make up for his disability. The film upends this stereotype by portraying Brad as a whole and complex person with a full range of human characteristics and ‘typical emotions’ and not as a superhuman simply possessing will power, determination and good attitude.

Although the film upends the stereotypical portrayals that prevail in mainstream films such as the pitiable individual, the supercrip, and the need to adjust and assimilate to the norm it does not rise above common pitfalls that can be encountered in western films of the twentieth- century. Firstly, the fact that the protagonist, Brad, is portrayed by a tic-free individual brings up the concept of “disability drag” as termed by Tobin Siebers (Siebers, 2008). Indeed, a concern that has been raised by scholars is a masquerade form of disability in which a non-disabled actor, this case a tic-free actor, portrays a disabled individual. Through this process the tic -free actor’s enactment is highlighted more than the lived experience of TS. Although the actor’s performance was realistic, nuanced and three dimensional, the mere fact that the film did not feature a protagonist with TS makes the stigma of TS more visible on screen. Secondly, the film propels pathos and sentimentality and does not resist the fairy-tale ending with the protagonist’s reconciliation with his father and his triumph in finding true love. However, the authenticity of the film was enhanced by highlighting that the plot was based on a true story. Indeed, “Front of the Class” purports to be a realistic biographical portrayal. Any criticism over the characters and the plot should also be approached within the realm of an art form that does not aim to accomplish a literal representation of reality.

Furthermore, the fact that the protagonist was a male and not a female needs to be taken into consideration. Studies have indicated that males with tics are seen more favourably than females with tics (Boudjouk, 2000). Therefore, the outcomes of the intervention might have also been influenced by the gender of the protagonist.

Despite the aforementioned limitations, the film conveys the message of empowerment and accurate educational information about TS.

2. Educational component and discussion (30 min)

Similar to the study of Ritterfeld & Jin, (2006) the screening of the film was followed by a short educational PowerPoint presentation (see Appendix U) about the condition of TS followed by an open discussion. The aim of the presentation was to provide factual information about TS, address questions adolescents may have about the condition and to disconfirm negative stereotypical beliefs. It followed the format of an educational PowerPoint slideshow that was produced by Tourette's Action, a UK charity that aims to improve the quality of life for individuals with TS. The presentation is developmentally appropriate for secondary students and freely available on the charity webpage[3]. Its content and language were developed based on the age and cognitive development of secondary students. Furthermore, it did not contain medicalised terminology and did not endorse the biogenetic model that can potentially increase stigma rather than ameliorate it (Angermeyer, Holzinger, Carta, & Schomerus, 2011). The presentation was also informed by the qualitative part of the PhD study and it aimed to address particular knowledge gaps or misconceptions that adolescents had. The session was conducted in an informal and open manner in order to provide the participants with the chance to ask questions and seek clarifications. Factual information was provided in a logical sequence during the lecture and the key points were emphasised in the slideshow in order to enhance students understanding about TS. The first topic concerned the symptomology of TS. The commonly held perception that TS is a "cursing disease" was challenged and it was emphasised that coprolalia is a common symptom amongst individuals with TS. Following this the possible causes of Tourette's were outlined. Although the biological basis of Tourette's was highlighted, it was the aim of the intervention to move beyond the medicalised approach and help students understand the biological, psychological and social aspects of TS. Therefore,

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[3] http://www.tourettes-action.org.uk/86-presentations-for-schools.html

a more holistic explanation about TS was provided with the onus placed on the contextual factors that exacerbate the symptomology, the idea being to try and minimise the distance between “us” (the participants) and “them” (people with Tourette’s). Commonly held questions derived from the first study data were also addressed: the course of the syndrome; the controllability of the symptomology and; medication and effective treatments were some of the topics discussed. Finally, famous people with the condition and the epidemiology of TS were presented.

Questions were encouraged throughout the presentation and a discussion was initiated drawing on the myths and the cumulative message about TS socially constructed by the media. Participants were encouraged to reflect upon the validity and accurateness of the information they had previously received about TS. It was important however to communicate to the participants that they were not to blame for their misconceptions about TS. This non-blaming approach was highlighted through acknowledging that TS-bias is deeply rooted in our society and easily internalised. The adolescents were active participants and their engagement with the speaker and with one another allowed the participants to reflect upon the new information that contradicted their previous believes.

7

Evaluating a School-based Educational Workshop Intervention About Tourette's Syndrome for Adolescents: A Randomised Cluster Control Trial

In the previous chapter the justification for the design of the intervention was provided. In this chapter an empirical evaluation of the intervention is reported.

7.1. Introduction

In the previous chapters, it was shown that no previous studies have examined the impact of educational interventions concerning attitudes towards TS on typically developing adolescents and have only investigated their efficacy on adults and children. Furthermore, methodological weaknesses of previous intervention research may have limited their validity. These weaknesses include not measuring Tourette's knowledge as a variable; not assessing if factors such as previous contact and gender are associated with stigma; relying solely on quantitate measures to assess the outcome of the intervention and finally, only utilising a pre- and post-test design and not measuring the maintenance of the effects over an extended period of time.

The programmes that have been empirically assessed scarcely evaluate knowledge and its impact on attitudes and only two studies which delve into Tourette's use a Knowledge Questionnaire (Holtz & Tessman, 2007; Nussey, Pistrang, & Murphy, 2014). Thus, although previous interventions have provided information concerning the condition, they have not assessed whether or not more accurate knowledge about the condition establishes better attitudes and behavioural intentions. Knowledge was therefore not experimentally manipulated and there was no definitive evidence to suggest that it can exert a causal influence on attitudes and behavioural intentions. In this study, more accurate knowledge about Tourette's syndrome was deemed to be an important outcome in itself. It was also viewed as a significant variable that could be used for a manipulation check to assess if more accurate knowledge can produce better attitudes and behavioural intentions.

Another limitation of previous studies is that they have not evaluated whether previous contact with a person with TS and other stigmatising conditions can moderate the effect of knowledge, attitudes and behavioural intentions. So, the extent to which familiarity can influence perceptions and subsequent desired social distance from individuals with TS has not been assessed. Due to the gap in TS literature, this research draws from studies on mental illness and disability in general. Research studies suggest that familiarity with people with a mental illness or a general disability is associated to more favourable attitudes and behavioural intentions towards that population (Angermeyer, Matschinger, & Corrigan, 2004; Martinez-Zambrano et al., 2013; Pinfold et al., 2003). Thus, people who have greater knowledge about mental illness or direct experience are less likely to endorse stereotypes and express a desire for social distance. Based on the aforementioned literature we can predict that increased familiarity with TS and/or other mental or developmental disorders will positively affect initial attitudes towards individuals with TS. Moreover, positive attitudes will foster a preference for lower social distance. The relevant studies also indicate that individuals that are familiar with people with stigmatising conditions are more responsive to anti-stigma interventions than their counterparts as evidenced by them reporting more favourable attitudes after the intervention.

Variables such as gender have not been evaluated in depth although it is well acknowledged that gender may play a prominent role in stigma endorsement (Pinfold et al., 2003). Females have tended to report more positive attitudes and it appears that following interventions, changes in stigmatising beliefs are augmented (Goreczny, Bender, Caruso, & Feinstein, 2011). Thus, these interventions have greater influence on females than on males.

Interventions have also hitherto assessed the transient impact rather than the longitudinal effects of the intervention. Thus, no anti-stigma intervention relating to TS have yet adopted a longitudinal design, and no follow-up data beyond the initial two-time points (baseline, post-intervention) have ever been obtained. Evaluating the sustainability of changes is very important since a number of anti-stigma interventions demonstrate that the degree of change may be brief and not stable over time (Thornicroft et al., 2016). Indeed, most interventions result in short-term attitudinal changes, with the impact depleting with time. In this study, long- term follow-up (9 weeks after the intervention) were used to assess if the initial gains could be sustained or if they attenuated over time.

Finally, interventions that aim to reduce TS stigma have tended to be based solely on quantitative methodology which aims only to assess the effectiveness of the intervention (Holtz & Tessman, 2007) and as such, have not delved into understanding the participants reasoning for their attitudes or behaviour and how the intervention could impact their line of reasoning. Therefore, the current study utilised a concurrent mixed- method design (Teddlie & Tashakkori, 2003) in which the two types of data (qualitative and quantitative) were collected simultaneously. This allowed for a more comprehensive understanding of how the educational intervention impacted tic-free adolescents.

The current study therefore aimed to move beyond the shortcomings of previous research and establish an innovative methodological approach. It is the first TS intervention study to utilise a concurrent mixed-method design and it is the only study to my knowledge that evaluates the long-term effectiveness of the intervention.

7.2. Aims and hypothesis

The aims of the research were as follows:

1. Firstly, to extend the knowledge base of tic-free adolescents' initial knowledge, attitudes and behavioural intentions towards an unfamiliar peer with TS;
2. Using a mixed- method design the study also sought to understand the participants' reasoning and to provide a tentative explanation for their attitudes and behavioural intentions;
3. The third aim of the study was to empirically evaluate the immediate impact of the educational workshop on the participants, though it was also pertinent to assess if initial gains can be sustained over a period of time after the intervention or if they are attenuated;
4. Finally, it was also deemed important to evaluate the moderating influence of gender and the mediating effect familiarity with individuals with TS and other stigmatising conditions might have on tic-free adolescents' attitude and behavioural intentions.

In light of the above aims, four hypotheses were identified:

1. Participants who received the intervention would have significantly better scores on measures relating to knowledge about TS, attitudes and behavioural intentions towards individuals with TS in comparison to participants who did not receive the intervention (waiting-list control group);
2. At the follow up stage (9 weeks later) the observed changes in scores relating to knowledge attitudes and behavioural intention for the intervention group would be

decreased from the scores obtained immediately after the intervention but also significantly increased in relation to baseline measures;

3. The perceivers' gender would affect their attitude and readiness to enter different types of social relationships with an individual with TS at a baseline level. Furthermore, gender would mediate the effect of the intervention.
4. That familiarity with TS and other mental or developmental conditions would affect the initial attitudes and behavioural intentions towards individuals with TS and would mediate the effect of the intervention.

7.3. Study design

The study followed a randomised cluster control trial with a waiting-list control group. Randomisation of the intervention occurred on a classroom level. Within the same school one class of Year Ten students was randomly allocated to the intervention condition, while the other class of the same grade formed the waiting-list control group. The decision to allocate the two classes to one of the two conditions (experimental or control) was taken by the head teacher of the school-based on random criteria. Randomisation was implemented at a classroom and not an individual level in order to minimise the risk of spill-over effects. However, it was acknowledged that peers within the same grade level are frequently taught in common groups and interact with each other and therefore the risk of diffusion of the treatment could not be eliminated (Pinto-Foltz, Logsdon, & Myers, 2011).

The two groups (intervention, control) were compared at three different intervals (T1= baseline, T2= immediately after the intervention, T3= 9 weeks after the intervention) in a period of 10 weeks. Students assigned to the control group continued with their timetable as usual and did not receive any form of "active" intervention during the intervention stage.

However, the intervention was implemented on the control group nine weeks later, after the follow up stage. Thus, in line with the waiting-list design the control group served as the comparison for the treatment group and later received the intervention (Elliott & Brown, 2002). Whilst previous studies have not used a waiting-list control group in this study both the school and I recognised that withholding a beneficial intervention from the control group and not implementing a waiting-list control group would be neither fair nor ethical. Indeed, when an intervention is viewed as being beneficial, even though there may be little empirical evidence to back it up, a random process to determine who receives the intervention first can be considered reasonable, only if everyone receives the intervention within an acceptable amount of time.

A concurrent mixed method design was used as defined by Tashakkori and Teddlie (2003). During each phase of the research process (baseline, post-intervention, follow-up) data were elicited through a questionnaire that included both rating scales (quantitative data) and open-ended questions (qualitative data). Both paradigms were utilised in order to obtain data that could substantially address the research questions and the breadth of the inquiry could be expanded (Greene, Caracelli, & Graham, 1989). The Likert scale allowed for answers pertaining to levels of agreement and disagreement and monitored changes in outcomes relating to knowledge, attitude and behavioural intentions. Qualitative questions on the other hand explored matters relating to ‘how?’ and ‘why?’. Taking into account the complex nature of attitudes and behavioural intentions, open-ended questions to avoid constraining the respondents’ frame of reference, and it allowed the participants to communicate more than standardised responses, to explain their choice of response and to express their main concerns in relation to the topic. Open-ended questions also helped obtain clarifications and information which might provide explanations for human attitude and behavioural intentions. They also encouraged deeper cognitive engagement and processing since the respondents would have to think and justify the reasons for endorsing or opposing an attitude or a behaviour. Finally, in comparison to one-to-one interviews open-ended survey questions offer greater anonymity and therefore potentially elicit more bias-free answers than one-to-one interviews or focus groups (Bryman, 2012).

7.4. Settings and participants

More than twenty secondary schools in both Kent and London were contacted via emails and telephone. Despite the schools' secretaries' assurance, which acted as a safeguard, that the head teacher would be contacting me to discuss the project further, speaking to a teacher or the head teacher of the school was essentially impossible. Only four of those schools yielded responses. They declined to take part in the study due to other research obligations, exam preparation and overloaded schedules. Only one school agreed to participate.

The participants were recruited from the same school where Study One took place, a co-educational, multi-cultural comprehensive secondary school in South East England. I arranged an hour-long meeting with the school's head teacher to ensure the feasibility of the logistics of the workshop but also to provide a synopsis of the study. The aims and significance of the research was pointed out as well as timeframe, and the requirements of the study for the school. Similar to Study One, the head teacher was asked to identify two classes which met the projects inclusion criteria, which stipulated that a child with identifiable symptoms of TS was not enrolled in them. This prevented indirectly stigmatising the student with TS and having the rest of the pupils unconsciously reflecting their thoughts about the specific child on their answers. There were no exclusionary criteria for participants in terms of gender, cultural and linguistic background or Special Educational Needs (SEN). However, study participants had to be fluent in English, in order to understand the questionnaire and the content of the intervention. Participants were also invited to participate regardless of their level of literacy skills as long as support was provided. The head teacher identified two Year Ten class that fulfilled the criteria and a total of 59 students were engaged in the project. Clark-Carter (1997) suggest that in order to obtain a medium effect size (d=0.5) using a two-tailed test in a between participants design, with power at 0.86 and alpha at 0.05, 30 participants would be required in each group. Also, in order to achieve a medium effect size (d=0.5) using a within subject design, so as to detect any difference between pre-intervention, post-intervention and follow-up measures, with power at 0.75, 30 participants would be required.

7.5. Procedure

The study was reviewed by the University of Kent Ethics Committee from which a favourable opinion was given. The submission included a description of the background, aims, objectives, measures and methodology and all relevant information sheets and consent forms accessible to the participants in terms of age (see appendix G-L).

Four weeks prior to the start of the study information sheets and opt-out consent forms were sent home to the parents/guardians of all eligible students. Parents/Guardians were asked to contact the researchers by phone or email if they had any questions about the study. They could also actively decline their child’s participation by signing the opt-out consent form (passive parental consent). Similar to Study One this method of obtaining parental permission was dictated by the particular policy of the school. It was also deemed appropriate since the research involved only minimal risk to participants. Under conditions of passive parental permission higher participation rates and non-biased samples are more likely to occur (Dillman, 2000). It was the aim of the study for students that were less academically capable, from diverse ethnic backgrounds, lower socioeconomic homes and from parents with lower education to be equally represented in the population (Shaw, Cross, Thomas, & Zubrick, 2015). By presenting the opinions of all the different racial minorities and using a non-biased demographic sample the true culture and ethos of the school would be revealed which might reflect the attitudes of the area's wider society.

Verbal opt-out assent was sought from the pupils. As opposed to Study One, potential participants were not required to sign an assent form to attest their refusal to participate in the study. Rather they were asked to state their assent verbally to the researcher or their teachers (a requested signature being deemed to place an unnecessary strain on the adolescents according to the University ethics committee). Ungar, Joffe, & Kodish, (2006) argue that there must be a clear distinction between how consent to participate should be sought from minors in comparison to adults. In the latter population, parental consent is also required and a signature from the minors should only be sought to indicate the autonomous authorization to take part in the study. In this study since parental consent had already been sought, it was decided that adolescents should not also be required to submit a signature when they may not fully comprehend the symbolic meaning of it. In other words, the minor's signature should not be used by the researcher as proof that the consent protocol was followed to the letter when it can distract the participant from engaging in the study.

Data collection commenced three weeks after the information sheets and consent forms had been distributed and it occurred at three different points in time over a period of two and a half months: one week prior to the intervention (baseline- T1), immediately after it (post- intervention T2) and two and a half months later (follow- up- T3) (see Figure 5, p. 178). The initial plan was for the follow up (T3) to be conducted a year post-intervention. However, it was made clear that the head teacher, who was committed to participating in the research, was going to be retiring and could not guarantee access to the school for the following academic year. Indeed, the incoming head teacher was negative towards conducting research as the study was not seen as a viable investment to the school. Therefore, T3 had to be arranged before the end of that specific school year. For the same reason it was also not possible to conduct T4, which may have produced further interesting results.

Baseline measures were taken one week before the actual intervention was implemented (T1). All students from the two Year Ten classes were gathered in a large hall within the school premises during school hours. None of the parents/guardians had opted their child out of the study. The researcher briefly introduced herself and the project and reminded the participants that their involvement was voluntary and they would not face any

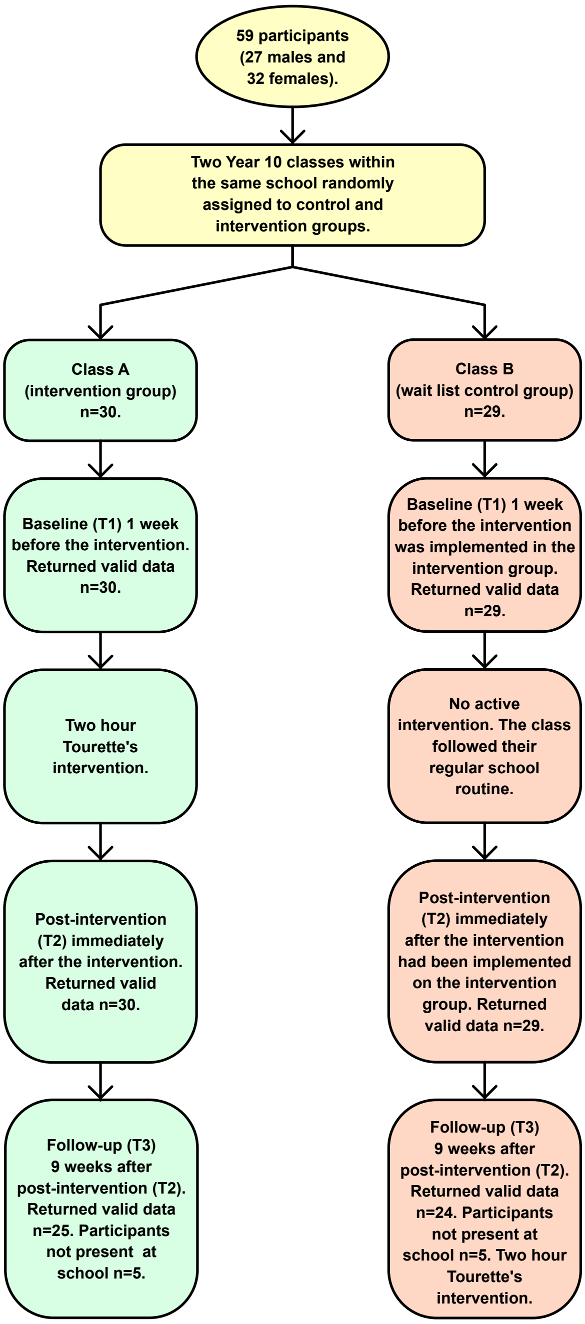

Figure 4 - Study procedure and attrition of participants.

negative consequences if they decided not to participate or drop-out of the study at any point. Furthermore, participants were informed that an alternative school activity would be provided for them if they chose not to participate. Nevertheless, none refused to participate. The head of the school who briefly came to show his support also highlighted the importance of the study. The high percentage of participation indicated the feasibility of the study since all Year Ten students that were eligible and invited to participate were enrolled in the study and none of them withdraw at any point of the study. On the other hand, retaining all adolescents' participation at the 9-week stage was not possible. Due to absenteeism from the school at the final collection time point, 17% of the participants did not participate in the follow-up.

All participants were shown the same short stimulus video as in Study One. Before the questionnaires were distributed participants were reminded to answer with honesty and without consulting with or viewing responses of other participants. It was also emphasised that their answers would be kept confidential (not be shared with anyone other than with the researcher and her supervisor). The students were not given any information about TS before being administered the questionnaires and given 20 minutes to complete the packet of questionnaires at which stage the demographic and the level of contact report had been included.

In the following week (T2) the head of the school randomly allocated one class to the intervention condition and one to the waiting-list control. The intervention group was initially shown the movie, which they viewed with great interest. Immediately after the viewing of the film the participants asked a lot of questions pertaining to TS which gave the researcher the impetus to commence the PowerPoint presentation. A short discussion followed where students were encouraged to talk about their conceptualisation about TS and their novel understanding of it based on the new information they had received. The packet of questionnaires was distributed immediately after this. In addition to the questionnaire about Knowledge, Attitudes and Behavioural Intentions participants were also given a questionnaire to and evaluate the intervention and its components.

The control group did not receive any active intervention at T2 and continued with its usual timetable. Data was gleaned from them during a 20 minutes session the same day.

Initially, the follow-up was scheduled 8 weeks after the intervention. However, the head teacher suggested that it be moved a week back because it coincided with the participants' exam time. The researcher happily complied. Data was gleaned from the intervention group during a 20-minute session and the intervention was implemented for the control group after the follow-up data were obtained (T3).

During all of the data collection points the researcher was present in the classroom to answer relevant questions and to ensure that participants did not collaborate with each other when answering the questionnaires. At T1 and T2 participants were accompanied by their regular teachers and two teaching assistants who supported particular individuals during the whole intervention. Although the questionnaires were designed to be completed by the pupils' unaided, two students who usually received academic support within the class were assisted during data collection by their teaching assistants who were briefly advised on how to support the participants without reflecting on their own opinion or on the potentially socially appropriate responses. Furthermore, the researcher read aloud all the questions to participants to accommodate students with literacy difficulties.

7.6. Measures

7.6.1. Demographic information (Appendix M)

Questions were asked concerning the pupils' age, sex, ethnicity and first language.

7.6.2. Level of contact with people with Tourette's syndrome and/or other intellectual/developmental disabilities and mental illnesses questionnaire (Appendix N)

These questions referred to the level of contact participants previously had with individuals with Tourette's syndrome as well as with people with other developmental disabilities (e.g. Autism, ADHD) or mental health illnesses (e.g. depression, anxiety disorder) which are also generally regarded as stigmatized. The intensity of familiarity was assessed using the Revised Level of Contact Report for Adolescents (Corrigan et al., 2005; Holmes, Corrigan, Williams, Canar, & Kubiak, 1999). For this specific study seven situations were provided that described different levels of contacts a person has had with a stigmatising condition. They ranged from minimal contact (e.g. I have never observed a person with TS) to personal contact and medium intimacy (e.g. I have been in a class with a person with Tourette's syndrome (e.g. I have been in a class with a person with Tourette's syndrome) to high intimacy (e.g. I have Tourette's syndrome). Participants were asked to check all the situations that they had experienced on each of the item lists and the index.

7.6.3. The Tourette's knowledge questionnaire (Appendix O)

Previous studies pertaining to Tourette's syndrome have not placed the emphasis on TS awareness and only a minority of them have utilised a relevant questionnaire (Holtz & Tessman, 2007; Nussey et al., 2014). For the purpose of this project a questionnaire was developed utilizing both the existing evidence base and the data gained from Study One. Commonly held misconceptions adolescents reported about the condition and queries they asked the researcher during the first study regarding the aetiological causes of TS, the genetic predisposition etc. were included in the questionnaire.

The Tourette's knowledge Questionnaire consisted of 18 items, eight multiple choice questions and ten true or false questions. The 'don't know' response option was included to reduce the probability of pupils answering correctly by chance. The content of the questions covered several aspects of the condition including the symptomology of TS, aetiology of the condition, the prognosis and the social aspect of the condition. Responses to these items were scored as either correct or incorrect. Possible scores ranged from 0 to 18, with higher scores indicating more accurate knowledge about the condition.

During the study participants were never given the right answers to the knowledge questionnaire in order to minimize subject learning and different questionnaires were also randomly ordered within the packet each time they were distributed in order to minimize the threat of testing.

7.6.4. Attitude questionnaire (Appendix P)

Adolescents' attitudes towards individuals were assessed using a 10-item self-report specifically designed for this research, since there were no existing standardised questionnaires pertaining to attitudes towards individuals with Tourette's syndrome, Study One was used to develop this measure by utilizing user-defined criteria. Thus, participants' statements and opinions as captured by the focus groups and free text writing were used to construct the questions. From a pool of statements and views pertaining to individuals with Tourette's syndrome the most relevant were chosen that reflected commonly held attitudes among the participants.

The questionnaire included statements that reflected domains of undesired differentness, benevolence, social restrictiveness and integration. Undesired differentness referred to statements that class people with TS as inferior to normal that require different handling (e.g. 'Just because they have Tourette's you shouldn't treat them any differently').

Benevolent views referred to opinions which suggest that individuals with TS should be viewed upon with kindliness and pity due to their misfortune (e.g. ‘I feel sorry for them as they can’t have a normal life’). Restrictiveness and integration, referred to opinions that assert that individuals with TS may pose a threat or a burden to their social surrounding and their presence must be met by some restrictions (e.g. ‘If the tics are really bad, then they should go to another school’). Half of the statements reflected a positive attitude towards the research object and half a negative attitude. Items were rated on a 5-point Likert scale ranging from total disagreement (1 “Strongly Disagree”) to total agreement (5 “Strongly Agree”). Internal reliability was reported at a Cronbach’s alpha of 0 .79 (acceptable), with test-retest reliability of 0.8 (good). Internal reliability was reported according to the George & Mallery, 2003 criteria: ≥0.90 excellent, 0.89 to ≥0.80 good, 0.79 to ≥ 0.70 acceptable, and 0.69 to 0.60 questionable.

Open-ended questions inquiring why the respondents answered the way they did accompanied every single question. This allowed the participants to uncover their line of thinking and permitted a more comprehensive understanding of the phenomenon being studied.

7.6.5. Behavioural intentions questionnaire (Appendix Q)

The questionnaire covered the students’ readiness and expectations to enter different types of social relationships with an individual with TS, informed by the Shared Activities Questionnaire – Revised (SAQ –R) (Ranson & Byrne, 2014), which has been written for a USA population. Questions therefore had to be modified to be more appropriate for a UK adolescent group. Focus groups from study One also served to identify relationship situations relevant to the everyday lives of adolescents and their importance to adolescents.

Instead of using a five-point Likert scale that assessed participants' willingness to socialise with individuals with TS (i.e. Would you?), a five-point likelihood scale (i.e. How likely is it that you would?) was utilised in order to increase the predictive power of an individual's behaviour (1=not likely at all, 5=very likely) (Warshaw & Davis, 1984). As opposed to scales that only assess intentions, a person's perceived likelihood of engaging in a specific behaviour was considered a more proximate predictor of behaviour (Courneya & McAuley, 1994; Sheppard, Hartwick, & Warshaw, 1988). In this way, it was expected that when answering questions pertaining to behavioural expectations people would take into consideration influential variables that might impact one's behaviour which might not have been considered when expressing intentions e.g. past behaviour, impact of the behaviour, peer pressure etc.

The questionnaire was composed of 10 statements, with a possible total score can ranging from 10 to 50 with higher scores revealing more favourable behavioural intentions. Internal reliability was reported at a Cronbach's alpha of 0.915 (excellent), with test-retest reliability of 0.77 (acceptable). Internal reliability was reported according to the George & Mallery, 2003 criteria: ≥0.90 excellent, 0.89 to ≥0.80 good, 0.79 to ≥ 0.70 acceptable, and 0.69 to 0.60 questionable.

Three activity domains were represented in the questionnaire: academic (e.g. work on an assignment with an individual with TS), general social (e.g. sit next to a person with TS in class) and active recreational (e.g. go to the cinema with an individual with TS). The different statements varied in the extent to which they required interactive behaviour and intimacy. Both group and dyadic activities were included within and outside school settings.

The behavioural intention questionnaire included open-ended questions in order to gain more in-depth understanding of the reasoning behind the participants' answers.

7.6.6. Film assessment (Appendix R)

In order to evaluate the feelings, the participants had developed towards the main character of the film, a short questionnaire was adapted from the study of Riiterfeld & Jin (2006). It comprised of five statements to assess empathy towards the main protagonist and were rated on a 5-point Likert scale anchored by Completely Disagree to Completely Agree, with higher scores showing greater sympathy. This questionnaire was only issued in data collection point T2, immediately after the intervention and was only be administered to the experimental group. The reliability of the questionnaire on the film “Front of the class” had not been tested.

7.6.7. Intervention evaluation (Appendix S)

A feedback questionnaire was developed to assess how helpful the participants considered the workshop. It also aimed to evaluate which of the components of the workshop the students considered more memorable and interesting. This questionnaire was issued in data collection point T2, immediately after the intervention and was only administered to the intervention group. Five statements assessing individuals’ views were rated on a 5 point Likert scale anchored by Completely Disagree to Completely Agree. An open-ended question which allows the participants to make any other comments they wish was also added.

7.7. Pilot study

The short stimulus video and the accompanying questionnaires were piloted on a group of five Year Ten adolescents from the school who did not participate in the final study. The adolescents were given a chance to evaluate the comprehensibility of the questionnaires and asked to note down anything that they didn’t understand. More specifically they were asked to indicate a) whether they knew what all the words meant b) whether all the questions made sense c) whether they understood what all the questions were asking d) whether they understood how to mark their answers.

All of the participants indicated that the questionnaire was comprehensible and one adolescent specifically commented that “it was easy to understand”.

The pilot study was also conducted in order to avoid a ceiling effect in the Knowledge Questionnaire, namely the questions being so easy that all the students answered them correctly or alternatively, a floor effect, that is, -too difficult for the students. The percentage of answering correctly on each question ranged from 25% to 75%.

The five adolescents were asked not to share the content of the questionnaires with the potential study two participants, so as to avoid them being prepared for the questions.

7.8. Data analysis

All quantitative data were entered and analysed using SPSS version 23, using ID numbers and not participants names. The assumption of normality was checked by calculations of skewness and Kurtosis, Kolmogorov Smirnov test and box-plots. Baseline, post-test and follow-up scores for all measures were compared for the two groups using non-parametric tests (due to outliers). For all statistical analyses the .05 alpha level was used for statistical significance.

The answers to the open-ended questions were analysed using inductive thematic analysis and the six-stage coding framework outlined by Braun & Clarke (2006). This was facilitated by NVivo (version 11), a software tool for managing qualitative research data. Thematic analysis was preferred over other qualitative approaches (e.g., grounded theory, case study) because it was our intention to compare the themes that emerged before the intervention to the ones that were developed after the intervention in order to interpret how and if the intervention impacted adolescents' reasoning in the experimental group.

During the first phase, every written response to any of the open-ended questions was copied to an NVivo file by the researcher and read over several times in order for her to immerse herself in the data before the coding commenced. In the next stage both the researcher and her first supervisor independently coded over each line of the data at a semantic level for the major ideas expressed. Next the codes were organised into main themes. This involved discarding some codes. Phase four included the merge of some themes in order for wider and more cohesive themes to be formed. In this phase of the analysis some themes that did not have enough data to support them were disregarded. Finally, phase five and six consisted of further defining and naming the themes as well as selecting quotes which clearly represented the identified issues within each theme.

In order to ensure inter-thematic reliability the researcher and her first supervisor compared codes, categories and emerging subthemes. Elongated discussions between the two followed until saturation, yielding the final themes and subthemes.

Inferences made from the qualitative and quantitative data set will be synthesised to meta-inferences at the end of the study.

7.9. Quantitative results

All quantitative data were entered and analysed using SPSS version 23, using anonymous ID numbers. The assumption of normality was checked using calculations of skewness and Kurtosis and Kolmogorov Smirnov test as well as box-plots. In most cases the data was normally distributed, but due to outliers, non-parametric tests were considered more appropriate. Baseline, post-test and follow-up scores for all measures were compared for the two groups using non-parametric tests: Mann-Whitney U tests were used to analyse the differences between the two groups for the three dependant variables at baseline, post-test, and follow-up intervals; six separate Friedman's analysis of variance (non-parametric equivalent of ANOVA) tests were conducted to assess the differences in each of the three dependent variables (Knowledge, Attitudes, and Behavioural Intentions) for both the intervention and the control group. Following the Friedman's, post hoc multiple comparison tests (Wilcoxon Signed Ranks tests) with Bonferroni's correction were then performed to determine where significant differences existed. The effect size of all the non-parametric tests was calculated manually following the formula $=Z/\sqrt{N}$, suggested by Fields (2009). Cohen's (1992) effect size estimates have been used to interpret the meaning of the r score: 0.10 small, 0.30 medium, 0.50 large.

For all statistical analyses the 0.05 alpha level was used for statistical significance except where adjustments to control the Type I error rate are specified.

7.9.1. Sample characteristics

A total of 59 students from Year Ten participated in the project (Table 6, p. 189). The mean age of the sample was 14.70 (SD=0.50, age range 14-16 years). The gender split was 54.20% females: 45.80% males.

The majority of the students (76.30%) identified themselves as being white British. Of the remainder, 15.30% reported being white of different background, 1.70% black British, 3.40% mixed background and 3.40% other background without providing any other information (Table 7, p. 189).

Table 6 - Demographic characteristics of study participants.

	Total	Male	Female
Sample Number	59	27	32
Age range	14–16	14–15	14–16
Mean Age	14.70	14.74	14.66
Standard Deviation	0.50	0.45	0.54

Table 7 - Ethnicity of study participants.

Ethnicity	White British	Any other white background	Black British	Mixed Background	Other
Number of participants (%)	45 (76.30%)	9 (15.30%)	1 (1.70%)	2 (3.40%)	2 (3.40%)

Table 8 - Study participants' first language.

First Language	**English**	**Latvian**	**Portuguese**	**Polish**	**Russian**	**Turkish**
Number of Participants (%)	45 (76.30%)	2 (3.40%)	2 (3.40%)	2 (3.40%)	1 (1.70%)	1 (1.70%)
First Language	**Romanian**	**Bulgarian**	**Shona**	**American**	**Other**	
Number of Participants (%)	1 (1.70%)	1 (1.70%)	1 (1.70%)	1 (1.70%)	1 (1.70%)	

The majority of participants (76.30%) reported English as their first language (Table 8, p. 189), whilst 21.10% stated their first language as other than English (3.40% Portuguese, 3.40% Latvian, 3.40% Polish, 1.70% Slovak, 1.70% Russian, 1.70% Turkish, 1.70% American, 1.70% Romanian, 1.70% Bulgarian, and 1.70% Shona). One student did not answer the relevant question.

Table 9 - Characteristics of intervention and control group.

Group	N (%)	Gender	N (%)	Personal Contact with individuals with TS/IDD or MI	N (%)
Intervention Group	30 (50.80%)	**Females** **Males**	19 (63.30%) 11 (36.70%)	**No personal contact with TS/IDD/MI** **Personal contact with TS/IDD/MI**	7 (23.33%) 23 (76.77%)
Control Group	29 (49.20%)	**Females** **Males**	13 (44.81%) 16 (55.29%)	**No personal contact with TS/IDD/MI** **Personal contact with TS/IDD/MI**	3 (10.30%) 26 (89.70%)

To obtain meaningful results regarding previous contact with individuals with stigmatising conditions, data was pooled to delineate two categories: no personal contact with individuals with stigmatising conditions (Tourette's syndrome (TS), Intellectual and/or Developmental Disabilities (IDD) or Mental Illness (MI)); and previous personal contact with individuals with stigmatising conditions.

From the 59 participants (Table 9, p. 190), 49 (83.10%) reported that they had experienced some form of personal contact with individuals with stigmatising conditions whilst 10 (16.90%) had never had any personal contact with individuals with TS, IDD or MI.

The participating school classes were allocated either to the intervention or the control group (see Methodology chapter). The Pearson Chi-Square indicated that both genders were represented equally in the intervention and control group (x(1)=2.04, p=0.15), as were individuals who had no personal contact or had personal contact with people with stigmatising conditions (x(1)=1.18, p=0.18).

7.9.2. Exploratory data analysis

The normality of distribution for the three questionnaires (Knowledge, Attitudes, and Behavioural Intentions) at the three time points (Baseline (T1), Post-Intervention (T2) and

Follow-up (T3)) were checked via the visual inspection of histograms, calculations of both skewness and kurtosis, and the use of Kolmogorov-Smirnov of normality and box plots.

7.9.2.1. Normality of distribution of variables

7.9.2.1.1. Distribution of total scores on the knowledge, attitude, and behavioural intentions questionnaires (Baseline T1)

The Knowledge Questionnaire total scores showed a normal distribution (see Figure 9, p. 351, Appendix T), with a skewness of -0.52 (SE= 0.31) and kurtosis of -0.92 (SE= 0.62). The one sample Kolmogorov-Smirnov test however rejected the null hypothesis that the Knowledge scores were normally distributed D (59)=0.12, n=59, p=0.04. The Attitudes Questionnaire scores (see Figure 10, p. 352, Appendix T) showed a normal distribution, with a skewness of -0.12 (SE=0.31) and a kurtosis of -0.30 (SE=0.61). The one sample Kolmogorov-Smirnov test also showed that the Attitudes scores were normally distributed D (59)=0.08, n=59, p= 0.20. The Behavioural Intentions Scores showed a normal distribution (see Figure 11, p. 353, Appendix T), with a skewness of –0.19 (SE=3.11) and kurtosis of 0.23 (SE=0.61). The one sample Kolmogorov-Smirnov test was also found non-significant D(59)=0.01, n=59, p=0.20, so the null hypothesis that the Behavioural Intentions scores were normally distributed was not rejected.

7.9.2.1.2. Distribution of Total Scores on the Knowledge, Attitude, and Behavioural Intentions Questionnaires (Post-Intervention T2)

The Knowledge scores (see Figure 12, p. 354 Appendix T) showed a normal distribution with a skewness of -0.31 (SE=0.31) and kurtosis of -0.94 (SE=0.61). When the one sample Kolmogorov-Smirnov test was calculated however, the null hypothesis that the Knowledge scores were normally distributed was rejected D (59)=0.15, n=59, p=0.001. Histogram displaying the distribution of Attitude Questionnaire scores after the intervention can be seen in Figure 13, (p.355) in the Appendix T. Distribution of Attitude Scores were found to be normally distributed (with skewness -0.07 (SE=0.31) and kurtosis -0. 66 (SE=0.61). The one sample Kolmogorov-Smirnov test also indicated that the Attitude scores did not deviate significantly from the norm D(59)=0.05, n=59, p=0.20. The scores on the Behavioural Intention Questionnaire (see Figure 14, p. 356, Appendix T) at the post-intervention stage were found to be normally distributed with skewness of -0.50 (SE= 0.31) and kurtosis of 0.02 (SE=0.61). The Kolmogorov-Smirnov were also found to be non-significant (D(59)=0.09, n=59, p=0.20), so the null hypothesis that the data were normally distributed was not rejected.

7.9.2.1.3. Distribution of Total Scores on the Knowledge, Attitude and Behavioural Intentions Questionnaires (Follow-up, T3)

Even though the skewness and kurtosis of the total scores on the Knowledge Questionnaire (see Figure 15, p. 357, Appendix T) at the follow up stage show that there is normality of distribution (skewness: 0.02 (SE=0.34); kurtosis -0.12 (SE=0.67)), an observation of the histogram and the calculations of the Kolmogorov-Smirnov test show that the data are not normally distributed. The Kolmogorov-Smirnov test was found to be significant D(49)=0.135, n=49 p=0.03; thus the null hypothesis that the data was normally distributed was rejected. Post-intervention Attitude Questionnaire scores were also examined for normality of distribution (see Figure 16, p. 358, Appendix T). Distribution of Attitudes scores were found to be normally distributed (with skewness -1.17 (SE=0.34) and kurtosis 5.13 (SE=0.67)). The one sample Kolmogorov-Smirnov test were also found to be non-significant (D(49)=0.12, n=49, p=0.09), so the null hypothesis that the data were normally distributed was not rejected. The skewness and kurtosis of the total scores on the Behavioural Intention Questionnaire show that there is normality of distribution (skewness -0.40 (SE= 0.34); kurtosis: 0.22 (SE= 0.67) (see Figure 17, p. 359, Appendix T). This was confirmed by the Kolmogorov-Smirnov test. The Behavioural Intentions Scores were found to be non-significant (D(49)=0.11, p= 0.15), so the null hypothesis that the data were normally distributed was not rejected.

7.9.3. Outliers

The presence of outliers was examined via box plots. Outliers were identified in many of the measures (see Figures 18-21, p. 360-363, Appendix T).

Many more outliers were apparent when the groups were separately analysed (Figures 6–8, p. 194-196). Due to the small sample size and the study's aim to identify the difference between three time points, it was decided not to remove the outliers. Therefore, non-parametric testing was used to examine the study's hypotheses.

7.9.4. Non-parametric between group testing

A Mann-Whitney U (non-parametric test equivalent to an independent t-test used for small sample sizes and when data is not normally distributed) was used to test for differences between the control and intervention groups for all independent variables at T1, T2 and T3. Differences between female and males and participants that had and did not have contact with people with stigmatising conditions were also explored.

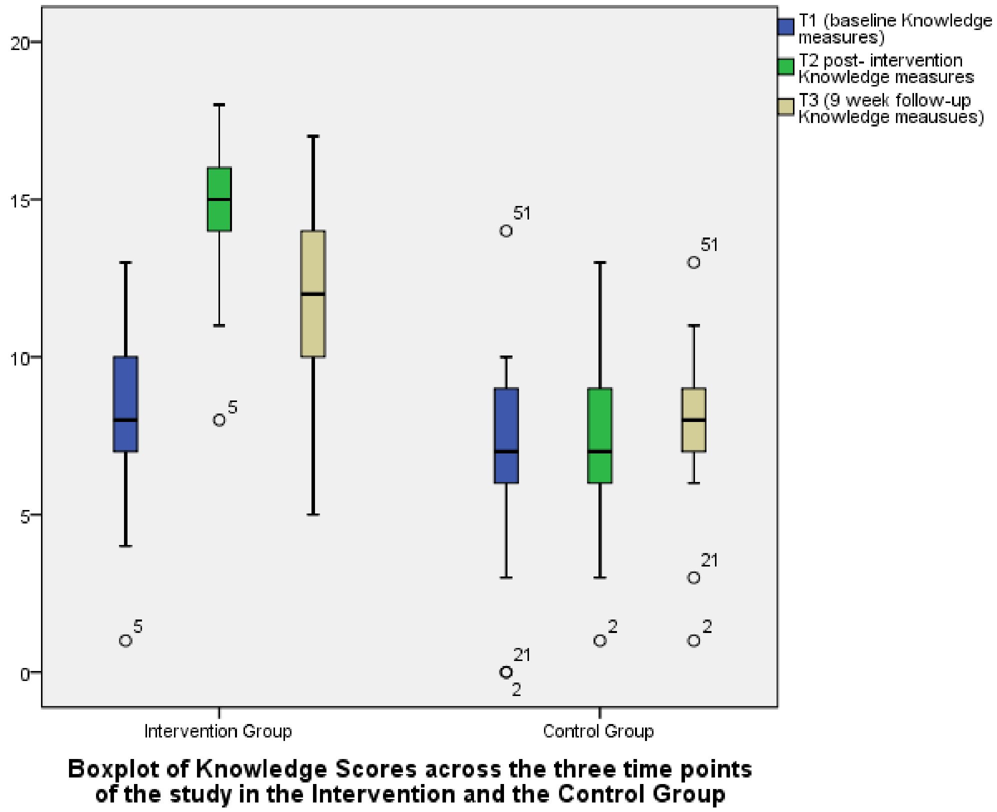

Figure 5 - Differences in knowledge scores across the different points of the intervention in the intervention and control group.

7.9.4.1. Baseline stage

The control group and intervention group did not differ significantly (Table 10, p. 196) to the distribution of their baseline responses on Knowledge (U=401.50, z=-0.51, p=0.61, r=-0.07), Attitudes (U=397, z=-0.58, p=0.56, r=-0.07) and Behavioural Intentions (U= 416.5, z=-0.28, p=0.78, r=0.04).

Similarly, gender (Table 11, p. 198) did not appear to impact on Knowledge (U= 400.50, z=-0.48 p=0.63, r=-0.06), Attitude (U=431, z=-0.01 p=0.99, r=-0.02) or Behavioural Intentions (U= 420, z=-0.17, p=0.86, r=-0.02) and so the findings did not support the initial hypothesis that gender affects adolescents' attitudes and willingness to engage in different types of social relationships with peers with TS.

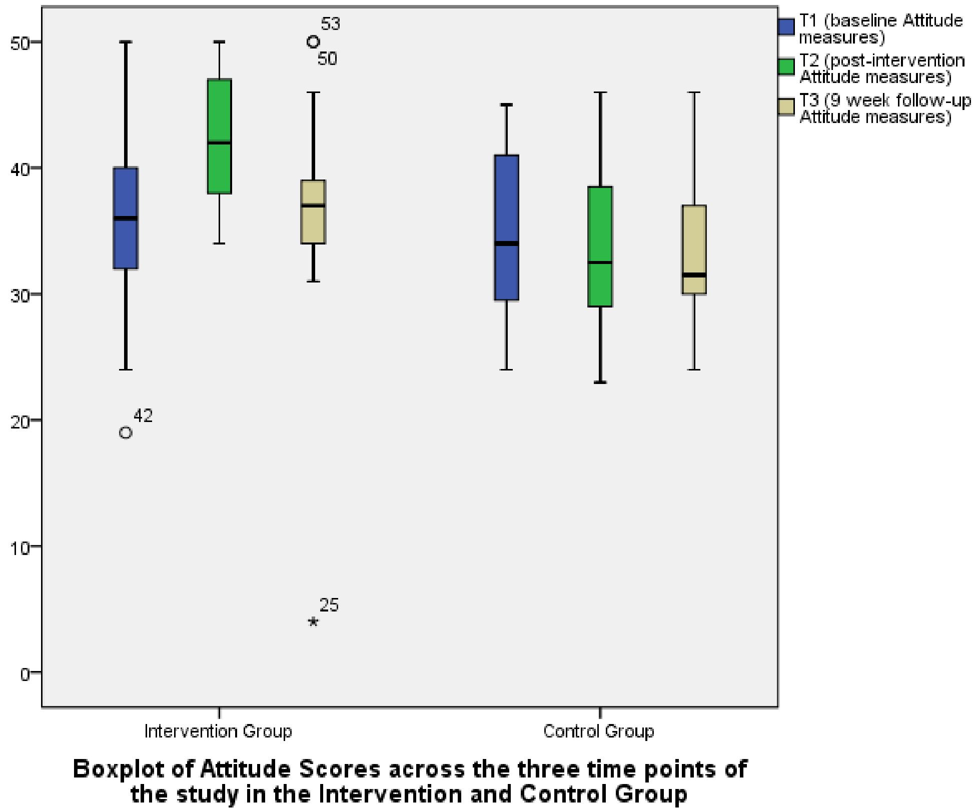

Figure 6 - Differences in attitude scores across the different time points of the study in the intervention and control group.

Previous personal contact with an individual with a stigmatising condition (TS/IDD/MI) was also not found to exert an impact on Knowledge (U= 158.50, z=-1.76, p=0.78, r=-0.23) (see Table 12, p. 199). However, familiarity was found to influence Attitudes towards individuals with TS (U= 115.50, z=-2.62, p=0.01, r=-0.34) and Behavioural Intentions (U=138.50, z=-2.16, p=0.03, r=-0.28). Students that had previous contact with individuals with stigmatising conditions were found to have better attitudes (Median=37.00) and Behavioural Intentions (Median=33.00) towards their peers with TS in comparison to their counterparts (Median=29.50 and Median 27.00, respectively). These findings confirmed the hypothesis that people who have had personal contact with individuals with stigmatising conditions have more positive attitudes towards peers with TS and are less compelled to seek social distance from them.

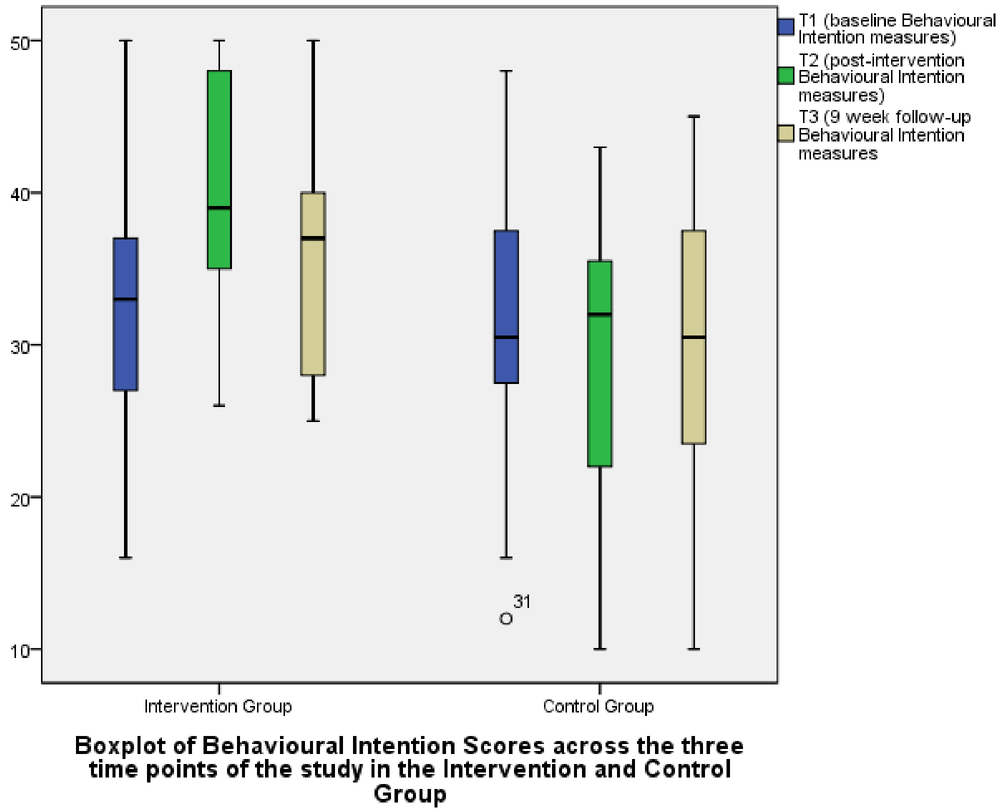

Figure 7 - Differences in behavioural intentions scores across the different time points of the study in the intervention and control group.

Table 10 - Comparison of intervention group with control group in all variables at pre-intervention stage.

Variable	N	Mean (SD)	Median	Range (min–max)	Mann-Whitney U test					
					M Rank	Sum ranks	U	Z	p	r
Intervention Group										
Knowledge scores (pre-intervention)	30	7.73 (2.53)	8.00	12.00 (1–13)	31.12	933.50	401.50	-0.51	0.61	-0.07
Control Group										

Knowledge scores (pre-intervention)	29	7.41 (3.12)	7.00	14.00 (0.00–14.00)	28.84	836.50				
Intervention Group										
Attitude scores (pre-intervention)	30	36.27 (6.90)	37.50	31.00 (19.00– 50.00)	31.27	938.00	397.00	-0.58	0.56	-0.07
Control Group										
Attitude scores (pre-intervention)	29	35.38 (6.22)	35.00	21.00 (24.00–45.00)	28.69	832.00				
Intervention Group										
Behavioural Intention scores (pre-intervention)	30	32.00 (8.70)	32.50	35.00 (15.00–50.00)	29.38	881.50	416.50	-0.28	0.78	-0.04
Control Group										
Behaviour Intentions scores (pre-intervention)	29	32.34 (7.91)	31.00	36.00 (12.00–48.00)	30.64	888.50				

Table 11 - Comparison of females and males in all variables at pre-intervention stage.

Variable	N	Mean (SD)	Median	Range (min-max)	Mann-Whitney U test					
					M Rank	Sum ranks	U	Z	p	r
Knowledge										
Females	32	7.28 (2.88)	7.00	13.00 (0.00–13.00)	29.02	928.50	400.50	-0.48	0.63	-0.06
Males	27	7.93 (2.74)	8.00	14.00 (0.00–14.00)	31.17	841.50				
Attitudes										
Females	32	35.69 (5.94)	36.50	27.00 (19–46)	29.97	959.00	431.00	-0.01	0.99	-0.02
Males	27	36 (7.27)	37.00	26.00 (24–50)	30.04	811.00				
Behavioural Intentions										
Females	32	32.19 (8.29)	33.00	33.00 (15–48)	30.36	971.50	420.50	-0.17	0.86	-0.02
Males	27	32.15 (8.36)	31.00	38.00 (12.00–50.00)	29.57	798.50				

Table 12 - Comparison of participants that have had personal contact with individuals with stigmatising conditions with participants that have not in all variables at pre-intervention stage.

Variable	N	Mean (SD)	Median	Range (min-max)	Mann-Whitney U test					
					M Rank	Sum ranks	U	Z	p	r
Knowledge										
Contact	49	7.88 (2.56)	8.00	14.00 (0.00–14.00)	31.77	1556.50	158.50	-1.76	0.08	-0.23
No contact	10	6.10 (3.63)	7.00	13.00 (0.00–13.00)	21.35	213.50				
Attitudes										
Contact	49	36.82 (6.25)	37.00	31.00 (19.00–50.00)	32.64	1599.50	115.50	-2.62	0.01	-0.34
No contact	10	31.00 (5.91)	29.50	19.00 (24.00–43.00)	17.05	170.50				
Behavioural Intentions										
Contact	49	33.29 (7.82)	33.00	38.00 (12.00–50.00)	32.17	1576.50	193.50	-2.16	0.03	-0.28
No contact	10	26.70 (8.48)	27.00	26.00 (15.00–41.00)	19.35	193.50				

7.9.4.2. Post-intervention stage (T2)

Post-implementation of the workshop on the intervention group resulted in significant and large differences (r=-0.78) between the control group, who had statistically less accurate Knowledge about TS (Median=8.00) (U= 37.00, z=-6.06, p<0.001) and the intervention group (Median= 15) (see Table 13, p. 200). The intervention group also had statistically less prejudicial attitudes (Median=42.00) towards individuals with TS (U=149.00, z=-4.34, p<0.001) in comparison to the control group (Median=33.00); this difference between the control and intervention group being large (r=-0.56). Finally, the adolescents' Behavioural Intentions scores at the post-intervention stage for the control group (Median=32.00) were statistically worse (U=172.00, z=-3.99, p<.001) than the Behavioural Intentions scores for the intervention group (Median=40.00). The difference between the control and intervention groups was found to be medium (r=-0.44).

Table 13 - Comparison of intervention group with control group in all variables at post-intervention stage.

Variable	N	Mean (SD)	Median	Range (min-max)	Mann-Whitney U test					
					M Rank	Sum of ranks	U	Z	p	r
Intervention Group										
Knowledge scores (post-intervention	30	14.43 (2.42)	15.00	10.00 (8.00-18.00)	43.27	1298.00	37.00	-6.06	<0.001	-0.78
Control Group										
Knowledge scores (post-intervention	29	7.79 (2.89)	8.00	12.00 (1.00-13.00)	16.28	472.00				
Intervention Group										
Attitude scores (post-intervention	30	42.10 (5.38)	42.00	16.00 (34.00-50.00)	39.53	1186.00	149.00	-4.34	<0.001	-0.56
Control Group										

Attitude scores (post-intervention	29	34.07 (6.21)	33.00	23.00 (23.00–46.00)	20.14	584.00				
Intervention Group										
Behavioural Intention scores (post-intervention	30	40.50 (7.58)	40.00	24.00 (26.00–50.00)	38.77	1163.00	172.00	-4.00	<0.01	-0.44
Control Group										
Behaviour Intentions scores (post-intervention	29	29.76 (9.19)	32.00	33.00 (10.00–43.00)	20.93	607.00				

7.9.4.3. Follow-up stage (T3)

Mann-Whitney U non-parametric analysis showed that at the follow up stage of the study, participants in the intervention group (Median=12) scored significantly different than adolescents in the control group in relation to Knowledge (U=123.50, z=-3.55, p<0.001, r=-0.51) (see Table 14, p. 202). The same was observed for Attitudes between control group and intervention group (Median=37.00) (U=167.50, z=-2.65, p=0.01, r=-0.38). Also, Behavioural Intentions in the intervention group (Median=37.00) differed significantly from the Behavioural Intentions of the control group (Median=30.50) at the follow-up stage (U=200.00, z=-2.01, p=0.04, r=-0.29).

Table 14 - Comparison of intervention group with control group in all variables at 9-weeks follow-up stage.

Variable	N	Mean (SD)	Median	Range (min-max)	Mann-Whitney U test					
					M Rank	Sum ranks	U	z	p	r
Intervention Group										
Knowledge scores (follow-up)	25	11.40 (3.27)	12.00	12.00 (5.00–17.00)	32.06	801.50	123.50	-3.55	<0.001	-0.51
Control Group										
Knowledge scores (follow-up)	24	7.87 (2.47)	8.00	12.00 (1.00–13.00)	17.65	423.50				
Intervention Group										
Attitude scores (follow-up)	25	37.20 (8.66)	37.00	46.00 (4.00–50.00)	30.30	757.50	167.50	-2.65	0.01	-0.38
Control Group										
Attitude scores (follow-up)	24	33.42 (5.78)	31.50	22.00 (24.00–46.00)	19.48	467.50				
Intervention Group										
Behavioural Intention scores (follow-up)	25	35.24 (7.12)	37.00	25.00 (25.00–50.00)	29.00	725.00	200.00	-2.01	0.04	-0.29
Control Group										
Behaviour Intentions scores (follow-up)	24	29.87 (9.26)	30.50	35.00 (10.00–45.00)	20.83	500.00				

7.9.5. Non-parametric within group testing

Having established that a significant difference exists between the intervention and the control group at T2 and T3 time points in all of the variables, a Friedman's non-parametric test was used to determine if statistically significant differences exist between the three time points for the intervention and the control group separately on the key variables. The Friedman test is the non-parametric alternative to a repeated measures of variance (ANOVA), which is the test of choice for larger sample sizes with normally distributed data. The Friedman test examines the ranks of the data from each time period of data collection to determine whether the variables share the same underlying continuous distribution. Wilcoxon signed-ranked tests were applied as post hoc tests to determine which time points differed across time for each of the two conditions.

7.9.5.1. Intervention group

7.9.5.1.1. Knowledge

The Friedman's test revealed that there was a statistically significant difference for Knowledge for the participants in the intervention condition at the three different time points (T1, T2, T3), $\chi2$ (2)=40.97, p<0.001 (Table 15, p. 204). Post hoc analysis with Wilcoxon signed-rank tests was conducted with a Bonferroni correction applied, resulting in a significance level set at p<0.017. Median scores for knowledge for T1, T2 and T3 were 8.00 (6.50–10.00), 15.00 (14.00–16.00) and 12.00 (9.00–14.00), respectively. There was a significant increase in Knowledge between T1 and T2 (z=-4.80, p<0.001, r=-0.62) and a significant decrease in Knowledge between T2 and T3 (z=-4.10, p<0.001, r=-0.55). Despite the overall reduction in knowledge between post-test and follow-up, there was still a statistically significant increase in knowledge between T1 and T3, the effect size of which is still large (z=-3.85, p<0.001, r=-0.52) (Table 16, p. 204).

Table 15 - Results of the Friedman ANOVA for the experimental group on the knowledge questionnaire.

Variable	N	χ^2	Df	p
Knowledge	25	40.97	2	<0.001

Table 16 - Paired Wilcoxon signed rank test for experimental groups' knowledge at different time points.

Variable	N	z-score	p-value	r
Knowledge Scores pre-test vs	30	-4.80	<0.001	-0.62
Knowledge Scores post-test	30			
Knowledge scores post-test vs	30	-4.10	<0.001	-0.55
Knowledge scores follow-up	25			
Knowledge scores pre-test vs	25	-3.85	<0.001	-0.52
Knowledge scores follow-up	30			

These results indicate that the two-hour anti-stigma program had a positive effect on participants' knowledge of TS. Furthermore, while the knowledge decreased in the weeks after the intervention, it maintained significantly above baseline measures in the follow-up 9 weeks later. This indicates the long-term effectiveness of the intervention in terms of Knowledge about TS.

A closer look at the distribution of responses to individual items of the measures at the three different time points of the study can be seen in Table 17 (p. 206). The majority of the students who received the intervention appeared to have a basic understanding of TS and in terms of awareness, most individuals at T1 did not espouse the stereotypical belief that everyone who has Tourette’s syndrome swears (86.70%). All of the intervention group believed that the severity of the tics deteriorates with age (100%) and that TS is not a hereditary condition (90.00%). Furthermore, 60.00% of the respondents in the intervention group believed that TS is caused by really bad childhood experiences. It is also important to mention that the majority of adolescents in the study believed that pupils with TS would be better suited in schools for children with similar issues, or special schools, or should be home schooled (86.70%) with only 13.30% believing that an individual with TS should be educated in a mainstream school.

At T2 whilst knowledge about TS improved significantly for the intervention group (see Table 17), approximately half of these participants reported that they thought that everyone who has tics has Tourette’s syndrome, and 60% of the participants believed that individuals with TS can never control their tics.

A gradual worsening in knowledge was detected in the 9-weeks follow-up. More noteworthy is the lingering belief that individuals with TS would not be suited for mainstream education.

Table 17 - Frequency of correct answers on the knowledge questionnaire amongst the intervention group at different time points.

Question Possible answers[4]	Baseline n=30 (% Correct)	Post-test n=30 (% Correct)	9-weeks follow-up n=25 (% Correct)
Tourette's syndrome is caused by really bad experiences during childhood. (**Not true**/ True/Don't Know)	n=12 (40.00%)	n=26 (86.70%) (z=-3.30, p<0.001) *	n=19 (76.00%) (z=-2.89, p=0.004) *
Everyone who has Tourette's syndrome swears. (**Not true**/ True/Don't Know)	n=26 (86.70%)	n=28 (93.30%) (z=0.82, p=0.14)	n=21 (84.00%) (z=-0.38, p=0.70)
You can catch Tourette's syndrome from other people. (**Not true**/ True/Don't Know)	n=22 (73.30%)	n=28 (93.30%) (z=2.45, p=0.01) *	n=23 (92.00%) (z=-1.41, p=0.16)
During adulthood, the tics get worse. (**Not true**/ True/Don't Know)	n=0 (0%)	n=20 (66.70%) (z=4.47, p<0.001) *	n=7 (29.20%) (z=-2.65, p<0.01) *
Lots of people who have Tourette's syndrome have other disorders as well. (Not true/ **True**/Don't Know)	n=4 (13.30%)	n=23 (76.70%) (z=4.36, p<0.001) *	n=9 (36.00%) (z=-2.33, p=0.02) *
Tourette's syndrome runs in families. (Not true/ **True**/Don't Know)	n=3 (10.00%)	n= 27 (90.00%) (z=4.60, p<0.001) *	n= 12 (48.00%) (z=-3.32, p<0.01) *
Everyone who has tics has Tourette's syndrome. (**Not true**/ True/Don't Know)	n=11 (36.70%)	n=16 (53.30%) (z=0.41, p=0.68)	n=18 (72.00%) (z=-2.31, p=0.02) *
People with Tourette's syndrome can never control their tics. (**Not true**/ True/Don't Know)	n=7 (23.30%)	n=9 (30.00%) (z=0.63, p=0.53)	n=9 (37.50%) (z=-2.00, p=0.05)
Tourette's syndrome is (a mental illness**, a condition that has do with the brain and the nervous system**, a physical disability)	n=18 (68.00%)	n=29 (96.70%) (z=2.45, p=0.14)	n=22 (88.00%) (z=1.00, p=0.32)
The ratio of TS in the UK (1/10, **1/100**, 1/1000)	n=16 (53.30%)	n=25 (83.30%) (z=1.73, p=0.08)	n=21 (84.00%) (z=1.13, p=0.26)
Symptomology in order to be diagnosed with TS (swear uncontrollably, **have both movement tics and tics that produce sounds**, have tics that produce sounds and swear uncontrollably	n=24 (80.00%)	n=30 (100%) (z=2.45, p=0.14)	n=22 (88.00%) (z=1.13, p=0.26)
The tics usually start (before the age of 3, **before the age of 18**, after the age of 18)	n=20 (66.70%)	n=25 (83.30%) (z= 1.39, p=0.17)	n=17 (68.00%) (z=-0.33, p=0.74)
What kind of people does Tourette's syndrome affect? (Only individuals with special educational needs, only individuals who crave attention, **Can potentially affect anyone**)	n=24 (80.00%)	n=29 (96.70%) (z=2.00, p=0.05)	n=20 (80.00%) (z=0.00, p=1.00)

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[4] The correct answers are highlighted.

How long does a person have to have symptoms before they are diagnosed with Tourette’s syndrome? (Five years, **One year,** Six months)	n=11 (36.70%)	n=26 (86.70%) (z= -3.64, p<0.001) *	n=18 (72.00%) (z=-2.71, p<0.01) *
How is Tourette’s syndrome diagnosed? (By an MRI, By a blood test, **By the symptoms**)	n=14 (46.70%)	n=29 (96.70%) (z=-3.50, p<0.001) *	n=21 (84.00%) (z=-2.71, p<0.01) *
What is the best school environment for a teenager with Tourette’s syndrome? (Home education, **mainstream school**, school for children with similar issues)	n=4 (13.30%)	n=21 (70.00%) (z= 4.12, p<0.001) *	n=14 (56.00%) (z=-3.16, p<0.01) *
The activities and the environment an individual with Tourette’s syndrome is in may affect how often the tics occur and how intense they are. (Not true/ **True**/Don’t Know)	n=11 (36.70%)	n=22 (73.30%) (z= 3.05, p<0.001) *	n=6 (24.00%) (z=-1.26, p=0.21)
Tourette’s syndrome is most effectively treated with medication. (**Not true**/ True/Don’t Know)	n=4 (13.30%)	n=18 (60.00%) (z=3.50, p<0.001) *	n=6 (24.00%) (Z=1.00, p=0.32)

7.9.5.1.2. Attitudes

The Friedman’s test revealed that there was a statistically significant difference for attitude for the participants in the intervention group at the 3 time points (T1, T2, T3), $\chi 2$ (2)= 19.83, p<0.001 (Table 18, p. 208). Post hoc analysis with Wilcoxon signed-rank tests was conducted with a Bonferroni correction applied, resulting in a significance level set at p<0.017. Median scores for Attitudes for T1, T2 and T3 were 36.00 (31.50–41), 42.00 (37.50–47.00) and 37.00 (33.50–41.00), respectively. There was a significant improvement in attitudes towards peers with Tourette’s syndrome from baseline to post-test and a large effect size (z=-4.30, p<0.001, r=-0.55). However, this improvement significantly decreased at T3 (z=-3.24, p=0.001, r=-0.44). Furthermore, the Wilcoxon signed-rank test indicated that participants’ attitudes in the intervention condition did not significantly differ between T1 and T3 (z=-1.66, p=0.10, r=-0.22) (Table 19, p. 208).

Results indicate that, following the completion of a two-hour anti-stigma workshop about Tourette's syndrome, there was a significant effect on attitudes towards peers with Tourette's syndrome. However, this observed attitudinal improvement was not maintained at T3 and the students returned to their baseline attitude levels (see Figure 7, p. 195).

Table 18 - Results of the Friedman's test for the experimental group on the attitude questionnaire.

Variable	N	χ^2	df	p
Attitude	25	19.83	2	<0.001

Table 19 - Paired Wilcoxon signed rank test for experimental groups' attitude at different time points.

Variable	N	z-score	p-value	r
Attitude Scores pre-test vs	30	-4.30	<0.001	-0.55
Attitude Scores post-test	30			
Attitude scores post-test vs	30	-3.24	0.001	-0.44
Attitude scores follow-up	25			
Attitude scores pre-test vs	30	-1.66	0.10	-0.22
Attitude scores follow-up	25			

Students' attitude scores as frequencies and percentages of pooled responses to individual items of the questionnaire at each time point of the study are displayed in Table 20 (p. 210). Comparison to baseline measures per item using the Wilcoxon test can be seen in Table 21(p. 212).

The baseline measures suggest that adolescents with TS may be vulnerable to negative peer attitudes but concurrently the views of adolescents were not extremely stigmatising. Interestingly, 40% of the participants believed that a person with TS could not have a normal life and were likely to pity them. Approximately one in two individuals that participated in the intervention group felt that individuals with TS would be able to acquire a job that would require them to speak in public. Similarly, only half of the participants believed that an individual with TS belonged in a mainstream school irrespectively of the severity of the tics. It should be emphasised that, similarly to the study of Schulze et al. (2003), for many of the questions, the majority of respondents stated that they were unsure about their views pertaining to individuals with TS (see Table 20-21, p. 210 and p. 212). This answer does not suggest negative views, it does indicate that the study participants were not ready to embrace positive views which might indicate that the intervention was warranted.

Immediately post-intervention, all items of the attitudes scores were significantly different from the baseline ones, except for three including Benevolence; with only 40% of the intervention group disagreeing with the statement that individuals with TS would not be able to live a normal life and therefore should be pitied. Fear towards individuals with TS showed no change, possibly because only a minority reported distress towards individuals with TS before the workshop and, therefore, this item had little room for change. Finally, 66.7% of the sample gave negative responses to the idea of an individual with severe TS being in mainstream school, demonstrating segregating attitudes.

Table 20 - Attitudes of the intervention Group (n=30) per item before the intervention, after the intervention, and at the 9-weeks follow-up (valid percentages reported).

Questions using user defined criteria	Pre-intervention n=30			Post-intervention n=30			9-weeks follow-up n=25		
	Strongly agree/Agree	**Neutral**	**Disagree/ Strongly Disagree**	**Strongly agree/Agree**	**Neutral**	**Disagree/ Strongly Disagree**	**Strongly agree/Agree**	**Neutral**	**Disagree/ Strongly Disagree**
"Having a person like that in my class would be very annoying"	n=6 (20.00%)	n=10 (33.30%)	n=14 (46.70%)	n=2 (6.70%)	n=7 (23.30%)	n=21 (70.00%)	n=4 (16.00%)	n=8 (32.00%)	n=13 (52.00%)
"People with Tourette's syndrome are just like everybody else and the tics shouldn't stop *them doing what they want to do"*	n=23 (76.60%)	n=5 (16.70%)	n=2 (6.70%)	n=29 (96.70%)	n=1 (3.30%)	n=0 (0%)	n=20 (80.00%)	n=4 (16.00%)	n=1 (4.00%)
"Just because they have Tourette's you shouldn't treat them differently"	n=24 (80.00%)	n=2 (67.70%)	n=4 (13.30%)	n=29 (96.70%)	n=1 (3.30%)	n=0 (0%)	n=18 (72.00%)	n=5 (20.00%)	n=2 (8.00%)
"I feel sorry for them as they can't have a normal life"	n=12 (40.00%)	n=13 (43.30%)	n=5 (16.70%)	n=13 (43.30%)	n=5 (16.70%)	n=12 (40.00%)	n=5 (20.00%)	n=8 (32.00%)	n=12 (48.00%)
"When I see a person with Tourette's syndrome, I feel very scared"	n=3 (10.00%)	n=6 (20.00%)	n=21 (70.00%)	n=3 (10.00%)	n=7 (23.30%)	n=20 (67.70%)	n=2 (8.00%)	n=6 (24.00%)	n=17 (68.00%)
I wouldn't think of people with Tourette's syndrome any different, wouldn't look at them any different	n=18 (60.00%)	n=7 (23.30%)	n= 5 (16.70%)	n=24 (80.00%)	n=4 (13.30%)	n=2 (6.70%)	n=22 (88.00%)	n=3 (12.00%)	n=0 (0%)

“If the tics are really bad, then they should go to another school”	n=3 (10.00%)	n=13 (43.30%)	n=14 (46.70%)	n=6 (20.00%)	n=4 (13.30%)	n=20 (66.70%)	n=3 (12.00%)	n=9 (36.00%)	n=13 (52.00%)
“People shouldn't look down on a person because she has tics, they’re not going to ruin their company”	n=19 (63.30%)	n=7 (23.30%)	n=3 (10.00%)	n=28 (93.30%)	n=1 (3.30%)	n=1 (3.30%)	n=21 (84.00%)	n=3 (12.00%)	n=1 (4.00%)
“If a person with Tourette’s syndrome wanted a job to speak in front of people he *wouldn’t be able to do it”*	n=5 (16.70%)	n=10 (33.30%)	n=14 (46.70%)	n=1 (3.30%)	n=4 (13.30%)	n=25 (83.30%)	n=1 (4.00%)	n=9 (36.00%)	n=15 (60.00%)
“People with Tourette’s syndrome can do just *as well in school”*	n=26 (86.70%)	n=2 (6.70%)	n=1 (1.33%)	n=30 (100%)	n=0 (0%)	n=0 (0%)	n=21 (87.50%)	n=3 (12.50%)	n=0 (0%)

Table 21 - Differences in attitude per item. Comparison to baseline measures using Wilcoxon signed-rank test.

	Post-intervention		9-weeks follow-up	
Attitude Questions	z-score	p-value	z-score	p-value
“Having a person like that in my class would be very annoying”	-2.77	0.01 *	-0.63	0.53
“People with Tourette’s syndrome are just like everybody else and the tics shouldn't stop them doing what they want *to do”*	-2.88	0.004 *	-0.09	0.93
“Just because they have Tourette’s you shouldn’t treat them differently”	-2.67	0.01 *	-0.35	0.73
“I feel sorry for them, as they can’t have a normal *life”*	-0.99	0.32	-2.28	0.02 *
“When I see a person with Tourette’s syndrome, I feel very scared”	-0.32	0.75	-0.91	0.36
“I wouldn't think of people with Tourette’s syndrome any different, wouldn't look at them any different”	-2.37	0.02 *	-2.81	0.005 *
“If the tics are really bad, then they should go to another school”	-1.05	0.29	-0.67	0.50
“People shouldn't look down on a person because she has tics, they’re not going to ruin their company”	-3.21	0.001 *	-1.72	0.08
“If a person with Tourette’s syndrome wanted a job to speak in front of people *he wouldn’t be able to do it”*	-3.14	0.002 *	-0.29	0.77
“People with Tourette’s syndrome can do just as well in school”	-2.97	0.003 *	-0.48	0.63

At T3 all of the Attitude items returned to baseline scores, with only two exceptions; although the workshop did not produce an immediate effect on reducing benevolent attitudes, it was effective in reducing it 9 weeks later. However, it should be noted that the percentage of people that were unsure or would induce pity on peers with TS them remained high.

7.9.5.1.3. Behavioural Intentions

The Friedman's test revealed that there was a statistically significant difference for behavioural intentions (Table 22, p. p. 214) for the participants in the intervention condition at the 3 different time points (T1, T2, T3), $\chi 2$ (2)=15.26, p<0.001. Post hoc analysis with Wilcoxon signed-rank tests was conducted with a Bonferroni correction applied, resulting in a significance level set at p<0.017. Median scores for Behavioural Intentions for T1, T2 and T3 were 33.00 (27–37), 39.00 (34–48) and 37.00 (28–40), respectively. There was a significant improvement in the participants' intention to engage with their peers with Tourette's syndrome from pre-test to post-test (z=-4.27, p<0.001, r=0.55). This improvement, however, significantly decreased at the 9-weeks follow-up (z=-3.33, p=0.001, r=-0.45). Furthermore, the Wilcoxon signed-rank test indicated that participants' attitudes in the intervention condition did not significantly differ between T1 and T3 (z=-1.66, p=0.06, r=-0.25). Namely, the participants' behavioural intentions returned to their baseline levels 9 weeks after the intervention had been implemented (Table 23, p. 214).

The results suggest that the intervention was effective in reducing the participants' desired social distance levels towards individuals with TS. However, the positive effect of the workshop could not be maintained 9 weeks after the intervention had been implemented (Table 23).

Students' behavioural intention scores as frequencies and percentages of pooled responses to individual items of the questionnaire at each time point of the study are displayed in Table 24 (p. 218). Comparison to baseline measures per item using the Wilcoxon test can be seen in Table 25 (p. 218).

Table 22 - Results of the Friedman's test for the experimental group on the behavioural intention questionnaire.

Variable	N	χ^2	df	p-value
Behavioural Intentions	25	15.26	2	<0.001

Table 23 - Paired Wilcoxon signed rank test for experimental groups' behavioural intentions at different time points.

Variable	N	z-score	p-value	r
Behavioural Intention Scores pre-test vs	30	-4.27	<0.001	-0.55
Behavioural Intention Scores post-test	30			
Attitude scores post-test vs	30	-3.33	0.001	-0.45
Attitude scores follow-up	25			
Attitude scores pre-test vs	30	-1.90	0.06	-0.25
Attitude scores follow-up	25			

As can be observed in Table 24, participants were uncertain about their behavioural intentions at the pre-intervention stage. This option does not adamantly suggest segregating views but, at the same time, indicates that the participants were not ready to engage with individuals with TS.

Although a specific trend cannot be traced in the students' Behavioural Intentions, it appears that they were more hesitant to engage in social and recreational activities which entailed public exposure. Indicatively, although 60% asserted that they would have a person with TS as a close friend only 43% would go to a shopping centre with them and only 20% of the participants said they would go to the cinema with them. Participants were also less willing to collaborate with students with TS in academic activities within school settings with only 30% of the intervention group reporting that they would want to work on an assignment with a person with TS. This might be linked to their lingering belief that the educational needs of an individual with TS might not be met within a mainstream school. Finally, greater hesitancy to include an individual with TS was observed in activities with an intimate overtone. Only 20% of the participants were willing to date an individual with TS.

An increased willingness to interact with people with TS was observed post-intervention with 83.30% of the students willing to spend time with an individual with TS during lunch time. Yet there was no statistically significant increase in willingness to go on a date with an individual with TS.

At T3 none of the items except one (60% willing to sit next to a person with TS) could be maintained significantly above baseline levels. Additionally, the percentages of uncertainty returned to baseline levels.

It must be emphasised that although the Mann-Witney U test indicates that the intervention group still differed significantly from the control group in all the variables, the Friedman test and post-hoc Wilcoxon indicated otherwise. Thus, 9 weeks after the workshop the intervention group's Attitudes and Behavioural Intentions returned to baseline levels. This might suggest a Type I error in the Mann-Whitney U test caused by the small sample size, in particular, there was a 17% drop-out of students in the 9-weeks follow-up.

Table 24 - Behavioural intentions of the intervention group (n=30) per item before the intervention, after the intervention, and at a 9-weeks follow-up.

	Pre-intervention n=30			Post-intervention n=30			9-weeks follow-up n=25		
How likely is it that you would	Not Likely at all/Unlikely	Unsure	Likely/ Very Likely	Not Likely at all/Unlikely	Unsure	Likely/ Very Likely	Not Likely at all/Unlikely	Unsure	Likely / Very Likely
go to the cinema with a person with Tourette's syndrome?	n=13 (43.30%)	n=11 (36.70%)	n=6 (20.00%)	n=6 (20.00%)	n=7 (23.30%)	n=17 (56.70%)	n=5 (20.00%)	n=9 (36.00%)	n=11 (44.00%)
want to sit next to a person with Tourette's syndrome in class?	n=8 (26.70%)	n=8 (26.70%)	n=14 (46.70%)	n=3 (10.00%)	n=5 (16.70%)	n=22 (73.30%)	n=1 (4.00%)	n=9 (36.00%)	n=15 (60.00%)
want a person with Tourette's syndrome as a close friend?	n=3 (10.00%)	n=9 (30.00%)	n=18 (60.00%)	n=0 (0%)	n=4 (13.30%)	n=26 (86.70%)	n=1 (4.00%)	n=8 (32.00%)	n=16 (64.00%)
want to date a person with Tourette's syndrome?	n=9 (30.00%)	n=15 (50.00%)	n=6 (20.00%)	n=8 (26.70%)	n=6 (20.00%)	n=16 (53.30%)	n=10 (40.00%)	n=8 (32.00%)	n=7 (28.00%)
want to go to a shopping centre with a person with Tourette's syndrome?	n=7 (23.30%)	n=10 (33.30%)	n=13 (43.00%)	n=1 (3.30%)	n=5 (16.70%)	n=24 (80.00%)	n=2 (8.00%)	n=7 (28.00%)	n=16 (64.00%)
want to work on an assignment with a person with Tourette's syndrome?	n=8 (26.70%)	n=13 (43.30%)	n=9 (30.00%)	n=1 (3.30%)	n=6 (20.00%)	n=23 (76.70%)	n=2 (8.00%)	n=9 (36.00%)	n=14 (56.00%)

ask a person with Tourette's syndrome to come over to your house to spend time together?	n=8 (26.70%)	n=8 (26.70%)	n=14 (46.70%)	n=0 (0%)	n=4 (13.30%)	n=26 (86.70%)	n=4 (16.00%)	n=6 (24.00%)	n=15 (60.00%)
introduce a person with Tourette's syndrome to your friends?	n=11 (36.70%)	n=3 (10.00%)	n=16 (53.30%)	n=0 (0%)	n=6 (20.00%)	n=24 (80.00%)	n=3 (12.00%)	n=5 (20.00%)	n=17 (68.00%)
introduce a person with Tourette's syndrome to your parents?	n=8 (26.70%)	n=6 (20.00%)	n=16 (53.30%)	n=2 (6.70%)	n=6 (20.00%)	n=22 (73.30%)	n=4 (16.00%)	n=8 (32.00%)	n=13 (52.00%)
spend time with a person with Tourette's syndrome during lunch and break time at school (i.e. sit and talk, etc.)?	n=7 (23.30%)	n=5 (16.70%)	n=18 (60.00%)	n=2 (6.70%)	n=3 (10.00%)	n=25 (83.30%)	n=1 (4.00%)	n=10 (40.00%)	n=14 (56.00%)

Table 25 - Differences in behavioural intentions per item. Comparison to baseline measures using Wilcoxon signed-rank test.

	Post-intervention		9-weeks follow-up	
Behavioural Intentions Questions <u>How likely is it that you would</u>	z-score	p-value	z-score	p-value
go to the cinema with a person with Tourette's syndrome? *	-2.85	0.004 *	-2.21	0.03 *
want to sit next to a person with Tourette's syndrome in class?	-3.19	0.001 *	-3.13	0.002 *
want a person with Tourette's syndrome as a close friend?	-2.89	0.004 *	0.00	1.00
want to date a person with Tourette's syndrome?	-2.09	0.04	-0.43	0.67
want to go to a shopping centre with a person with Tourette's syndrome?	-0.43	0.001 *	-1.64	0.10
want to work on an assignment with a person with Tourette's syndrome?	-3.50	<0.001 *	-1.33	0.18
ask a person with Tourette's syndrome to come over to your house to spend time together?	-3.38	0.001 *	-1.57	0.12
introduce a person with Tourette's syndrome to your friends?	-3.78	<0.001 *	-1.50	0.13
introduce a person with Tourette's syndrome to your parents?	-2.74	0.01 *	-0.16	0.87
spend time with a person with Tourette's syndrome during lunch and break time at school (i.e. sit and talk, etc.)?	-2.86	0.004 *	-0.24	0.81

7.9.5.2. Control group

7.9.5.2.1. Knowledge

The Friedman's test revealed that there was a statistically significant difference for Knowledge about Tourette's syndrome (Table 26, p. 219) for the participants in the control group at the three different time points (T1, T2, T3), $\chi 2$ (2)=8.68, p=0.01. Post hoc analysis

Table 26 - Paired Wilcoxon signed rank test for control group's knowledge at different time points.

Variable	N	z-score	p-value	r
Knowledge Scores pre-test vs	30	-1.23	0.22	-0.15
Knowledge Scores post-test	30			
Knowledge scores post-test vs	30	-1.98	0.05	-0.27
Knowledge scores follow-up	25			
Knowledge scores pre-test vs	25	-2.61	0.01	-0.35
Knowledge scores follow-up	30			

with Wilcoxon signed-rank tests was conducted with a Bonferroni correction applied, resulting in a significance level set at p<0.017. There were no significant differences between T1 and T2 (z=-1.23, p= 0.22, r=-0.15) nor T2 and T3 (z=-1.98, p= 0.05, r=-0.27), but there was an increase in knowledge about Tourette's syndrome between T1 and T3 in the control group (z=-2.61, p= 0.01, r=-0.35). This suggests that the control group was more knowledgeable about TS 9 weeks after the intervention, indicating that the program produced spill-over effects to non-targeted students.

7.9.5.2.2. Attitudes and behavioural intentions

The Friedman's test indicated that in the control group there was non-significant main effect for Attitudes at the three different time points (T1, T2, T3), $\chi2$ (2)=1.61, p=0.45 or behavioural intentions, $\chi2$ (2)=1.56, p=0.46).

7.9.6. Film assessment and intervention evaluation

The short evaluation questionnaire administered to the intervention group indicated that most participants enjoyed the workshop (76.70%), especially the movie about a person with TS (66.70%). The intervention group also reported that their knowledge about TS improved after the workshop (76.50%). Nevertheless, only around half of them reported that their attitudes had changed because of the interventions. This highlights the resilience of negative attitudes (Table 27, p. 220).

Table 27 - Intervention assessment.

Question	Strongly Agree/Agree Number of Participants (%)	Neutral Number of Participants (%)	Disagree/Strongly Disagree Number of Participants (%)
My knowledge about TS has improved as a result of the workshop	23 (76.70%)	3 (10.00%)	4 (13.30%)
My attitude towards individuals with TS has improved as a result of the intervention	17 (56.70%)	9 (30.00%)	4 (13.30%)
I enjoyed participating in the workshop	23 (76.70%)	6 (20.00%)	1 (3.30%)
The film was the most interesting/useful part of the workshop	20 (66.70%)	8 (26.70%)	2 (6.70%)
The presentation with the slides was the most interesting/useful part of the workshop	7 (23.30%)	15 (50.00%)	8 (26.70%)

Furthermore, the questionnaire assessing the feelings that the participants developed towards Brad (the focus individual of the movie) indicated that the majority felt moved (73.30%) by him and could understand how he was feeling (83.30%). Nevertheless, the majority (60%) of the intervention group could not relate to the movie's protagonist indicating a lack of empathy towards someone different (Table 28, p. 221).

Table 28 - Movie assessment questionnaire.

Question	Strongly Agree/Agree Number of Participants (%)	Neutral Number of Participants (%)	Disagree/Strongly Disagree Number of Participants (%)
I developed positive feelings towards the main character of the film	23 (76.70%)	6 (20.00%)	0 (0%)
I could relate to the main character of the film	12 (40.00%)	7 (23.30%)	11 (36.70%)
I felt moved by the main character of the film	22 (73.30%)	4 (13.30%)	4 (13.30%)
I felt empathy towards the main character of the film	23 (76.70%)	4 (13.30%)	2 (6.70%)
I could understand how the main character of the film was feeling	25 (83.30%)	3 (10.00%)	2 (6.70%)

7.10. Qualitative results

Data for both the intervention and control groups were analysed separately for each time point. The themes that emerged at the baseline stage were found to be the same for both groups and were therefore combined to present a more comprehensive picture of the findings. Emergent themes from the control group post-intervention were identical to the pre-intervention themes and are therefore not presented separately. At the 9-weeks follow-up stage of the study, qualitative data collected was insufficient for any themes to emerge.

Two baseline themes emerged from the answers that 59 Year Ten students provided to the open-ended questions. However, just one theme emerged from the post intervention data from 30 participants from the intervention group.

7.10.1. Baseline (T1) themes

Two main themes emerged which highlighted the duality of participants' thoughts and feelings towards people with TS. Whilst on the one hand, individuals acknowledged that people with TS possessed characteristics that rendered them similar to themselves, on the other hand, they perceived people with TS as fundamentally different from the norm and in need of healing. Similarly, although the adolescent participants advocated for equal rights of individuals with TS, they also viewed social participation and inclusion as an exclusive entitlement for individuals that fit in the realm of normalcy.

Prior to the intervention, the participants asserted that people with TS were 'like' everybody else but 'lacking'; i.e. people with TS resembled their tic-free counterparts, but they were simultaneously conceived as fundamentally different. TS was viewed from a medicalised perspective, as a deficiency that needed to be resolved. Although the participants believed in social justice and maintained that individuals with TS should be granted with equal opportunities, they also felt that the inevitable insufficiency that the tics caused would impair them in both a social and educational context. In other words, they considered that people with TS could only be accepted into society and integrated into mainstream education if they were aligned with the norm, and their undesirable difference alleviated.

People with TS were also viewed as an excludable population that could be relegated to a more restrictive and segregated setting. A dual system of education was firmly supported, whereby students that interrupted “normal” pupils’ education should be placed in a segregated setting. As demonstrated below, the participants also indicated that whilst they were, in practice unwilling to accept diversity, they believed that individuals with TS would be socially valued through a process of normalisation. In this way, participants ratified the medical model by which “deviant others” need to be rehabilitated in order to return to normalisation and claim their place within mainstream education and society (Oliver, 1996).

7.10.1.1. Theme I: “I can tell you 10000 ways by which we are the same but also different”

A duality of thought was apparent in the participants’ perception about individuals with TS; the majority viewing people with TS as similar to themselves, but simultaneously as fundamentally different from what they (the participants) conceptualised as normal. The following two subthemes explain this duality:

Sameness and humanness

At baseline, the majority of participants asserted that individuals with TS “are still human beings”; this phrase being repeated by 13 of the pupils participating, to explain the reasons why they thought that people with TS should not be treated differently from their peers. Similarly to themselves, individuals with Tourette's were seen as fully human as explained below by one participant:

> *TP27: …they are humans along with everyone else and they probably don't want* to be singled out.

In other words, TS was not conceptualised as something that overshadowed individuals' fundamental human characteristics and swallowed their character's traits, and they should not therefore be excluded from social justice and participation.

It appeared from the data that humanness was bestowed to individuals with TS by the participants partly because they were not perceived as culpable for their condition; although tics were seen as undesirable, they were not considered to be under the individual's control. Rather, tics were viewed as being caused by external and uncontrollable by the individual.

> TP57: I think it could be annoying however, I would remember that it's not their fault and it's uncontrollable.

In alignment with the first part of the study, pupils attributed the causes of TS to factors beyond the individuals control and formed their attitudes and perceptions accordingly.

Differentness – *"They're the copies of us, but different"*

Individuals with TS were simultaneously conceptualised as different and were evaluated and measured based on what was viewed as the norm, namely themselves. By regarding TS as a deficit and viewing the condition through a medicalised approach as a misfortune, many participants presumed that they would have to assume the role of the benevolent friend and provide social support for the individual with TS.

Indeed, even when people with TS were considered "no less humans" than everybody else, a direct or indirect comparison was conducted with what they perceived as normal. Participants exempted individuals with TS from the spectrum of normalcy, although they clearly acknowledged their humanness. The tics were viewed as aberrant behaviour. This highlights that conformity and uniformity were highly valued:

> TP47: They are just different.
>
> TP3: They're almost regular people
>
> TP8: Because they are still human beings and should be treated similarly to normal people.

Whilst none of the pupils actually defined what they meant by ‘normal’, the distinguishing characteristics that their peers with TS possessed makes them similar to normal, but not normal. The focus was placed on their differentness and distinct characteristics. Their uniqueness, which rendered them hypervisible, was unvalued and mostly viewed through a medicalised approach.

Participants perceived themselves to fall within the boundaries of normality while individuals with TS were more or less marginalised depending on the disruptiveness of the tics. Indeed, pupils in the study believed that individuals with TS’s rights to social participation and social justice were inevitably tied to their proximity to normalcy. Participants’ intention to include individuals with TS in their lives and their reciprocal activities was mitigated by the severity of the tics:

> TP36: Depends on how bad the tics are.

The boundaries between the normal and those who were considered divergent from this normalcy were sharp and unambiguous, and the only way individuals with TS could penetrate the former category was through rehabilitation. Indeed, the tics were viewed as a personal problem that needed to be treated by experts and healed in order to eliminate and ameliorate the pernicious effects of the tics. The need for rehabilitation was pointed out by reducing individuals with TS to patients that were perceived as defective by medical regimes. Medical cure was viewed as the quintessential solution to the problem in order to restore the natural order of things:

> TP4: Needs tablets to get better.
>
> TP9: Needs their medication.

By viewing TS through the medical paradigm, the onus was put on the inability of a person with TS to perform as the average person and a normative approach was adapted whereby people with TS were viewed as inadequate:

> *TP17: Can they do everything that we can do? I don't think so.*
>
> *TP44: There are some things that person with Tourette's sy*ndrome can't do. e.g. things with concertation.
>
> *TP36: They can't always do what they want.*

The conceptualisation of TS as a form of impairment resulted in the individuals with the condition being viewed as vulnerable and, subsequently, dependent on their counterparts (Murray, 2004). In most cases, a form of friendship was delineated in which the typically developing adolescent would assume a 'mothering role' and 'tolerate' and 'endure' the individual with TS. This meant that friendship as described by the pupils was not defined by reciprocity but by a dependency model whereby the pupils would provide social and emotional support to them whilst at the same time pitying them:

> TP12: I would work with them, so they feel wanted.
>
> TP9: I will help them to have the experience of friendship.
>
> TP46: I would tolerate their Tourette's.
>
> TP3: It would take a decent person to be there for them, and to put up with them.

> *TP22: If I was in their place, I don't know how I would live.*

Providing assistance to an individual with TS was a unilateral decision and participants did not take into consideration the perspective of the individual whom they were intending to help. By assuming to know the social needs of people with TS, the pupils indicated that they did not denote that the intended recipient of the support might want to initiate it themselves or might not want it and could choose to refuse it. Such notions that it is acceptable to provide unsolicited help may constitute a potential threat to the self-esteem and self-worth of people with TS (Roloff, 2000). Although this support might be well intended, it may be perceived as patronising by the recipient. In alignment with the medical model, the majority of the participants also insinuated that people with TS would be experiencing unfulfilling lives. In turn, participants appeared to feel compelled to place the onus on how they could potentially help them improve them:

> TP40: I feel sorry for them. They can't have a normal life like us.
>
> *TP16: They are people that can't have a normal life, I'm fortunate enough to* have a normal life, so I can help them.
>
> TP38: Because she has the tics, no one can help her. Just the doctors.

In line with the discourse of pity some of the participants envisioned individuals with TS to possess unrealistically positive qualities and attributes in comparison to their normal peers. They asserted that they would be more interesting, more intelligent or even more generous than typically developing adolescents. This might have been viewed as a form of compensation to make up for having TS. Alternatively, in align with the "sympathy effect", which is a form of positive bias, participants might have been aware that individuals with TS would be stereotyped and wanted to respond in kindness towards their disadvantaged individuality:

> TP11: They might be smarter.
>
> *TP44: They probably have the best personalities. Better than everybody else's.*
>
> TP31: I'm sure that they have better ideas than everybody else.
>
> TP13: Probably the tics make that person better than normal people.

Goldstein and Johnson (1997) have pointed out that by crediting exaggeratedly positive attributes on individuals with differentiating characteristics, the gap between "them" and "us" is widened. Thus, stigmatisation and prejudice is not only formed when one is attributed with a negative label. By assigning an individual an overly positive identity, a person is still stereotyped and set apart from his/her peers. The individual is still stripped from his personality and lumped into a category based on his "otherness".

7.10.1.2. Theme II: "Rights" and "Access"

Rights

The majority of the participants asserted that individuals with TS should have the same rights as their typically developing peers, believing that all discriminatory and prejudicial attitudes should be ameliorated and their peers with TS should be ensured equal treatment:

> TP60: Even if they have Tourette's all should be treated as if they are the same. Everyone has to have equal treatment.

Participants based their line of reasoning on the fundamental premise that all pupils are equal and consequentially asserted sameness in treatment irrespectively of the personal characteristics and additional needs of individuals with TS. Thus, it was emphasised that people with TS should not be judged based on their individual circumstances:

> TP17: Well, if they swear in class they should be punished like everyone else.

Thus, social justice was solely conceptualised as an act of eliminating discriminatory barriers for individuals with differentiating characteristics. Human rights however were conceptualised as an abstract concept and participants did not discuss any practical strategies to bring about change. The participants placed emphasis on the concept of equality and not equity. Thus, it was not acknowledged by pupils that redistribution of resources may be warranted in order for equal educational opportunities for TS to be provided.

Access to mainstream education and society

Although the participants strongly advocated for the rights of every individual to receive education, they believed that due to their deficit individuals with TS should be treated in special schools that 'protects' pupils. Alternatively, they could be home schooled:

> TP20: They still deserve education. Just not with other normal people.
>
> TP47: They might benefit from home school.
>
> *TP8: People with Tourette's could do well in school but only if they are in a* specialised school for their problem.

The above quotes exemplify that the participants did not regard the school as having any responsibility to respond to the pupils' characteristics in an equitable manner in order for them to accomplish their full potential. A form of ableism is noted here in relation to mainstream schools. In the participants' views people that fell outside the range of what was considered normative should be excluded from mainstream schools. Participants could not envision a classroom where sameness and homogeneity did not prevail. The hyper-focus on the needs of the 'normal pupils' and the concept of ableism precluded access for individuals with TS to the general mainstream school:

> TP27: It would constantly distract everyone, and the teacher.

Participants did not view mainstream schools as a place that should be equipped in order to allow all pupils to participate. A separatist view of education was therefore put forward. People with TS were expected to stop ticking and adjust to the normal behaviour otherwise restricted to a school environment with similar others:

> TP23: They should stop or should go to a school with children with similar issues.

A resistance to inclusive education was revealed in the participants' opinions about the right of their peers with TS to be included in mainstream classrooms. Despite their apparent general egalitarian views, humanistic perceptions about individuals with TS and ideals about social justice, they considered that the presence of individuals with TS within their classroom would decrease their academic achievements. Students with TS were viewed as an ongoing disruptive force and participants were concerned that the presence of an individual with TS in the classroom would make it harder for them to concentrate. They felt that inclusion would impede their learning and have a negative personal effect on them:

> *TP6: If it's affecting others' learning, it's not fair.*

The participants worried that the inclusion of an individual with TS in mainstream school might also result in typically developing adolescents being deprived of their rights. Within the classroom, the rights of "normal" pupils (entitled to be taught, and to concentrate with minimal distractions) should be given precedence over the right of an individual with TS to participate in mainstream education. The arguments reflect those segregationists; that mainstream students' academic achievements might be adversely affected by the presence of individuals with differentiating characteristics.

> *TP8: They would distract me from my work and I wouldn't do well anymore.*

This argument which has been predominantly disputed by the empirical evidence (Kalambouka, Farrell, Dyson, & Kaplan, 2007) was described as the main barrier for inclusion. It is noteworthy that the participants did not consider the positive emotional and social benefits of inclusive settings for mainstream students.

Some viewed segregation as a form of protection from an unwelcoming mainstream educational system. The participants strongly believed that the ethos of mainstream school would fail individuals with TS and under the auspices of their own protection they endorsed their marginalisation. Although this form of ostracism was based on a benevolent logic it still promoted segregation:

> TP17: They would be bullied. So, they need to go to another school.
>
> TP22: They will be laughed at too much.

Participants viewed the right of students with TS to social participation and inclusion as being dependant on their ability to assimilate to the norm. So as long as their deviating characteristics were hidden away, and as long as a ‘passing or covering behaviour’ was adopted by individuals with TS, they could be socially integrated. Thus, it was expected that a person with TS should ‘pass’ as a tic-free, normal individual and to deliberately conceal their condition and supress their tics in order to be accepted and become part of the ‘normal’ group’:

> TP45: It depends how the person can control the tics
>
> TP19: They could do their best to control the tics.
>
> TP27: They should try and be normal.

7.10.2. Post-intervention (T2) theme for the intervention group

One main theme emerged as a result of the intervention. The experimental group continued to view individuals with TS as different thus, the existence of a differentiating characteristic was not negated. However, they were not necessarily seen as “others”. Being different was more respected and assimilation did not appear to be a social necessity. On the contrary, participants appeared to be keener to challenge the devaluation of individuals with TS and the quest for the assimilative cure was less prominent. Diversity started to be conceptualised as something ordinary and the concept of normalisation was deconstructed. The participants did not require pupils with TS to adjust to the predefined requirements of the school in order to fit into their school culture and ethos. After the intervention, pupils with TS were viewed as part of the school community and not just ‘a problem’ that their schools had to manage or eliminate. Although after the intervention segregation was not strongly rejected, it was more vividly questioned. The virtues of inclusion were better conceptualised and participants acknowledged the responsibility the school and society held in order to make inclusion possible.

7.10.2.1. Theme I: Moving towards inclusiveness and embracing diversity

Before the intervention, the majority of the participants did not appear to discern the beneficial effects of heterogeneity in the classroom. Immediately after the intervention however, traditional disability and ability views were challenged and perceived more critically by the intervention group in that the students appeared to be more resistant towards the narrow confines of normalcy and more appreciative towards diversity by embracing their own and their peers differentiating characteristics and uniqueness’s. They were more inclined to view normalcy as a social construct that should be questioned:

> TP54: They can have a normal life, but honestly, is there such a thing as normal life?

It was also apparent that they were more amenable to broadening the concept of normalcy and to include their peers with TS within the category. The concept of differentness and normalcy were therefore destabilised. Participants were keener to embrace diversity by recognising and accepting both the similarities and differences to individuals with TS:

> TP49: They are normal people.
>
> TP13: It's just another person, isn't it?
>
> TP37: They are us.

Although the participants acknowledged that TS was part of their peers’ social identity, the diagnosis was considered as something that did not overshadow the individuality of the person. They accepted the differentiating characteristics of the individual with TS and viewed them secondary to the persons’ humanness. Furthermore, they acknowledged that everyone has multiple and complex identities and cannot solely be characterised by any aspect of it:

> TP14: You wouldn't treat someone with a broken arm differently. So why would you treat someone with Tourette's?

Furthermore, participants were more reflective in their answers by acknowledging that each individual possessed unique characteristics that others may view as either desirable or undesirable:

> TP14: No one is perfect and everyone has their flaws.
>
> TP24: Everyone is special and different in their own way.

Characteristically, one participant acknowledged that similar to TS, her ethnicity might be perceived by some to be an undesirable characteristic:

> TP13: I am Portuguese, so other people may not like me because of that.

In the majority of cases post-intervention, participants in the experimental group reported that their decision to socialise with an individual with TS would depend upon the attributes of the individual with TS as well as their common interests. Thus, the criteria for selecting a typically developing friend would also be applied to individuals with TS. The tics were no longer considered the most salient aspect of the individual and people with TS were assessed based on their character:

> TP54: As long as they treat me right I will do the same.
>
> TP23: As long as they can tolerate me I can tolerate them.

Immediately after the intervention, the experimental group acknowledged that inclusion does not solely depend on the individual but also on the resources that are offered in order to accommodate his/her needs. Barriers were regarded as mostly imposed by society and predicated on issues of resources and prejudice:

> TP36: Being included depends on what the school can offer.
>
> *TP54: If they don't get teased, they will be fine.*

It was acknowledged that inclusion could be impeded due to poor attitudes and TS was not seen as a formidable tragedy. On the contrary, prejudicial attributes were:

> TP14: I don't feel scared of them. I feel on edge of how other people treat them.

7.11. Discussion

The current study evaluated the short and long-term impact of an easily delivered intervention on tic-free adolescents' knowledge, attitudes and behavioural intentions towards individuals with Tourette's syndrome. It also assessed whether gender and personal acquaintance with an individual with a stigmatised condition (LD/MI or DD) impacted upon stigma endorsement and explored differences in the effects of the intervention based on the aforementioned variables. The study design, a concurrent mixed-method design with a 9-weeks follow-up was chosen in order to overcome some of the limitations of previous interventions, namely the exclusive use of quantitative data and short-term results. Indeed, this is the first study to my knowledge that aimed to assess the long-term impact of implementing an anti-stigma Tourette's syndrome workshop in a secondary school in the UK. This study is also novel in eliciting a nuanced level of understanding about attitudes and behavioural intentions of adolescents through combining quantitative and qualitative data during the interpretation stage of the analysis. Finally, by using a waiting-list control group design all participants received the intervention, overcoming the disadvantage of a non-treatment control group. In this way, I therefore managed to balance the need for a control group with the school requirement that stipulates all participants should benefit from the intervention.

Due to an average of neutral responses (midpoint answers in the five point Likert scale) at the baseline stage, one could argue that the intervention was not warranted and would only be needed if the sample responded with mostly negative attitudes. In other words that midpoint answers which could also indicate indifference or unwillingness to answer, are better than negative responses. However, it is important to consider that the averages indicating neutral scores were based on data with considerable variance and students' negative attitudes, which could impede school belonging for individuals with TS, still need to be addressed. Furthermore, numerous studies (Schulze, Richter-Werling, Matschinger, & Angermeyer, 2003) have indicated that midpoint options could be concealing the open expression of negative attitudes and prejudice for students who wish to present themselves as socially or politically ‘‘correct’’. Indeed, the questionnaires that were used in this study tackle blatant prejudicial attitudes and participants’ answers may have been restrained due to the overt nature of the questions.

Overall, the study indicated that this short and easily delivered intervention can have a positive impact on typically developing adolescents’ Knowledge, Attitudes and Behavioural Intentions towards individuals with TS. Significant changes were particularly true for students with previous personal contact with individuals with stigmatising conditions. These results substantiate the value of contact theory (Allport, 1979) and education in eliminating stigma and discrimination, and expand its applicability in the area of TS.

Immediately after the intervention the control groups’ adamant opinions about the need for individuals with Tourette’s to be `healed’ and normalised in order to fit into mainstream society’s stringent criteria were replaced by statements that questioned the concept of normalcy and contested the usefulness of homogeneity. With regard to the follow-up results, the picture was more diverse. Participants’ Knowledge of TS had decreased by the follow-up data collection point but was still significantly above baseline measures. The improvement recorded on the Attitude and Behavioural Intention items upon completion of the intervention was not sustained at the follow-up, where students almost returned to their baseline answers.

7.11.1. Short-term effects of the intervention

First, as predicted, the workshop was found to have a positive impact as indicated by an improvement in Knowledge, Attitudes and Behavioural Intentions towards peers with Tourette's syndrome immediately after the intervention had been implemented. Following the workshop, the intervention group demonstrated significantly greater knowledge about TS, and less prejudicial attitudes but also more readiness to enter different types of social relationships with someone who had TS in comparison to the control group. The short-term results of the study were in align with those that have found some efficacy for short, school-based anti-stigma interventions using a combination of education and indirect contact (Economou et al., 2012; Stuart, 2006).

The qualitative data elicited from the participants was used to explore and provide a depth of understanding of the social phenomenon under study and helped to interpret the changes observed in the quantitative data after the intervention. Thus, the open-ended data enabled this elaboration process. The participants' answers indicated that attitude changes were facilitated by disputing the value of homogeneity and questioning the need to assimilate to the norm. Indeed, immediately after the intervention the experimental group was more inclined to deconstruct the notion of normalcy and was insightful enough to see the uniqueness of all human beings and even themselves. TS was viewed as another marker of human diversity. People with TS were viewed beyond their label, as multidimensional, whole individuals and their deficit as only one element of their social identity. Some adolescents further questioned whether the barriers imposed on an individual with TS were due to their internal deficit or whether hurdles were imposed by society and the school.

It should be highlighted that although the intervention was efficient in changing adolescents' overall attitudes and behavioural intentions towards individuals with TS immediately after the intervention, participants were still hesitant to accept that mainstream schools could accommodate for the needs of students with TS and were reluctant to engage in personal and intimate relationships with their peers with TS after the intervention. This might indicate that attitudes that endorse unconditional social participation of individuals with TS are more resilient to change.

7.11.2. Long-term effects of the intervention

The original study hypothesis asserted that individuals in the intervention group would continue to experience a positive change as regards all variables from post-test to follow up. The follow-up findings were, however, diverse and non-conclusive. On the one hand, the retention of knowledge about TS after 9 weeks of the intervention was maintained significantly over the baseline levels, with participants continuing to hold more accurate conceptions of several aspects of TS. These findings made the study the first to provide RCT evidence for the long-term effects of an easily delivered intervention on the awareness of TS. On the other hand, attitudes and behavioural intentions could not be sustained over time. The 9-weeks follow-up indicated that the effects of the intervention lessened with time, almost returning to baseline levels. Diminishing effects during follow-up periods is a common finding in intervention studies (Chan, Mak, & Law, 2009; Economou et al., 2012). It appears that attitudes and behavioural intentions are more resilient to change than knowledge and possibly necessitate a different approach for targeting it. As it has been stated before, the film was chosen as a part of the intervention in order to enhance persuasion through creating an experiential learning situation and engendering an empathic emotional response to individuals with TS. Vivid emotional responses from the movie were expected not only to facilitate attitudinal change, but also to leave a rich and strong memory trace. Although the image of an individual with TS could have been imprinted in the participants' minds, this may not have resulted in such a strong emotion that would endure throughout the course of the intervention and be evident at the 9-weeks follow-up and beyond. Possibly a booster-session in the following weeks to its implementation could have prevented the positive effects of the intervention from eroding. The short duration of the workshop, which is acknowledged as one of its limitations, could account for the unsustainability of the results – although the optimal duration of an intervention is not yet clear. Furthermore, prejudicial attitudes about Tourette's syndrome that are well established within the wider society may have impacted the positive attitude change during the 9 weeks period.

7.11.3. External circumstances that may have influenced the effects of the intervention

It should also be emphasised that before the follow-up data were collected, the participants' attitudes may have been influenced by incidents of racism against ethnic minority school students. These occurrences, which were reported to have been fuelled as a result of the European Union referendum, could have violated the participants' sense of safety and belonging and made them less receptive to acceptance of people who were different from themselves. Taking into consideration that the follow-up data was collected only four days after these incidents, which took place within the school premises and were reported in the local newspapers, it is likely that the interplay of these factors might have affected the results. Moreover, during the follow-up stage, the study was underpowered due to high attrition rates. Therefore, a lack of statistical significance may be a fabrication caused by a small sample size.

Further research is needed to investigate strategies to sustain these short-term improvements in students' attitudes towards their peers with Tourette's syndrome. However, the program could be considered as an initial step for future development and a building block in a more comprehensive process of social change.

7.11.4. Gender and personal acquaintance with an individual with a stigmatised condition as variables predicting attitudes and behavioural intentions

Contrary to predictions, the findings of this study suggest that gender does not play a prominent role in stigma endorsement; the study failed to detect a substantial difference between male and female attitudes and behavioural intentions towards individuals with TS. This is in contrast to the majority of previous studies addressing the aforementioned question that have reported significant differences in gender and have highlighted the potential implication that could have on the planning of anti-stigma programs. Prior studies suggest that school-aged females tend to express more accepting and tolerant attitudes towards individuals with stigmatising conditions and males are more likely to show greater prejudicial behavioural intentions (Krajewski & Flaherty, 2000; Reis, 1988; Townsend, Wilton, & Vakilirad, 1993). Furthermore, females report greater changes in stigmatising beliefs following the intervention. The current study findings indicated that, in relation to TS, stereotypes and desire for social distance are equally prevalent among females and males. The effects of the intervention were not mediated by gender either, these findings being in alignment with Friedrich et al., (1996) and Boudjouk et al., (2000), which report that in childhood and adolescence gender may not be a prominent variable in shaping attitudes and desired social distance towards individuals with TS. This could indicate that TS is viewed differently from other stigmatising conditions and that females feel less impelled to be protective towards this population. Further studies should explore the relationship between gender and stigma endorsement in relation to TS in more depth.

This study provides further support to the link between personal experience with stigmatising conditions and favourable attitudes. Tic-free adolescents’ acquaintances with someone with an intellectual disability, a mental illness or a developmental disorder, was found to be a positive and important influence on their attitudes towards their peers with TS. Moreover, participants who had prior contact with stigmatised groups were more sensitive to the provision of information about TS and significantly more positive outcomes were observed in the realm of attitude and behavioural intentions after the intervention. It should be emphasised that although at the baseline stage adolescents with experiential knowledge of marginalised groups were not necessarily more knowledgable about TS, they were more inclined to interact with individuals with TS. This might indicate that contact rather than knowledge is a more important component in attitudes and therefore should be a cornerstone in anti-stigma interventions. Furthermore, less prejudicial attitudes by individuals that have previous experience with individuals with stigmatised conditions might suggest the existence of 'secondary transfer effects' (STE) (Pettigrew, 2009). According to STE theory, the effects of intergroup contact does not only influence prejudice towards that specific population but also towards other (secondary) outgroups that were not initially involved in the intergroup encounter. Thus, contact with other stigmatised outgroup members (individuals with ID or mental illness) might have reduced prejudice toward individuals with TS and a generalisation of contact effects might be indicated. Future research should assess if the current intervention has the potential to reduce prejudice in general. In particular, the extent to which indirect contact with an individual with TS is associated with more positive attitudes towards individuals with other stigmatised conditions overall. Positive results could indicate the relevance and importance of implementing such an intervention in secondary schools regardless of whether stigmatisation of individuals with TS is a particular concern of the school/class involved.

7.11.5. Evaluation of the workshop by the participants

Pupils' evaluation of the workshop revealed that the movie was considered to be the most interesting and engaging part of the workshop. Indirect contact, which was facilitated through the film was therefore assessed as more constructive than the educational element of the workshop. However, it should be emphasised that the fact that Brad's life was cinematised may have been interpreted by some pupils as an indication that he was unrepresentative of the wider TS community and an atypical person with TS. Contact with individuals that are perceived as atypical in a group may promote subtyping and help further confirm the general stereotyping about a specific population. Despite these limitations in the contact condition, the film had a positive effect on more general attitudes towards individuals with TS. Participants' views on the acceptability of the intervention's content was assessed immediately after it had been implemented. The data suggested that more than 76% percent of the experimental group enjoyed taking part in the workshop, which indicated the acceptability of the workshop to the participants.

7.12. Limitations of this study

Although the results of this study are encouraging, several methodological limitations should be considered. The stimulus video that was shown to the participants accentuated the differentiating characteristics of the targeted individual and understated the positive characteristics. Moreover, female adolescents with TS are rated less socially acceptable and are more highly scrutinised for their deviant behaviour than adolescent males with the condition (Boudjouk et al., 2000). This may have urged participants to endorse more negative attitudes and express low levels of intended engagement. Furthermore, variables such as perceived attractiveness of the targeted individual may have moderated varying degrees of prejudice. However, the variation within the data and the absence of floor or ceiling effects suggests that individual differences still existed within the pupils' responses.

In relation to the students' participation, although all pupils from the two classes decided to take part, it is acknowledged that it would have been difficult for the pupils to resist given the head teacher's support of the study. Therefore, all the participants might not have been fully engaged in the study.

As is the case in most studies that use mixed-model design, more missing data was observed in open-ended questions than in closed and a more diversified set of answers were provided. A larger number of respondents avoided providing an answer to justify their choice and communicated their opinion simply by choosing between an explicit set of alternatives. More specifically, only 37% of the questions were answered by the participants at the pre-intervention stage (435 answers were provided out of 1180). In most cases, the responses consisted of one or two clause sentences. Immediately after the intervention, the experimental group answered 26% of the questions (154 answers out of the 600) and the control group 21% (124 answers out of 580). Response rates dropped even lower at the follow-up stage of the study, in which only 14.5% of the questions were answered by the intervention group (87 out of 600) and 8% by the control group (46 out of 580). The phenomenon of qualitative missing data in mixed-design surveys can be due to the demand of more cognitive engagement in comparison to closed-ended questions. Furthermore, in contradiction to interviews where the investigator prompts the participants to answer a specific question, in self-administered questionnaire it is harder to provide a motive for the participants (Reja, Manfreda, Hlebec, & Vehovar, 2003; Schuman & Scott, 1987).

7.12.1. Effects of attrition

A limitation of this study was the attrition that occurred at the follow-up data collection point. The rate of attrition from pre/post to follow-up was 17%. This resulted in a total sample of 49. This means that the study underpowered at the follow-up stage of the research; low statistical power increases the likelihood that a statistically significant finding is actually falsely positive as was observed (for a given p-value). Due to the limited number of participants and the fact that it was underpowered at the follow-up stage, the study can be characterised as an exploratory rather than a large-scale trial.

7.12.2. Spill-over effects

Although contamination bias was specifically considered when designing the study and the participants of the intervention group were specifically asked not to discuss the content of the interventions with their peers, spill over effects were observed during the follow up stage as far as knowledge is concerned. Due to fact that only one year group from one school was used, there was inevitably an opportunity for diffusion of the information among the participants of the study. Thus, pupils from the same year group would be communicating with each other during the school day, breaks and other social opportunities. This may have exposed the non-targeted control group to some information about the condition and may explain their improved literacy on TS at the follow-up stage of the data collection. Although spill-over effects may jeopardise the validity of the findings, they can also be considered promising as they provide some suggestive evidence that this kind of workshop can facilitate a process involved in untargeted knowledge awareness. Alternatively, the intervention might have brought about an increased interest in TS and participants from the control group might have been liable to the Hawthorne effect. However, a presence of a Hawthorne effect at the post-intervention stage makes it possible to argue more confidently that the implementation of such a program could have positive effects for the wider school. Future research needs to develop multilevel experimental designs that enable the researcher to measure the spill-over effects and gauge the exact effect of the workshop.

7.12.3. Generalisability of the results

It was not feasible to include another school in the study and cluster randomisation was applied at a class level within the same school. Conclusions would have been strengthened if the design had included several schools. More than 20 secondary schools were contacted in order to participate in the research. Whilst reasons varied for their refusing to participate, the majority of them cited issues relating to their capacity to support the research. This highlights the challenges of collaborating with schools as regards TS interventions (or for that matter, any interventions). It also has implications for the generalisability of the results. Future studies should examine if positive results could be extrapolated to a representative sample of adolescents throughout the UK. Thus, they should aim to widen their recruitment scope and engage a wider range of schools in order to assess if it’s a suitable intervention for wider dissemination.

7.12.4. Limitations of the questionnaires used

The Attitude and Behavioural Intention questionnaires used by the study were unstandardized and had not been psychometrically tested. However, all questionnaires were piloted before their initial use to determine clarity, initial reliability, and the need for further revision. The questionnaires showed good validity and reliability scores, however, psychometric assessment of the instrument needs to be conducted for future research. It should be noted nevertheless that, due to high percentage of non-response rates on the open-ended questions, the feasibility of the qualitative part of the questionnaires should to be questioned. Furthermore, most open-ended answers provided were inadequate and, in most cases, insufficient to provide a comprehensive explanation for the participants' Attitudes and Behavioural Intentions. Low response rates may suggest that the qualitative data obtained might not have represented the views of the entire group and it could indicate the occurrence of a non-response error. Thus, the opinion of the respondents of the open-ended questions might differ from the views of those who were non-respondents. It is difficult to determine if non-response error occurred and if there were meaningful differences between those who commented and those who did not in this study. However, it is pertinent to point out that recent studies suggest that individuals with limited English language skills are less likely to voice their opinion in open-ended questions (Reja et al., 2003). This might suggest that the voices of the less fluent population were underrepresented in the open-ended questionnaire and the opinions of students that were not articulate enough to put forward an answer were not heard. On the contrary, those with more profound opinions may provide more verbose responses.

The high rates of non-response indicate that individual interviews with the participants would have been more appropriate for the purpose of the study. Interviewers could have motivated and prompted the participants to explain their line of reasoning. Future studies need to use interviews and/or focus groups to elicit how TS is perceived by adolescents before and after the intervention.

Furthermore, the questionnaires used measured overt and blatant discriminatory attitudes, which did not tap into subtle and covert forms of discrimination that adolescents may harbour against people with TS. Future studies need to investigate implicit attitudes that adolescents may have towards individuals with TS that are less likely to be expressed due to social desirability bias, societal sanctions and the desire to maintain a positive self-perception.

A further limitation relates to the self-report questionnaire assessing previous contact with individuals with stigmatising conditions in that it was based on the assumption that participants share a definition of the diagnostic categories of intellectual disability and mental illness and were able to clearly differentiate them from other categories, such as specific learning difficulties (e.g., dyslexia). However, in contrast to previous studies, it did not examine previous contact to individuals with stigmatising conditions as a dichotomous variable (present or absent only). By asking the participants to indicate the types of contact they have had, the possibility of them subjectively interpreting the concept of "contact" is lowered. Future studies should also examine if factors such as the quality of contact, the frequency and the number of people with stigmatising conditions the participants know might influence their attitudes. Future studies should also consider the impact of other demographic variables (e.g., educational attainment, parents' educational attainment and socio-economic status ethnicity previous experience with inclusive education, religion) and their mediating effects on stigma. Thus, although these variables are not easy to manipulate, in contrast to knowledge, future research should assess if interventions need to be built on the interaction of several types of variables.

Another limitation of the study is that since behaviours were not directly observed, it cannot be denoted whether or not adolescents may have provided socially desirable responses or learned responses which do not reflect actual peer acceptance of pupils with TS. Therefore, there is considerable scope for conducting naturalistic research through observation within school settings in order to unveil attitudinal changes and factors that might impact inclusion and acceptance.

Finally, the efficacy of the workshop cannot be extrapolated to all age groups in secondary schools since this study delved into a specific age range, 14 to 16-year-old pupils who, arguably, had developed abstract reasoning but had not yet consolidated their attitudes

(Jean, 1972). Previous studies indicate that age and maturity levels are determinants of attitudes and desired social distance with younger pupils being more prone to prejudiced beliefs (Skre et al., 2013). Although it was the initial aim of the study to assess the effectiveness of the workshop across secondary age group, this was not possible due to the school concern about statutory examinations from year eleven onwards.

7.13. Summary

The aim of this chapter was to evaluate the effectiveness of the intervention in raising awareness about TS and improving attitudes and behavioural intentions towards adolescents with TS. A total of 59 students from two Year Ten classes in a secondary school in South East England were randomly allocated to an intervention and control waiting-list condition. Data were collected at three time points: two weeks before the intervention, upon the completion of the intervention and at a 9-weeks follow-up.

The intervention yielded substantial changes in students’ knowledge, attitudes and behavioural Intentions. The intervention yielded substantial changes in students. However, only changes in knowledge were maintained at the 9-weeks follow-up. Future research is warranted to find ways maintain the positive results of the intervention.

8

Summary and Discussion

In this chapter, the main aims and findings of the thesis will be discussed. Considerations of the limitations of the study are outlined and recommendations for future research, practice and policy are presented.

The purpose of this research was threefold. Firstly, it aimed to explore typically developing adolescents' perception of their peers with Tourette's syndrome (TS); to understand possible misconceptions they might have in relation to the condition and to construct a model that explains their attitudes towards this population. Secondly, the study sought to develop an intervention based on the aforementioned data; the purpose of the intervention – which was developed out of the misconceptions adolescents had of TS, and the negative attitudes and behavioural intentions they might have fostered towards individuals with TS – being to improve typically developing adolescents' knowledge, attitudes and behavioural intentions towards their peers with TS. To this end, a substantive theory was constructed based on adolescents' perceptions that guided the development of a culturally and developmentally appropriate Tourette's anti-stigma intervention. To my knowledge, this is the first study to have utilised user-defined criteria to develop an intervention – rather than an intervention based solely on theoretical principles – to ameliorate TS stigma. The final aim of the study was to examine the impact of the intervention on adolescents' knowledge and improving their attitudes and behavioural intentions towards a peer with TS. The feasibility and efficiency of the intervention was evaluated using a mixed-method approach as well as a post-intervention follow-up after 9 weeks. Mixed-methods were chosen since the purpose was not only to assess whether or not the intervention had a positive effect on the participants but also how and why the intervention may have worked. Few follow-up measures exist beyond the immediate post-intervention period in relation to TS anti-stigma interventions. This is the first study therefore that assessed whether the improvements in knowledge, attitude and behavioural intentions are long-lasting, or dissipate shortly after the intervention. In order to address these aims two empirical studies were conducted.

In Study One free text writing and focus groups were used to elicit the views of twenty-two Year Ten students from a secondary school in South East England in regards to their perceptions about individuals with Tourette’s syndrome. Grounded theory was utilised to develop an analytical framework that provided directions for designing the intervention and the research measures for the second study. Study Two assessed the intervention using a cluster randomised control trial and mixed-model design.

8.1. Study One: Development of a meaningful intervention

A systematic review undertaken on the topic of stigma in youths with TS (Chapter 2), highlighted that there is insufficient research pertaining to stereotypes, prejudice, and discrimination in relation to TS. The few studies found for the systematic review indicated that individuals without TS exhibit unfavourable attitudes towards individuals with TS in comparison to typically developing peers. Young people with TS themselves described some form of devaluation from others as a response to their condition. However, the systematic review did not account for why individuals with TS have tended to be stigmatised by their peers as well as the general public. The studies that met the inclusion criteria of the systematic review used mainly quantitative methods and were unable to provide information as to the origins of stigma. Furthermore, they did not explore the reasoning behind the “stigmatisers” behaviour, and did not evaluate which motives are urging tic-free individuals to exclude people with TS from social interaction. In other words, they did not identify modifiable causal factors associated with negative attitudes. Furthermore, misconceptions held by typically developing adolescents about the condition have not been identified by the aforementioned studies. Arguably, misconceptions and commonly held beliefs about the condition of TS should be at the heart of any effective anti-stigma intervention. To overcome the shortcomings of previous research, it was decided that a qualitative study was warranted to help provide a more iterative, richer understanding of how typically developing peers perceive TS.

An exploratory study was conducted in a co-educational, multi-cultural comprehensive secondary school in South East England. The opinions of twenty-two students from Year Ten were elicited after the screening of a 4-minute video of an adolescent girl displaying tics. Through free-text writing and focus groups, the participants expressed their opinions in relation to individuals with TS. Then, an explanatory theoretical model was developed using grounded theory. Findings revealed that misconceptions about the condition appeared to be caused by how TS has been depicted in mainstream popular culture. Through misrepresentations and exaggerations in contemporary popular media, people with Tourette’s were conceptualised as straying from the boundaries of normalcy. Nevertheless, the participants still felt the needed to ask questions about the condition, which suggests that the information they had received through public media was insufficient, and that the tasks of the study induced interest about the topic of TS. Adolescents who conceived the tics beyond the individual’s control perceived their peers with TS as being deprived of agency and TS was situated within a discourse of pity. Individuals with the condition were viewed as in need of support and were perceived as people with confined life opportunities. The participants expressed a desire to provide for, care for, and protect individuals with

Tourette’s while simultaneously revealing a dominant relationship towards their subordinated peers with TS. Therefore, a benevolent form of stigma was unveiled; people with TS were viewed as unable to cope with life demands and were in need of a benevolent figure to support them. Although participants expressed a desire to help their peers with TS, they also asserted that they would avoid initiating meaningful social relationships with them due to fear of ‘social contamination’. Indeed, the apprehension of being viewed as tainted and, subsequently, avoided and socially excluded due to the relationship they had with an individual with TS hindered them from including individuals with TS in their social group. On the other hand, adolescents who viewed those with TS as accountable for the condition expressed a plenary desire for social distance. The findings therefore suggested that although adolescents with TS might not be actively bullied within the school, they would be peripheral to the social networks of those without TS. Thus, it would have been challenging for adolescents with TS to form intimate relationships, which is a distinctive characteristic of the relationships made during adolescence and have long and short-term implications for their socio-emotional functioning (Sullivan, 2013).

The findings of Study One pinpointed to the directions the subsequent intervention needed to follow. For example, the nature of control that individuals have over their tics was an important theme derived from the adolescents in study one, which needed to be incorporated into a meaningful intervention, as were the answers to questions adolescents had about the condition. Perhaps most importantly, the discourse of pity and paternalism in relation to TS was an issue which needed to be addressed. The intervention was also informed by the systematic review of previous school-based interventions that aimed to reduce stigma in behaviourally defined conditions (see Chapter 3). This demonstrated that although there were no hard or fast rules in relation to the length of intervention, direct and/or indirect contact and education seemed to be the most effective approaches to reducing stigma.

Informed by all of the above findings, the subsequent two-hour long intervention was therefore developed to consist of three components. Firstly, indirect contact with an individual with TS through the viewing of a film was used to enable participants to identity with their peers with the condition. It also aimed to reduce ‘pity’ towards and ‘disempowerment’ of people with TS. The second component entailed an educational element through a PowerPoint presentation. This aimed to demythologise stereotypes about TS and explain the condition in more detail. Finally, a student led discussion was geared towards answering any questions the participants might have had. The extent to which the intervention worked was the focus of Study Two, outlined below.

8.2. Study Two: Assessment and evaluation of the intervention

The effectiveness of the intervention in increasing adolescents’ knowledge about TS and improving their attitudes and behavioural intentions towards their peers with TS was assessed in Study Two using a randomised cluster control trial with a concurrent mixed-method design. This study also sought to explore differences of gender and previous familiarity with TS and other mental or developmental con in relation to attitudes and behavioural intentions. A total of 59 students from the same school used in Study One (though these adolescents were different to those who participated in the first study) were divided into a waiting-list control group and anti-stigma intervention group. Data were collected at three time points: one week prior to the intervention (T1); immediately after it (T2); and nine weeks later (T3). Similar to Study One, this study was unique in that it was the first to examine the effectiveness of an anti-stigma intervention among secondary school students in the UK.

The results of this study indicated that the intervention produced significant changes in knowledge, attitudes and behavioural intentions upon completion of the intervention. Qualitative findings indicated that the pupils in the intervention group were less eager to define normalcy and more willing to embrace diversity. TS was viewed as a secondary characteristic of an individual that could not overshadow their personality. It was no longer viewed as a problem that society needed to deal with.

The follow-up measures indicated that only the changes in knowledge were maintained after 9 weeks, suggesting that attitudes and behavioural intentions are more resilient to change than knowledge and possibly necessitate a combination of different approaches. Unfortunately, the qualitative data were not sufficient in explaining the reasons why attitudes and behavioural intentions dissipate over time.

The results of the study highlight that stigma around TS is powerful, pervasive and difficult to change. Indeed, stigma is a multi-dimensional construct and although can be reduced through education cannot be radically eliminated solely by that. Furthermore, the study indicated that, although a targeted approach (only a specific group) was used, any future interventions should be more universal and address a wider range of people. The participants of the study were exposed to a positive representation of TS only once and might have been thereafter bombarded with the pejorative stereotypes by which TS has been represented in the social media. Although the intervention could have temporarily changed their perception about TS, their constant contact with their wider social network and their involvement in their social community would have made it very hard for these changes to be sustainable. In other words, the individuals that participate in anti-stigma interventions cannot be secluded or shielded from the wider stigma that is cultivated within society.

Taking on a more universal approach could help reform the culture about TS. In particular the media representations that have a powerful influence on the public attitudes that need to be challenged. Families also may need to be involved as advocates of social change. Changing stigma cannot be done quickly and is a difficult process that requires engagement from different parties. Thus, marked short-term improvement in reported attitudes can be achieved, but maintaining these effects is a major challenge.

While the results of both studies are encouraging and have provided several new insights, there were limitations to the research process which are outlined below.

8.3. Limitations

As acknowledged previously, the main limitation of the research was that its findings cannot be extrapolated to a representative sample of adolescents in the UK. The individuals that took part in the whole study were from a pool of students from a specific school that represented a sample of convenience rather than a probability sample. Whilst the recommended guidelines for contacting schools were followed (Oats & Riaz 2016; Rice et al., 2007): gatekeepers in a range of schools were approached; detailed study information packs were sent out; and face-to-face meetings with key teachers were arranged so that questions and/or concerns about the research could be raised and addressed, only one school agreed to participate. This highlights the difficulties researchers have in accessing an increasingly closed environment. Furthermore, schools are becoming more fearful of over-scrutiny via the Office for Standards in Education (Ofsted) and arguably the antithetical goals that are placed on them: on the one hand, high academic achievement scores (especially GCSE) are demanded of their pupils and, on the other hand, the need for them to respond to the diverse needs of all the students. The very low response rate in participation (more than twenty schools were contacted, out of which only one participated) is also a common but under-discussed finding among school-based studies, especially for early career researchers who have not previously worked in schools, and who are unlikely to have a wide contact network to draw from. Oats and Riaz (2016), who encountered similar difficulties in gaining access to schools, contacted the Cabinet Secretary for Education and Lifelong Learning out of frustration. As a result, a gatekeeper was assigned to the research, who contacted the schools and agencies on behalf of the researcher, introducing the research and its potential impact. This ensured a wider pool of school participation. Future researchers should consider using similar strategies. In order for a more comprehensive and generalizable account of stigma and TS in relation to school-age adolescents, any subsequent study would therefore need to incorporate a wider range of participants from a broader age-range and different regions as well as different schools (e.g. grammar schools, public schools). Nevertheless, despite the rather narrow sample pool employed in this study, given that research of any kind in relation to stigma and adolescents with TS is in its infancy, it can be argued that exploratory trials were warranted before using large scale trials.

Another limitation of the research is its ecological validity due to the artificiality of the stimulus that introduced the participants to the symptomology of TS. Although the video of an individual with TS (stimulus) that was shown to the participants accurately depicted TS, it remained unrepresentative of real world situations. Thus, no other characteristics or attributes of the individual are shown that might have created a more comprehensive and balanced portrayal of the person. This could have been prevented if a naturalistic setting had been used. However, using a naturalistic study design in which a student with TS is enrolled in the class gives rises to other professional and ethical concerns. For example, the student with TS might have been indirectly stigmatised since the rest of the class of pupils could have unconsciously been reflecting their thoughts about the specific person in their answers rather than about the condition. Furthermore, since the study used paper-and-pencil measures rather than direct measures of behaviour, only reported intentions to behave in a certain way were assessed. Although intentions are reliably associated with behaviour (Fishbein, Martin & Icek Ajzen, 1975), directly evaluating the behaviour can provide more ecological validity. Paper-and- pencil measures might have also allowed the participants to provide socially desirable responses. It is common for individuals to over-report socially desirable attitudes and behaviours in questionnaire items to present a more positive image of themselves, instead of responding in an honest manner (Krumpal, 2013). Therefore, although the findings suggest that the intervention increased students' willingness to socialise with individuals with TS, whether the program had an impact on actual behaviour cannot be demonstrated.

Another limitation relating to Study Two was the use of the phrase "feel sorry for" in the attitude questionnaire (Appendix P); the phrase aiming to represent a negative emotion. Alternatively, pity may incorporate a subjective meaning and be regarded as a positive feeling that motivates others to help. In other words, participants in this study might have believed that pity was a benevolent response to a disadvantaged 'other'. As no additional questions relating to pity were included in the attitude questionnaire, it is difficult to assess the meaning of pity each participant had in mind. Future studies need to explore how the notion of pity is conceptualised by adolescents and include more questions relating to it.

The findings of Study Two indicate that, although the participants' attitudes changed immediately after the intervention, the vast majority believed that people with TS could not be well accommodated within mainstream school and a special school would be more appropriate for them. This suggests that pupils with TS might still be disadvantaged, feel undervalued and have a limited sense of belonging in comparison to typically developing adolescents. The question of whether more positive attitudes and behavioural intentions towards individuals with TS automatically translates into a better sense of belonging for those with TS within a mainstream school is yet to be explored and should be addressed. Future research should therefore include measurements of school belonging when implementing an anti-stigma/psychosocial intervention to ascertain whether such strategies have impacted this variable. The results from this research also suggested ways to improve the educational part of the intervention, including placing more emphasis on the law relating to students with disabilities; and how each student is entitled to receive the same type of education irrespective of disability. Mindful of the fact that the students can help to construct the ethos and culture of a school, any educational intervention should address how culture can change through teaching students to be more responsive to diversity.

Future studies also need to include measures to evaluate implicit attitudes pertaining to individuals with Tourette's syndrome to provide a broad evaluation of the anti-stigma impact. The studies in this thesis used explicit (i.e., consciously controllable) measures that may lead to self-presentation bias. Recent studies (Murch et al., 2017; Wilson & Scior, 2014) suggest that stigma may manifest itself more on implicit than explicit measures and that a more thorough assessment of the former measure may be warranted.

Although the studies for this thesis evaluated the stereotypes and prejudice adolescents may have in regards to individuals with TS, they did not investigate what age these attitudes may have emerged. Future studies should trace how and when these prejudices emerge in order to develop preventative and early intervention initiatives instead of reactive interventions targeting already established attitudes. Future anti-stigma interventions need to evaluate the diverse components involved in stigmatisation for each separate population. There is a need to understand the framework in which stigma is created and base the interventions on these variables.

8.4. Future research

The findings of this thesis have pinpointed crucial areas of research that have been understudied and have consequentially guided new research proposals described below.

Firstly, the findings indicated that misconceptions about TS may deprive individuals with TS from basic human rights. The essential question to be explored is whether the rights of people with TS to social inclusion was the only human right being violated or whether access to health and social care were also variables being violated. Indeed, although TS can be associated with mobility difficulties, unemployment issues, lower socioeconomic status, poorer psychosocial functioning and diminished quality of life, it is popularly only known as ‘the cursing disease’. A new study, funded by the NIHR School of Social Care Research (SSCR) (Project Reference C088/CM/UKJB-P118) commenced in October of 2017 with myself as the Principle Investigator (PI). Building on the findings of this PhD thesis, this new funded study is expected to highlight service deficits and merits in relation to this specific population. It also aims to understand the difficulties adults with TS experience and their access to social care, and how receiving care may affect an individual’s everyday life, personhood and embodiment. The objective of this study is to provide data which will also inform current policy and practice regarding Tourette’s syndrome. The SSCR project is a small scale and exploratory study: an England-wide online survey of 50 adults with TS, followed by in-depth interviews from a subsample; 10 interviews with those who receive social care and 10 interviews with those who do not receive social care. Since there is a scarcity of research that examines TS through a social, non-medicalised perspective this new research is innovative and designed using public and patient involvement (PPI), which included several group meetings held via Tourettes Action.

This new SSCR project should therefore alert practitioners to the full range of people’s needs and help identify attributes essential for service delivery. These findings could also further help meet the challenge of responding to people’s holistic needs by recommending how social care services can be improved. It is expected that the findings could also be incorporated into social care training to help professionals’ understanding of TS; the overall objective being to help improve the quality of life of adults with TS.

Secondly, the findings presented in Chapter 2 highlighted that there are currently no empirical studies that assess TS-stigma from the perspective of those who endure stigmatisation. Whilst there is a large body of research that involves self-reports from the public about attitudes towards this population, there are no studies to my knowledge that delve into the first-hand accounts of the experience of TS stigma. There is also a scarcity of knowledge pertaining to the extent, nature and impact of Tourette’s stigma. Similarly, factors that influence the level of stigma have not yet been explored. The coping mechanisms of people with TS when experiencing stigmatisation is also unknown. Thus, no studies to date have evaluated whether people with TS employ specific strategies to protect themselves from rejection and discrimination or ameliorate its consequences by accessing support at an interpersonal level (e.g., informal care and support), or at a governmental/structural level (e.g., social care services, legal and policy interventions, rights-based approaches) or at a community level (e.g., community education). Furthermore, the effectiveness of those support systems in eliminating stigma from the perspective of the stigmatised has not been evaluated. Therefore, a research grant proposal was submitted to the Tourettes Action research award Scheme for 2017/18. The proposal has been successful at the first stage of review and the final results will be announced within the next few months.

Research pertaining to Tourette’s syndrome in relation to stigma could also be introduced to the new Unit for Stigma Research at the University College of London

(UCLUS). This new department is dedicated to generating stigma theory and tackling the stigma of Intellectual and Developmental Disabilities as well as Mental Health Problems. A perspective on the stigma of TS could further advance the theory production in the stigma field.

Finally, dissemination of early findings of this thesis (see Malli. Forrster- Jones & Murphy, 2015; Malli & Forrester-Jones, 2017) led the CEO of Tourettes Action, Suzzane Dobson, to develop a national campaign aimed at raising awareness about TS and dispelling myths and stereotypes about the condition as well as assessing its effectiveness. The intervention will be developed in co-operation with individuals with Tourette's and be based on direct contact with people with TS. The intervention aims to inform the general public through poetry reading, theatre performances, lectures, art exhibitions and screening of movies. Also, specially chosen target groups (e.g. police officers, students, employers, landlords etc.) will be approached in order to receive the intervention. This project has been discussed with the National Lottery Fund and met with great enthusiasm. A full proposal will be submitted within the next few months. Other recommendations for research include evaluating the effectiveness of the intervention in the different groups the intervention has been introduced using randomised control trials.

8.5. Recommendations for practice

The findings presented in this thesis offer a number of practical implications for education and practice. Firstly, it would appear that indirect contact with individuals with TS can be effective in destigmatising the condition. Although this form of contact does not permit two-way communication, it still offers a level of consistency and greater control over the content of the discussion and therefore is appropriate in the context of a school curriculum. The foundations to implementing such an easily deliverable programme appeared to be linked to school staff willingness to collaborate. If secondary school teachers are hesitant to implement the workshop because of their own perceived limited knowledge and confidence about TS, educational seminars for secondary school staff could be added. This 'train the trainers' approach would arguably move the intervention towards a multi-model educational approach involving both teaching staff and students as well as sensitising local educational authorities to TS. An additional component of the intervention might be 'media boxes', similar to the Bavarian Anti-Stigma Action campaign for mental illness (BASTA). These boxes contain background information, movies, books etc. that help teachers understand and familiarise themselves with TS.

Efforts could also be made to integrate this TS intervention into Personal, Social, Health and Economic curriculum (PSHE). The primary aim of PSHE is to promote the well-being of pupils through addressing key issues of concern, currently around drugs and education. It also aims to shape pupils' attitudes and values by providing them with accessible information that enhances their understanding and is orientated towards creating a positive school ethos that enables each student to fulfil their potential (National Children's Bureau, 2006). Taking into consideration that teachers are provided with the flexibility to determine how best to deliver PSHE education and the positive findings of this study, this easily reproduced and adaptable intervention could be both incorporated into PSHE, or teaching methods incorporated within it, could be translated into teaching other PSHE topics.

8.6. Recommendations for policy

This thesis has highlighted that individuals with Tourette's syndrome may be at risk of social exclusion and in turn, face challenges in having their basic human rights met. The right not to be discriminated against, as well as access to health and social care are some of the rights that might be violated (UK Human Rights Act (HRA) 1988) in this population. Although TS may be considered a disability under UK law in accordance to the 2010 Equality Act, there is currently no specific government commitment or policy guidance that safeguards the human rights of this population, ensuring their inclusion in wider society. Thus, there is currently no policy that aims to improve the quality of life of people with TS, such as white papers Valuing People (HM Government, 2001) and Valuing People Now (HM Government, 2009). Furthermore, there is no reference to the condition in the Health and Social Care Act of 2012 indicating a general lack of governmental directives in relation to this population.

Although it is acknowledged that TS is considered a distinct neurological condition, people with the condition can account for up to one percent of the population. This makes the condition almost as prevalent as Autism (Baron-Cohen et al., 2008), signifying the importance of instigating specific policy guidelines for this population.

8.7. Conclusions

The strength of the studies presented in this thesis was their novelty in creating a tailor-made intervention for adolescents and in providing an additional TS anti-stigma intervention. The research demonstrated that indirect contact and education can be a cost-effective method to reduce stigma and misconceptions about TS.

Following an exploratory study to better understand how typically developing adolescents perceive individuals with TS, a tailor-made intervention was designed. However, only knowledge improvement was maintained after 9 weeks, suggesting that changes in attributes and behaviour intentions are more resilient to change. Nevertheless, the intervention had a positive impact in raising awareness about TS and demythologising stereotypes about the condition.

The findings of the research highlight the advantages of creating tailor-made interventions. Understanding the origins of stigma in a specific subgroup of the population can help locate and target specific areas of misconception and prejudice. It also helps to build upon the theory of stigma and understand the process of how it may be formed. Since stigma in TS is a new field, it was necessary to explore the perceptions towards individuals with TS before an intervention could be designed.

Moreover, the research highlights the egregious effects of stigma on individuals with TS but also highlights that misconceptions can be successfully changed. While there were significant changes observed in knowledge, attitudes and behavioural intentions immediately after the intervention, future research needs to concentrate on how these positive results can be maintained over a longer period of time. If stigma is challenged from a young age, discrimination can surely be avoided in the general public and subsequent generations. Finally, lest we forget the people with TS in this thesis, here is a quote from someone with TS from my latest study about the Social Care needs of individuals with TS that highlights the debilitating effects of TS stigma and the need to ameliorate it:

> *“Yeah, the stigma is created by the Daily Mail, and the Mirror and the Sun and all those red banner newspapers. Oh, and Chanel 5…For them I’m just a walking joke…But until somebody gets to know me and they realise that I’m just a man. I’ve got the smarts about me and I’m not just the tics and the swearing and stuff like that. But yeah, I mean people can misinterpret my Tourette’s as* being drunk or being on drugs or something like that. So, all I want is for them *to understand and know what TS is.”*

9

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Appendix A

University of Kent | Tizard Centre

T: +44 (0)1227 827373
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www.kent.ac.uk/tizard

Participants information sheet

Title of Research: **Adolescents' perception of their peers with Tourette's syndrome and their knowledge about the disorder. An exploratory study.**

Researcher: Melina Malli, mm758@kent.ac.uk, 01227 824770

Supervisor: Dr R.V.E Forrester-Jones, R.V.E.Forrester-Jones@kent.ac.uk, 01227 823194

We are inviting you to take part in our research study. Before you decide if you want to take part in the study, it is important that you understand what the study is about and what we will ask you to do**. Please** take time to **read the information carefully** and discuss it with your family or friends if you want to. Feel free to ask us if there is anything you don't understand.

Who are we?

My name is Melina Malli and I am a PhD student at the University of Kent. Dr Rachel Forrester-Jones is my supervisor and she is a Reader at the University of Kent.

What is the study about?

We want to know **your views on people with Tourette's syndrome**. The study has been approved by the Tizard Ethics Committee from the University of Kent.

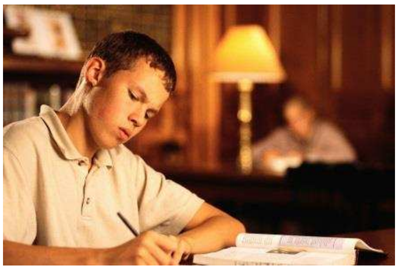
Do I have to take part?

No, it's up to you whether you take part or not. If you decide not to take part you will continue with your curriculum as usual. If you decide to take part you will be asked to sign a consent form. However, you can still change your mind and stop participating in this study at any time without giving a reason.

What will I be asked to do if I decide to take part?

Everybody who takes part in the study gets to see a short video of a teenager who has Tourette's syndrome.

Afterwards you will be asked to write down your views about that person. This will take approximately **10 minutes**.



All the participants in the study will be divided into groups. Each group will be given a number and be seen by the researcher in a random order. In the group you will have the chance to **talk about** your thoughts with your friends and the researcher. You will also get to hear stories about people with Tourette's syndrome. We would like to know your views on those as well. These discussions will last about **45 minutes** and they will be **audio recorded**.

Will my taking part in the study be kept confidential?

Yes. A code will be used in place of your name. All the information pertaining to you will be kept safely locked away in a filing cabinet at the University of Kent. All of the responses to questions will be safely stored in a separate cabinet and only accessed by the research team. **You will not be identified in any of the reports/publications of the project**.

What will happen to the results of the research study?

The information you provide us with will be used to produce part of the researcher's PhD, and might appear in scientific magazines. However, your name will not be included.

What happens now if I decide that I want to take part?

If you agree to take part please sign the assent form.

Who to contact

If you or your parent(s)/guardian(s) have any questions about taking part in the study please do not hesitate to get in contact with hesitate to get in contact with me on **01227 824770** or email mm758@kent.ac.uk. You can also speak to my supervisor, Dr Rachel Forrester-Jones on 01227823194 or e-mail R.V.E.Forrester-Jones@kent.ac.uk.

Thank you for reading the information sheet!

Appendix B

University of Kent | Tizard Centre

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Information sheet for Head Teacher

Study Tittle: Adolescents' perception of their peers with Tourette's syndrome and their knowledge about the disorder. An exploratory study.

Researcher: Melina Malli, mm758@kent.ac.uk, 01227 824770

Supervisor: Dr R.V.E Forrester-Jones, R.V.E.Forrester-Jones@kent.ac.uk, 01227 823194

This document explains what we are doing in the research project and sets out what will be involved in the school.

Who are we?

My name is Melina Malli and I am a PhD student at the University of Kent. Dr Rachel Forrester-Jones is my supervisor and she is a Reader at the University of Kent.

What is the purpose of this study?

We want to find out **adolescents' views on people with Tourette's syndrome**. The study has been **approved by the Tizards ethics Committee from the University of Kent**.

What sort of children do we need?

As part of the research we are looking for a **Year Ten** class comprising **of 20-35 adolescents** between 14 to 15 years of age. It is imperative for the study that **none of the children in that particular class have Tourette's syndrome** in order to avoid stigmatisation. It is also important that all the participants of the study can speak English fluently.

Who will give consent for the child to take part?

The researchers will adhere **to school policy regarding good practice and parental consent**.

What will be involved?

We will take every care to reduce to minimum disruption to school routine. Initially every student in the class that has chosen to participate in the study gets to **see a short video of a teenager who has Tourette's syndrome**. This video should **not last more than 5 minutes**. Afterwards the participants will be asked to **write down their views about that person**. This will take approximately **10 minutes.**

Their teacher will be asked to divide all the participants into groups of 6 to 8 pupils based on their friendships. It is anticipated that 4-5 focus groups will be formed for the purpose of the study. Each group will be given a number and be seen by the researcher in a random order. These groups will be taken out of the class during regular classroom activities into a comfortable spare space within the school. In the groups they will have the chance to **talk about their thoughts** about people with Tourette's syndrome with their friends and the researcher. They will also get to hear stories about people with Tourette's syndrome. We would like to know their views on those as well. **These discussions will last about 45 minutes and they will be audio recorded**.

Since the project will run through regular class hours, an alternative school activity will be provided to the pupils that choose not to consent to participate.

Who will run the research sessions?

The research team would like to assure you that all the researchers coming into the school will have CRB clearance for working with children. Melina Malli will be the main researcher and running the group discussions.

Will all pupils' details and responses be kept confidential?

Yes. A code will be used in place of the pupils' names. All the information pertaining to the pupils will be kept safely locked away in a filing cabinet at the University of Kent. All of the responses to questions will be safely stored in a separate cabinet and only accessed by the research team**. The pupils and the school will not be identified in any of the reports/publications of the project**.

Who to contact

If you have any queries about any aspect of the study please do not hesitate to get in contact with me on 01227 824770 or email mm758@kent.ac.uk. You can also speak to my supervisor, Dr Rachel Forrester-Jones on 0122783194 or e-mail R.V.E .Forrester-Jones@kent.ac.uk.

Thank you for reading the information sheet!

Appendix C

University of Kent | Tizard Centre

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Parent(s)/ guardian(s) information sheet

Study Tittle: Adolescents' perception of their peers with Tourette's syndrome and their knowledge about the disorder. An exploratory study.

Researcher: Melina Malli, mm758@kent.ac.uk, 01227 824770

Supervisor: Dr R.V.E Forrester-Jones, R.V.E.Forrester-Jones@kent.ac.uk, 01227 823194

Your child is being invited to take part in a research study. Before you decide whether or not to let your child participate in this study it is important for you to understand why the research is being done and what it will involve. **Please take time to read the following information** carefully and discuss it with others. Ask us if there is anything that is not clear or if you would like more information.

Who are we?

My name is Melina Malli and I am a PhD student at the University of Kent. Dr Rachel Forrester-Jones is my supervisor and she is a Reader at the University of Kent.

What is the purpose of the study?

We want to find out **adolescents' views on their peers with Tourette's syndrome.** The study has been approved by the Tizard Ethics Committee of the University of Kent.

Does my child have to take part?

It is up to you to decide whether or not your child can take part. You are free to withdraw your child at any time and without giving a reason by contacting Mr Somers. A decision to withdraw, or a decision not to take part will not result in any negative consequences.

What if my child does not want to participate?

In addition to your permission, your child must agree to participate in the study. If you child does not want to participate she/he will not be included in the study and there will be no penalty. If your child initially agrees to be in the study they can change their mind later without any negative consequences.

What is my child going to be asked to do?

Everybody who takes part in the study gets to see a short video of a teenager who has Tourette's syndrome. Afterwards they will be asked to **write down their views** about that person. This will take approximately **10 minutes.**

All the participants in the study will later be **divided into groups**. Each group will be given a number and be seen by the researcher in a random order. In the groups they will have the **chance to talk about their thoughts about people with Tourette's syndrome** with their friends and the researcher. They will also get **to hear stories about people with Tourette's syndrome**. **We would like to know their views on those as well**. These discussions will last about **45 minutes and they will be audio recorded**.

How will your child's privacy and confidentiality be protected if s/he participates in this research study?

A code will be used in place of your child's name. All the information pertaining to your child will be kept safely locked away in a filing cabinet at the University of Kent. All of the responses to the questions will be safely stored in a separate cabinet and only accessed by the research team. **Your child will not be identified in any of the reports/publications of the project**.

What will happen to the results of the research study?

The information the adolescents provide us with will be used to produce part of the researcher's PhD, and might appear in scientific magazines. However, their names will not be included.

What happens now if I decide that my child doesn't want to take part?

If you don't agree for your child to take part **please return the slip**.

Who to contact

If you have any queries about any aspect of the study please **do not hesitate** to get in contact with me on **01227 824770** or email me at **mm758@kent.ac.uk**. You can also speak to my supervisor, Dr Rachel Forrester-Jones on 0122783194 or e-mail R.V.E.Forrester-Jones@kent.ac.uk**.**

Thank you for reading the information sheet!

Appendix D

University of Kent
Tizard Centre

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Assent Form

Title of Research: **Adolescents' perception of their peers with Tourette's syndrome and their knowledge about the disorder. An exploratory study.**

Researcher: Melina Malli, mm758@kent.ac.uk, 01227 824770

Supervisor: Dr R.V.E Forrester-Jones, R.V.E.Forrester-Jones@kent.ac.uk, 01227 823194

Please sign this form if you

- ☐ have understood what this project is about
- ☐ have understood what you will be asked to do in the study
- ☐ have asked all the questions you want
- ☐ have had your questions answered
- ☐ have talked to your parent(s)/ guardian(s) about this project
- ☐ are happy to take part in the research

Your signature Printed Name

Date

Appendix E

University of Kent
Tizard Centre

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www.kent.ac.uk/tizard

Consent form for parents/carer

Title of the study : Adolescents' perception of their peers with Tourette's syndrome and their knowledge about the disorder. An exploratory study.

Researcher: Melina Malli, mm758@kent.ac.uk, 01227 824770

Supervisor: Dr R.V.E Forrester-Jones, R.V.E.Forrester-Jones@kent.ac.uk, 01227 823194

Dear Parent/Carer

I am writing to ask if your child would take part in the above study.

Students participating will:

- particpate in a group discussion with the researcher and some of his/her classmates that will be audio- recorded.
- complete an anonymised free text writing exercise.

Please contact me if you DO NOT wish them to take part.

Andy Somers

Principal

Appendix F

Vignettes and interview schedule of the focus groups

1) Pat and Pat's friends are hanging out during break time, just socializing and having fun. Jamie wanders near them and it is apparent to Pat that Jamie wishes to hang out with them as well. Pat is considering asking Jamie to join them, but decides not to because of Jamie's tics.

2) Chris is alone at home and eager to watch a film on the TV. None of Chris's friends are available but Chris does want the company. Bobbie is the next door neighbour that has happened to pay him a visit to ask Chris something about school homework. It's obvious that Bobbie wants to watch the movie with Jamie. Chris is considering asking Bobbie to stay but decides not to because of Bobbie's tics.

Interview schedule

Do you think it is ok that Jamie/ Bobby was not invited?

Why do you think it was ok/was not ok?

What would you do in Pat's/Chris's shoes?

Do you think she/he would need your help or would he be able to sort it out by himself?

What do you know about Tourette's syndrome?

What is your main source of information about the disorder? Do you think it's their fault they act the way they do?

Do you think a person with Tourette's is as capable as any other person?

Would it be easy for you to hang out with somebody with Tourette's syndrome?

Would you be comfortable presenting him/her to your other friends?

Do you think it would be easy for you to fall in love with somebody with Tourette's syndrome?

Appendix G

Researcher: Melina Malli, mm758@kent.ac.uk, 01227 824770

Supervisor: Dr R.V.E Forrester-Jones, R.V.E.Forrester-Jones@kent.ac.uk, 01227 823194

Participants' information sheet

Group A

Evaluating a school-based Educational Workshop about Tourette's syndrome for Adolescents

---

We are inviting you to take part in our research study. **Before you decide** if you want to take part in the study, it is important that you understand what the study is about and what we will ask you to do. **Please read the information carefully and discuss** it with your family or friends if you want to. Feel free to ask us if there is there is anything you don't understand.

Who are we?

My name is Melina Malli and I am a PhD student at the University of Kent. Dr Rachel Forrester-Jones is my supervisor and she is a Reader at the University of Kent.

What is the study about?

We want to know whether an educational workshop could add to your knowledge and understanding about Tourette's syndrome. The study has been approved by the Tizard Centre Ethics Committee from the University of Kent.

Do I have to take part?

No, it's up to you whether you take part or not. If you decide not to take part please inform your teacher or Melina and we will make sure you are not included in the study. In this case you will continue with your curriculum as usual. If we do not hear from you, you will be included in the study. However, you can still change your mind and stop participating at any stage of the study without giving a reason up to the point when the information has been analysed anonymously.

What happens now if I decide to take part?

If you decide to take part in the study you will watch a short video of a teenager who has Tourette's syndrome (3-4 minutes). Afterwards you will be asked to **fill in questionnaires** regarding your knowledge about Tourette's syndrome and your perception of people with Tourette's syndrome. They will only take approximately 20 minutes to complete. We will give you the same questionnaires to complete before the workshop, immediately after the workshop, and 3-4 months after the workshop. This will help us find out if the workshop was useful and discover ways to make it better in the future.

The workshop: During the workshop you will get to:

- watch a film about a person with Tourette's syndrome (Film title: Front of the Class) and
- learn about the disorder through a short educational PowerPoint presentation and a discussion with the researcher.

The workshop should not take more than 2 hours.

What are the possible disadvantages and risks of taking part?

Although the workshop is designed to be a positive experience, some may find that the film might be emotional. However, the teacher, the school counsellor and I will be available to offer support if you are feeling upset. Please keep in mind that you can stop participating at any time up to the point when the information has been analysed anonymously.

Will my taking part in the study be kept confidential?

Yes. A code will be used in place of your name. All the information about you will be kept safely locked away in a filing cabinet at the **University of Kent and in password protected encrypted electronic format**. All of the responses to the questions will be safely stored in a separate cabinet and only accessed by the research team. **You will not be identified in any of the reports/publications about the project.**

What will happen to the results of the research study?

The information you provide will be used to produce part of my PhD, and might appear in scientific magazines. At the end of the study an accessible summary of the results will be sent to the school with a request that the report be distributed to all the pupils who participated in the study. However, your name will not be included.

Who to contact

If you or your parent(s)/guardian(s) have any questions about taking part in the study please do not hesitate to get in contact with me on **01227 824770** or email mm758@kent.ac.uk. You can also speak to my supervisor, Dr Rachel Forrester-Jones on **01227 823194** or e-mail R.V.E.Forrester-Jones@kent.ac.uk. If you would like to speak to someone outside of the research team, you can contact the Tizard Centre Ethics Committee by contacting Jo Ruffels, j.ruffels@kent.ac.uk.

Thank you for reading the information sheet!!

Appendix H

Researcher: Melina Malli, mm758@kent.ac.uk, 01227 824770

Supervisor: Dr R.V.E Forrester-Jones, R.V.E.Forrester-Jones@kent.ac.uk, 01227 823194

Participants' information sheet

Group B

Evaluating a school-based Educational Workshop about Tourette's syndrome for Adolescents

---

We are inviting you to take part in our research study. **Before you decide** if you want to take part in the study, it is important that you understand what the study is about and what we will ask you to do. **Please read the information carefully and discuss** it with your family or friends if you want to. Feel free to ask us if there is there is anything you don't understand.

Who are we?

My name is Melina Malli and I am a PhD student at the University of Kent. Dr Rachel Forrester- Jones is my supervisor and she is a Reader at the University of Kent.

What is the study about?

We want to know whether an educational workshop could add to your knowledge and understanding about Tourette's syndrome. The study has been approved by the Tizard Centre Ethics Committee from the University of Kent.

Do I have to take part?

No, it's up to you whether you take part or not. If you decide not to take part please inform your teacher or Melina and we will make sure you are not included in the study. In this case you will continue with your curriculum as usual. If we do not hear from you, you will be included in the study. However, you can still change your mind and stop participating at any stage of the study without giving a reason up to the point when the information has been analysed anonymously.

What happens now if I decide to take part?

If you decide to take part in the study you will watch a short video of a teenager who has Tourette's syndrome (3-4 minutes). Afterwards you will be asked to **fill in questionnaires** regarding your knowledge about Tourette's syndrome and your **perception of people with Tourette's syndrome**. They will only take approximately 20 minutes to complete and will help us find out if the workshop was useful and discover ways to make it better in the future. We will give you the same questionnaires to complete three times within the next 3-4 months.

After 3-4 months the workshop will take place.

The workshop: **During the workshop you will get to:**

- watch a film about a person with Tourette's syndrome (Film title: Front of the Class) and
- learn about the disorder through a short educational PowerPoint presentation and a discussion with the researcher.

The workshop should not take more than **2 hours**.

What are the possible disadvantages and risks of taking part?

Although the workshop is designed to be a positive experience, some may find that the film might be emotional. However, the teacher, the school counsellor and I will be available to offer support if you are feeling upset. Please keep in mind that you can stop participating at any time up to the point when the information has been analysed anonymously.

Will my taking part in the study be kept confidential?

Yes. A code will be used in place of your name. All the information about you will be kept safely locked away in a filing cabinet at the **University of Kent and in password protected encrypted electronic format**. All of the responses to the questions will be safely stored in a separate cabinet and only accessed by the research team. **You will not be identified in any of the reports/publications about the project.**

What will happen to the results of the research study?

The information you provide will be used to produce part of my PhD, and might appear in scientific magazines. At the end of the study an accessible summary of the results will be sent to the school with a request that the report be distributed to all the pupils who participated in the study. However, your name will not be included.

Who to contact

If you or your parent(s)/guardian(s) have any questions about taking part in the study please do not hesitate to get in contact with me on **01227 824770** or email **mm758@kent.ac.uk**. You can also speak to my supervisor, Dr Rachel Forrester-Jones on **01227 823194** or e-mail **R.V.E.Forrester-Jones@kent.ac.uk**. If you would like to speak to someone outside of the research team, you can contact the Tizard Centre Ethics Committee by contacting Jo Ruffels, j.ruffels@kent.ac.uk.

Thank you for reading the information sheet!

Appendix I

Researcher: Melina Malli, mm758@kent.ac.uk, 01227 824770

Supervisor: Dr R.V.E Forrester-Jones, R.V.E.Forrester-Jones@kent.ac.uk, 01227 823194

Information sheet for Parent(s)/ Guardian(s)

Group A

Evaluating a school-based Educational Workshop about Tourette's syndrome for Adolescents

---

We are inviting your child to take part in a research study. Before you decide whether or not to let your child participate in this study it is important for you to understand why the research is being done and what it will involve. **Please take the time to read the following information** carefully and discuss it with others. Ask us if there is anything that is not clear or if you would like more information.

Who are we?

My name is **Melina Malli** and I am a **PhD student** at the University of Kent. **Dr Rachel Forrester-Jones** is my supervisor and she is a **Reader at the University of Kent**.

What is the purpose of the study?

We want to know **whether an educational workshop about Tourette's syndrome is useful for educational and social purposes. The study has been approved by the Tizard Centre Ethics Committee from the University of Kent.**

Does my child have to take part?

It is up to you to decide whether or not your child can take part. If you decide that they cannot take part please sign the Consent form that says this and your child will continue with his/her curriculum as usual. If you do not sign this form your child will be included in the study. However, you are free to withdraw your child or your child's data at any stage of the study and without giving a reason up to the point when the information has been analysed anonymously. A decision to withdraw, or a decision not to take part will not result in any negative consequences.

What if my child does not want to participate?

In addition to your permission, your child must agree to participate in the study. If your child does not want to participate she/he will not be included in the study and there will be no penalty. If your child initially agrees to be in the study they can change their mind later without any negative consequences.

What is my child going to be asked to do?

If you choose for your child to take part in the study she/he will view a short video of a teenager who has Tourette's syndrome (3-4 minutes). Afterwards we will be asking your child to **complete questionnaires** regarding her/his knowledge about Tourette's syndrome and his/ her perception of people with Tourette's syndrome. The questionnaires will take approximately 20 minutes to complete. They will be distributed at three time points; before the workshop, immediately after the workshop and 3-4 months after the workshop. This will help us find out if the workshop was useful and discover ways to make it better in the future.

The workshop entails viewing a film about a person with Tourette's syndrome (Film title: Front of the Class) and learning about the subject through a short educational PowerPoint presentation and a discussion with the researcher. The workshop should not take more than 2 hours and will be implemented during the students' regular school hours.

What are the possible disadvantages and risks of my child taking part in the workshop?

Although the workshop is designed to be a positive experience for your child, some may find that the film might be emotional. However, the researcher, the teacher and the school counsellor will be available to offer support if your child is feeling upset. Please keep in mind that your child can stop participating at any time up to the point when the information has been analysed anonymously.

How will my child's privacy and confidentiality be protected if she/he participates in this research study?

A code will be used in place of your child's name. All the information pertaining to your child will be kept safely locked away in a filing cabinet at the University of Kent and in password protected encrypted electronic format. All of the responses to the questions will be safely stored in a separate cabinet and only accessed by the research team. **Your child will not be identified in any of the reports/publications about the project**.

What will happen to the results of the research study?

The information the adolescents provide will be used to produce part of the researcher's PhD, and might appear in scientific magazines. At the end of the study an accessible summary of the results will be sent to the school with a request that the report be distributed to all the pupils who participated in the study. However, their names will not be included.

Who to contact

If you have any queries about any aspect of the study please **do not hesitate** to get in contact with me on **01227 824770** or email me at **mm758@kent.ac.uk**. You can also speak to my supervisor, Dr Rachel Forrester-Jones on **01227 823194** or e-mail **R.V.E.Forrester-Jones@kent.ac.uk. If you would like to speak to someone outside of the research team, you can contact the Tizard Centre Ethics Committee by contacting Jo Ruffels, j.ruffels@kent.ac.uk.**

Thank you for reading the information sheet!

Appendix J

Researcher: Melina Malli, mm758@kent.ac.uk, 01227 824770

Supervisor: Dr R.V.E Forrester-Jones, R.V.E.Forrester-Jones@kent.ac.uk, 01227 823194

Information sheet for Parent(s)/Guardian(s)

Group B

Evaluating a school-based Educational Workshop about Tourette's syndrome for Adolescents

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We are inviting your child to take part in a research study. Before you decide whether or not to let your child participate in this study it is important for you to understand why the research is being done and what it will involve. **Please take the time to read the following information** carefully and discuss it with others. Ask us if there is anything that is not clear or if you would like more information.

Who are we?

My name is **Melina Malli** and I am a PhD student at the University of Kent. **Dr Rachel Forrester-Jones** is my supervisor and she is a **Reader at the University of Kent**.

What is the purpose of the study?

We want to know **whether an educational workshop about Tourette's syndrome is useful for educational and social purposes. The study has been approved by the Tizard Centre Ethics Committee from the University of Kent.**

Does my child have to take part?

It is up to you to decide whether or not your child can take part. If you decide that they cannot take part please sign the Consent form that says this and your child will continue with his/her curriculum as usual. If you do not sign this form your child will be included in the study. However, you are free to withdraw your child or your child's data at any stage of the study and without giving a reason up to the point when the information has been analysed anonymously. A decision to withdraw, or a decision not to take part will not result in any negative consequences.

What if my child does not want to participate?

In addition to your permission, your child must agree to participate in the study. If your child does not want to participate she/he will not be included in the study and there will be no penalty. If your child initially agrees to be in the study they can change their mind later without any negative consequences.

What is my child going to be asked to do?

If you choose for your child to take part in the study she/he will view a short video of a teenager who has Tourette's syndrome (3-4 minutes). Afterwards we will be asking them to **complete questionnaires** regarding their knowledge about Tourette's syndrome and their perception of people with Tourette's syndrome. This will take approximately 20 minutes to complete and will help us find out if the workshop was useful and discover ways to make it better in the future. The same questionnaires will be distributed three times within the next 3-4 months.

After 3-4 months the workshop will take place. **The workshop** entails viewing a film about a person with Tourette's syndrome (Film title: Front of the Class) and learning about the subject through a short educational PowerPoint presentation and a discussion with the researcher. The workshop should not take more than 2 hours and will be implemented during the students' regular school hours.

What are the possible disadvantages and risks of my child taking part in the workshop?

Although the workshop is designed to be a positive experience for your child, some may find that the film might be emotional. However, the researcher, the teacher and the school counsellor will be available to offer support if your child is feeling upset. Please keep in mind that your child can stop participating at any time up to the point when the information has been analysed anonymously.

How will my child's privacy and confidentiality be protected if she/he participates in this research study?

A code will be used in place of your child's name. All the information pertaining to your child will be kept safely locked away in a filing cabinet at the University of Kent and in password protected encrypted electronic format. All of the responses to the questions will be safely stored in a separate cabinet and only accessed by the research team. **Your child will not be identified in any of the reports/publications about the project**.

What will happen to the results of the research study?

The information the adolescents provide will be used to produce part of the researcher's PhD, and might appear in scientific magazines. At the end of the study an accessible summary of the results will be sent to the school with a request that the report be distributed to all the pupils who participated in the study. However, their names will not be included.

Who to contact

If you have any queries about any aspect of the study please **do not hesitate** to get in contact with me on **01227 824770** or email me at **mm758@kent.ac.uk**. You can also speak to my supervisor, Dr Rachel Forrester-Jones on **01227 823194** or e-mail **R.V.E.Forrester-Jones@kent.ac.uk.** If you would like to speak to someone outside of the research team, you can contact the Tizard Centre Ethics Committee by contacting Jo Ruffels, j.ruffels@kent.ac.uk.

Thank you for reading the information sheet!

Appendix K

Researcher: Melina Malli, mm758@kent.ac.uk, 01227 824770

Supervisor: Dr R.V.E Forrester-Jones, R.V.E.Forrester-Jones@kent.ac.uk, 01227 823194

Consent form for Parent(s)/ Guardian(s)

Evaluating a school-based Educational Workshop about Tourette's syndrome for Adolescents

Researcher: Melina Malli, mm758@kent.ac.uk, 01227 824770

Supervisor: Dr R.V.E Forrester-Jones, R.V.E.Forrester-Jones@kent.ac.uk, 01227 823194

Dear Parent/Carer,

If you have read the **Information Sheet** and **understood what the study is about and DO NOT wish for your child to take part, please complete this form or alternatively contact the headteacher of the school.**

Please initial in the space provided below:

1. I confirm that I have read and understand the information sheet for the above study. ………………….

2. I **DO NOT** wish for my child to take part in the above study. …………………

Name of parent:

Name of child:

Signature: Date

Appendix L

Researcher: Melina Malli, mm758@kent.ac.uk, 01227 824770

Supervisor: Dr R.V.E Forrester-Jones, R.V.E.Forrester-Jones@kent.ac.uk, 01227 823194

Information sheet for Headteacher

Evaluating a school-based Educational Workshop about Tourette's syndrome for Adolescents

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This document explains what we are doing in the research project and sets out what will be involved in the school.

Who are we?

My name is **Melina Malli** and I am a **PhD** student at the **University of Kent**. **Dr Rachel Forrester-Jones** is my supervisor and she is a **Reader** at the **University of Kent.**

What is the purpose of this study?

We want to know whether an educational workshop about Tourette's syndrome is useful for educational and social purposes. The study has been approved by the Tizards Centre Ethics Committee from the University of Kent.

What does the study entail?

We will take every care to **to keep disruption in the school to a minumum.** Initially every student in the class that has chosen to participate in the study will see a short video of a teenager who has Tourette's syndrome. This video should not last more than 3-4 minutes. Afterwards we will be asking them to **complete questionnaires** regarding their knowledge about Tourette's syndrome and their perception of people with Tourette's syndrome. They will take approximately 20 minutes to complete and will be distributed three times to the participants. This will help us find out if the workshop is useful and find ways to make it better in the future.

During the workshop the pupils will **watch a film about a person with Tourette's syndrome** (Film title: Front of the Class) and learn about the disorder through a short educational Powerpoint presentation and a discussions with the researcher. The workshop should not take more than 2 hours.

What will be involved?

The study requires **two year ten classes to take part**. **One of the classes will do the workshop right away and the other will do it 3 to 4 months later**. All of the pupils that take part in the study will also be asked to complete questionnaires at three time points.

This table will show you when both classes will do each thing throughout the study:

	The class that does the workshop right away	**The class that does the workshop 3 to 4 months later**
Complete the questionnaires	✓	✓
One week later participate in the workshop	✓	✗
Complete the questionnaires immediately after the workshop	✓	✓
Complete the questionnaires **3 to 4 months later**	✓	✓
Participate in the workshop 3 to 4 months later	✗	✓

What are the possible disadvantages and risks for the pupils taking part in the workshop?

Although the workshop is designed to provide a positive experience to the participants, some may find that the film might be emotional. In light of that possibility, it is suggested that a teacher and the school counsellor are available to offer support.

Who will run the workshop?

The workshop will be run by myself. I am a qualified teacher with many years of experience working with adolescents and hold a DBS clearance for working with pupils.

Who do we want to take part?

As part of the research we are looking for **two year ten classes** consisting of **20-35 adolescents** aged 14 to 15 years old. It is imperative for the study that **none of the children in those particular classes have Tourette's syndrome** in order to avoid stigmatisation. It is also important that all the participants can speak English.

Who will give consent for the child to take part?

The researchers will adhere **to school policy regarding good practice and parental consent**.

Will all pupils' details and responses be kept confidential?

Yes. A code will be used in place of the pupils' names. All the information pertaining to the pupils will be kept safely locked away in a filing cabinet at the University of Kent and in password protected encrypted electronic format. All of the responses to the questionnaires will be safely stored in a separate cabinet and only accessed by the research team**. The pupils and the school will not be identified in any of the reports/publications of the project**.

Who to contact

If you have any queries about any aspect of the study please do not hesitate to get in contact with me on **01227 824770** or email **mm758@kent.ac.uk**. You can also speak to my supervisor, Dr Rachel Forrester-Jones on **01227 823194** or e-mail **R.V.E.Forrester-Jones@kent.ac.uk**. If you would like to speak to someone outside of the research team, you can contact the Tizard Centre Ethics Committee by contacting Jo Ruffels, j.ruffels@kent.ac.uk.

Thank you for reading the information sheet!

Appendix M

Demographic information

I would first like to obtain some background information about you

Age………………………………………………

Gender:

Male ☐

Female ☐

……………………………………………………………………………………………………

How would you describe your ethnicity? Please circle the response that best describes your ethnicity

White British
Any other white background
Black British
Any other black background
Asian British
Any other Asian background
Roma
Czech
Slovak
Mixed Background
Other **Please specify**

What is your first language?

Appendix N

Level of contact with people with Tourette's syndrome and other intellectual / developmental disabilities and mental illnesses Questionnaire

Please read each of the questions below and circle the response that best describes your level of contact.

1. How much contact have you had with people with Tourette's syndrome?	
1.	I have never observed a person with Tourette's syndrome.
2.	I have watched a TV show that included a person with Tourette's syndrome.
3.	I have been in a class with a person with Tourette's syndrome.
4.	A friend of the family / of mine has Tourette's syndrome.
5.	I have a relative with Tourette's syndrome.
6.	I live with a person with Tourette's syndrome.
7.	I have Tourette's syndrome.

2. How much contact have you had with people with intellectual/developmental disabilities (e.g. Autism, ADHD)?	
1.	I have never observed a person with an intellectual/developmental disability.
2.	I have watched a TV show that included a person with an intellectual/developmental disability.
3.	I have been in a class with a person with an intellectual/developmental disability.
4.	A friend of the family/of mine has an intellectual/developmental disability.
5.	I have a relative with an intellectual/developmental disability.
6.	I live with a person with an intellectual/developmental disability.

7.	I have an intellectual/developmental disability.
3. How much contact have you had with people with mental illnesses (e.g. depression, anxiety disorder)?	
1.	I have never observed a person with a mental illness.
2.	I have watched a TV show that included a person with a mental illness.
3.	I have been in a class with a person with a mental illness.
4.	A friend of the family/of mine has a mental illness.
5.	I have a relative with a mental illness.
6.	I live with a person with a mental illness.
7.	I have a mental illness.

Appendix O

THE TOURETTE'S KNOWLEDGE QUESTIONNAIRE

We would like to know what you know about Tourette's syndrome. Please circle one answer for each question.

1. Tourette's syndrome is caused by really bad experiences during childhood.
   NOT TRUE TRUE DON'T KNOW

2. Everyone who has Tourette's syndrome swears.
   NOT TRUE TRUE DON'T KNOW

3. You can catch Tourette's syndrome from other people.
   NOT TRUE TRUE DON'T KNOW

4. During adulthood the tics get worse.
   NOT TRUE TRUE DON'T KNOW

5. Lots of people who have Tourette's syndrome have other disorders as well.
   NOT TRUE TRUE DON'T KNOW

6. Tourette's syndrome runs in families.
   NOT TRUE TRUE DON'T KNOW

7. Everyone who has tics has Tourette's syndrome.
   NOT TRUE TRUE DON'T KNOW

8. People with Tourette's syndrome can never control their tics.
   NOT TRUE TRUE DON'T KNOW

9. Tourette's syndrome is

A mental illness
A condition that has do with the brain and the nervous system
A physical disability

10. About ____________ children in England have Tourette's syndrome.
1/10
1/100
1/1000

11. In order to be diagnosed as having Tourette's syndrome you have to
swear uncontrollably
have both movement tics and tics that produce sounds
have tics that produce sounds and swear uncontrollably

12. The tics usually start
before the age of 3
before the age of 18
after the age of 18

13. Tourette's syndrome affects
Only individuals with special educational needs
Only individuals who crave attention
Can potentially affect anyone

14. How long does a person have to have symptoms before they are diagnosed with Tourette's syndrome?
Five years
One year
Six months

15. How is Tourette's syndrome diagnosed?
By an MRI
By a blood test
By the symptoms

16. What is the best school environment for a teenager with Tourette's syndrome?
Home education
Schools for children with similar issues
A mainstream school

17. The activities and the environment an individual with Tourette's syndrome is in may affect how often the tics occur and how intense they are.

NOT TRUE	TRUE	DON’T KNOW

18. Tourette’s syndrome is most effectively treated with medication.

NOT TRUE	TRUE	DON’T KNOW

Appendix P

Attitude Questionnaire

Below are a number of statements regarding attitudes towards people with Tourette's syndrome. Please circle how much you agree or disagree with each of the following statements and explain why.

1. *"Having a person like that in my class would be very annoying"*

1. Strongly Disagree   2.Disagree   3. Neutral   4. Agree   5. Strongly Agree

Why?____
____
____
____

2. *"People with Tourette's syndrome are just like everybody else and the tics shouldn't stop them doing what they want to do"*

1. Strongly Disagree   2.Disagree   3. Neutral   4. Agree   5. Strongly Agree

Why?____
____
____
____

3. *"Just because they have Tourette's you shouldn't treat them differently"*

1. Strongly Disagree   2.Disagree   3. Neutral   4. Agree   5. Strongly Agree

Why?____
____
____
____

4. “ *It makes me feel sad as they can’t have a normal life*”

1. Strongly Disagree	2.Disagree	3. Neutral	4. Agree	5. Strongly Agree

Why?____
____
____
____

5. “*When I see a person with Tourette’s syndrome, I feel very scared*”

1. Strongly Disagree	2.Disagree	3. Neutral	4. Agree	5. Strongly Agree

Why?____
____
____
____

6. “*I wouldn't think of people with Tourette’s syndrome any different, wouldn't look at them any different*”

1. Strongly Disagree	2.Disagree	3. Neutral	4. Agree	5. Strongly Agree

Why?____
____
____
____

7. *"If the tics are really bad, then they should go to another school"*

1. Strongly Disagree	2.Disagree	3. Neutral	4. Agree	5. Strongly Agree

Why?____
____
____
____

8. *"People shouldn't look down on a person because she has tics, they're not going to ruin their company"*

1. Strongly Disagree	2.Disagree	3. Neutral	4. Agree	5. Strongly Agree

Why?____
____
____
____

9. *"If a person with Tourette's syndrome wanted a job to speak in front of people he wouldn't be able to do it"*

1. Strongly Disagree	2.Disagree	3. Neutral	4. Agree	5. Strongly Agree

Why?____
____
____
____

10. *“People with Tourette’s syndrome can do just as well in school ”*

1. Strongly Disagree	2.Disagree	3. Neutral	4. Agree	5. Strongly Agree

Why?____
____
____
____

Appendix Q

Behavioural Intentions Questionnaire

Please circle the answer that shows how you feel about doing each of these things with your peers with Tourette's syndrome.

1. How likely is it that you would go to the cinema with a person with Tourette's syndrome?

1. Not likely at all	2. Unlikely	3. Unsure	4. Likely	5. Very likely

Why do you say that?

____
____
____
____

2. How likely is it that you would want to sit next to a person with Tourette's syndrome in class?

1. Not likely at all	2. Unlikely	3. Unsure	4. Likely	5. Very likely

Why do you say that?

____
____
____

3. How likely is it that you would want a person with Tourette's syndrome as a close friend?

1. Not likely at all	2. Unlikely	3. Unsure	4. Likely	5. Very likely

Why do you say that?

____

____

____

4. How likely is it that you would want to date a person with Tourette's syndrome?

1. Not likely at all	2. Unlikely	3. Unsure	4. Likely	5. Very likely

Why do you say that?

____

____

____

5. How likely is it that you would want to go to a shopping centre with a person with Tourette's syndrome?

1. Not likely at all	2. Unlikely	3. Unsure	4. Likely	5. Very likely

Why do you say that?

____

____

____

____

6. How likely is it that you would want to work on an assignment with a person with Tourette's syndrome?

1. Not likely at all	2. Unlikely	3. Unsure	4. Likely	5. Very likely

Why do you say that?

____

____

____

7. How likely is it that you would ask a person with Tourette's syndrome to come over to your house to spend time together?

1. Not likely at all	2. Unlikely	3. Unsure	4. Likely	5. Very likely

Why do you say that?

____

____

____

8. How likely is it that you would introduce a person with Tourette’s syndrome to your friends?

1. Not likely at all	2. Unlikely	3. Unsure	4. Likely	5. Very likely

Why do you say that?

____

____

____

9. How likely is it that you would introduce a person with Tourette’s syndrome to your parents?

1. Not likely at all	2. Unlikely	3. Unsure	4. Likely	5. Very likely

Why do you say that?

____

____

____

10. How likely is it that you would spend time with a person with Tourette’s syndrome during lunch and break time at school (i.e. sit and talk, etc.)?

1. Not likely at all	2. Unlikely	3. Unsure	4. Likely	5. Very likely

Why do you say that?

____

Appendix R

Participant's ID_______________________

Film assessment

Please complete the following questionnaire based on the person in the film. Please circle how much you agree or disagree with each of the following. Please circle one answer for each question.

1. I didn't develop good feelings towards Brad.

1. Strongly Disagree 2.Disagree 3. Neutral 4. Agree 5. Strongly Agree

2. I could relate to Brad.

1. Strongly Disagree 2.Disagree 3. Neutral 4. Agree 5. Strongly Agree

3. I felt moved by Brad.

1. Strongly Disagree 2.Disagree 3. Neutral 4. Agree 5. Strongly Agree

4. I felt empathy towards Brad.

1. Strongly Disagree 2.Disagree 3. Neutral 4. Agree 5. Strongly Agree

5. I could not understand the way Brad was feeling.

1. Strongly Disagree 2.Disagree 3. Neutral 4. Agree 5. Strongly Agree

Appendix S

INTERVENTION ASSESSMENT

Below are a number of statements regarding your opinion about the workshop. Please circle how much you agree or disagree with each of the following statements.

1. My knowledge about Tourette’s syndrome has improved as a result of the workshop.

1.	2.	3.	4.	5.
Strongly agree	Agree	Neutral	Disagree	Strongly disagree

2. My attitude towards individuals with Tourette’s syndrome has not improved as a result of the workshop.

1.	2.	3.	4.	5.
Strongly agree	Agree	Neutral	Disagree	Strongly disagree

3. I enjoyed participating in the workshop.

1.	2.	3.	4.	5.
Strongly agree	Agree	Neutral	Disagree	Strongly disagree

4. The film was the most interesting/useful part of the workshop.

1.	2.	3.	4.	5.
Strongly agree	Agree	Neutral	Disagree	Strongly disagree

5. The presentation with the slides was the most interesting/useful part of the workshop.

1.	2.	3.	4.	5.
Strongly disagree	Disagree	Neutral	Agree	Strongly agree

Is there anything else you would like to add?

____

____

____

____

____

Appendix T

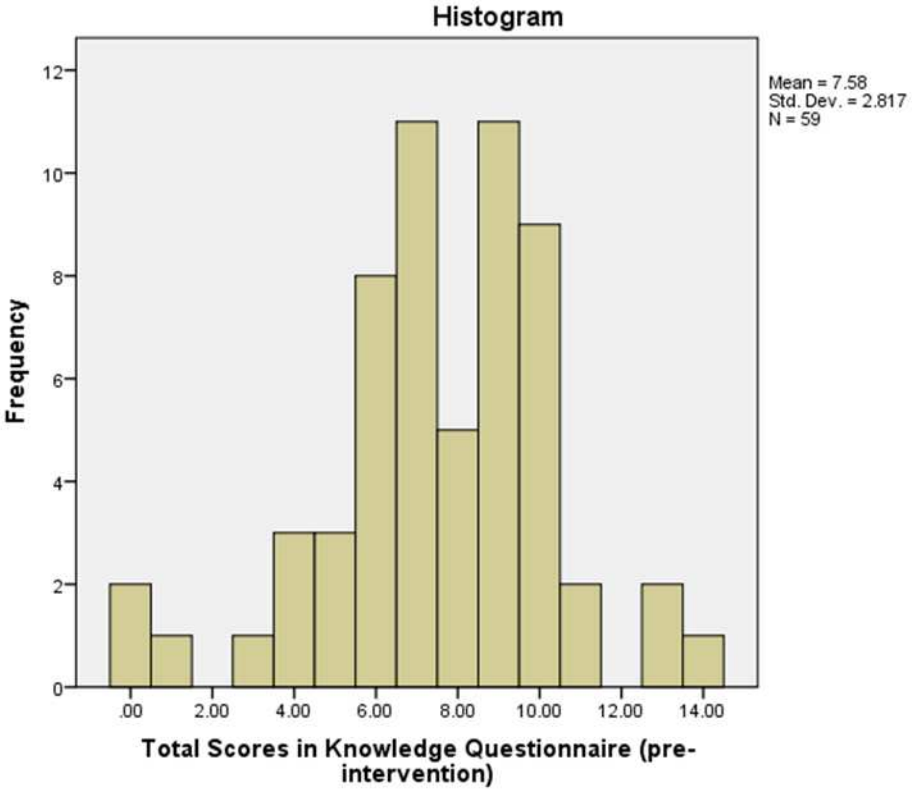


Figure 8 - Histogram displaying the participants' total score on the knowledge questionnaire (pre-intervention).

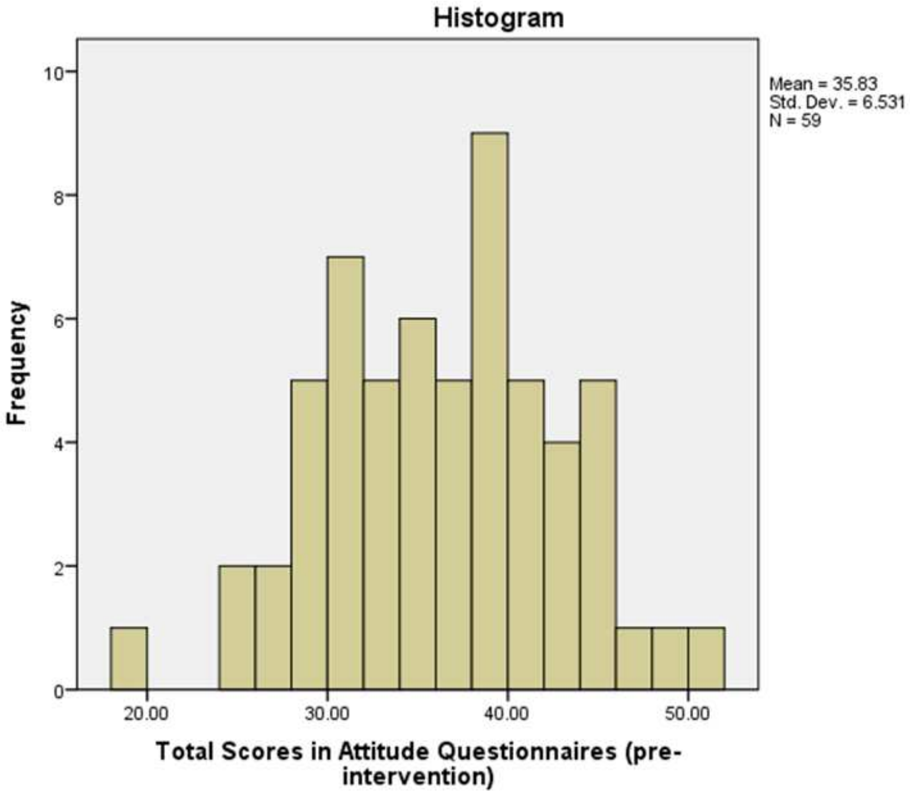


Figure 9 - Histogram displaying the participants' total scores on the attitude questionnaire (pre-intervention).

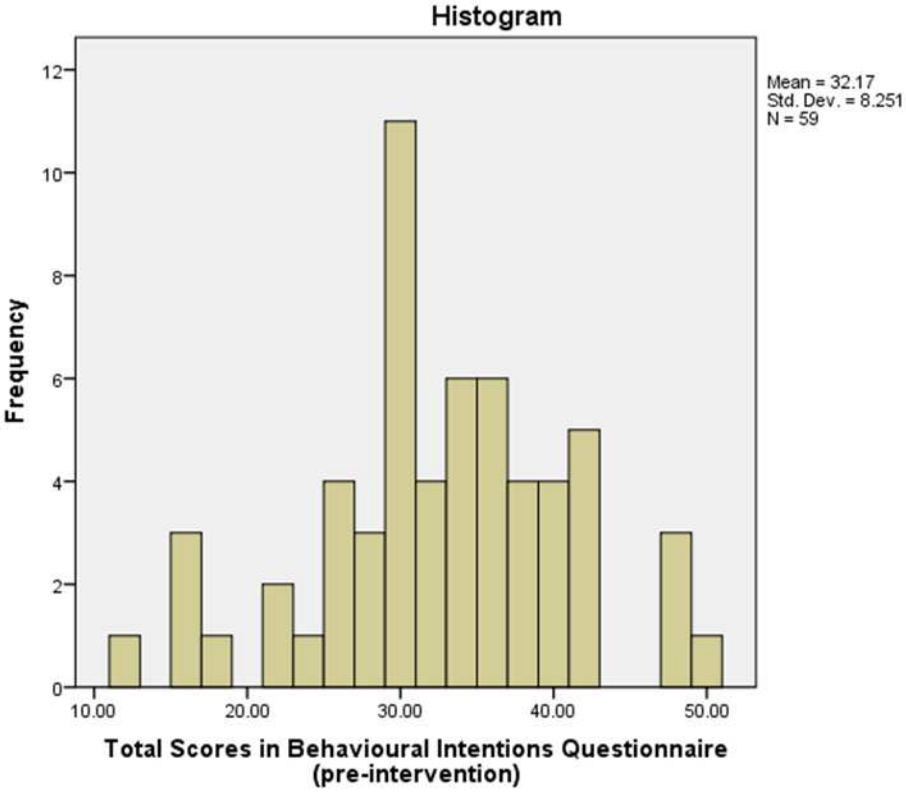


Figure 10 - Histogram displaying the participants' total scores on the behavioural intentions questionnaire (pre-intervention).

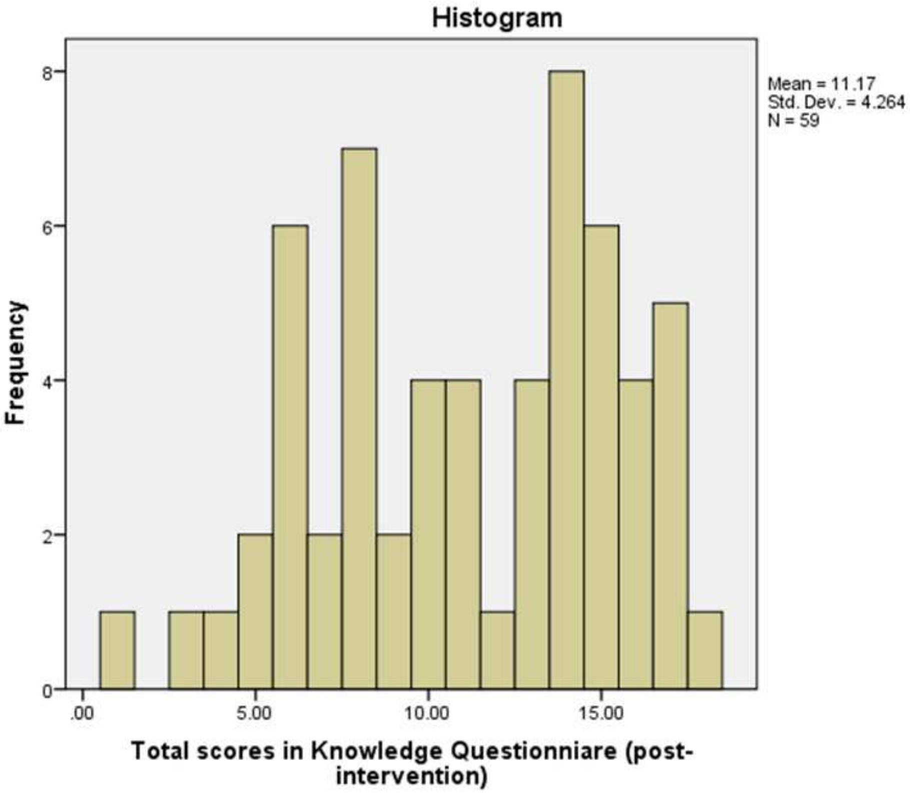


Figure 11 - Histogram displaying the participants' total score on the knowledge questionnaire (post-intervention).

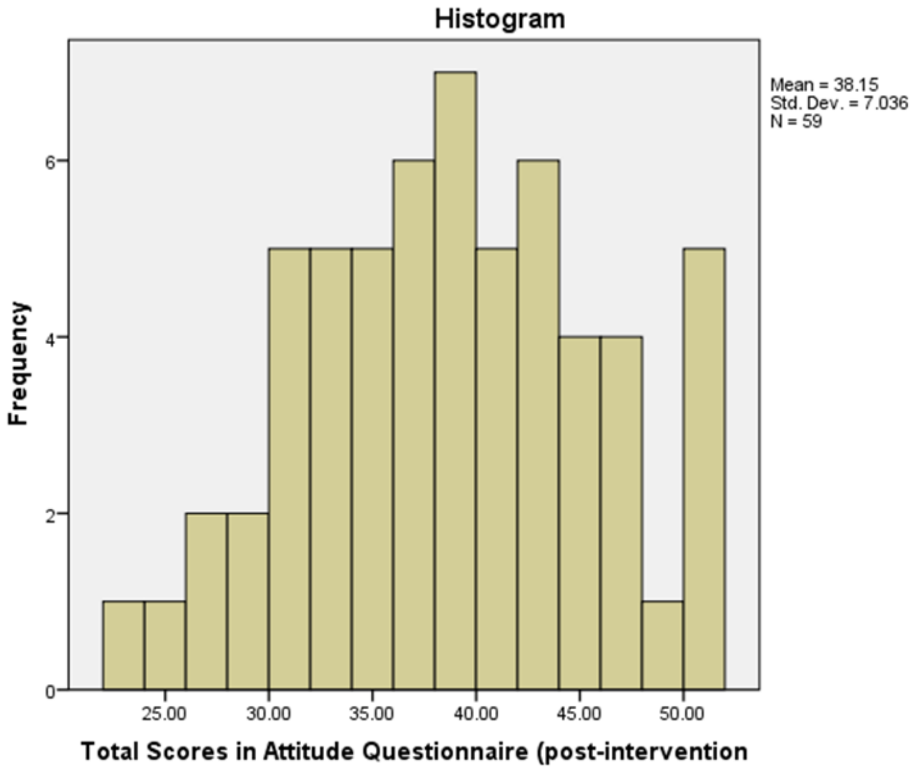


Figure 12 - Histogram displaying the participants' total scores on the attitudes questionnaire (post-intervention).

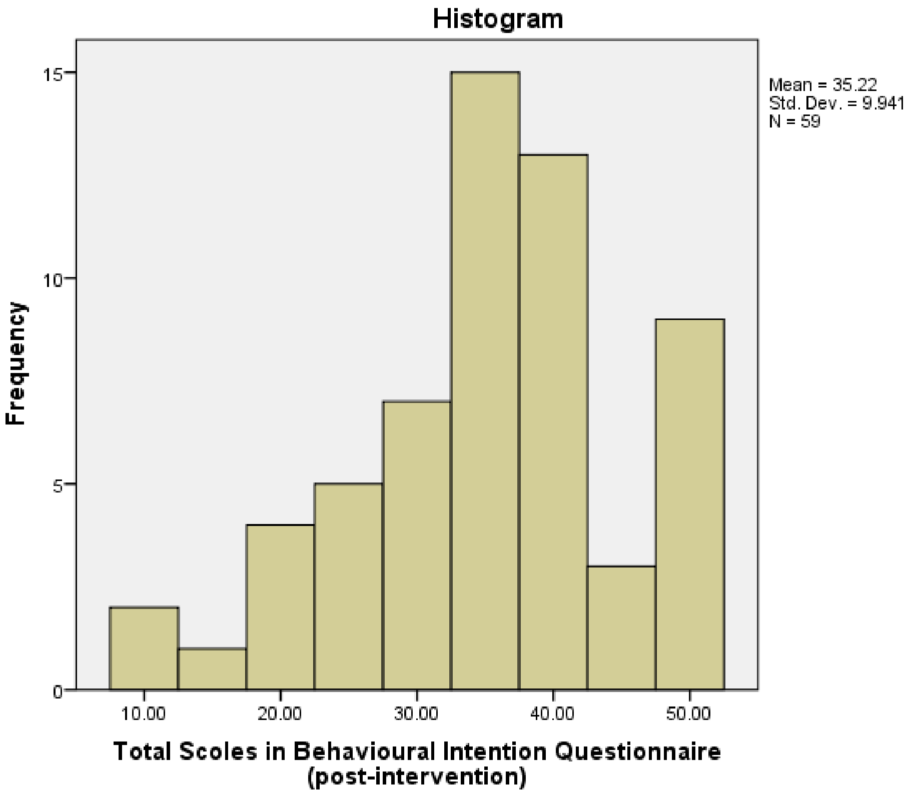


Figure 13 - Histogram displaying the participants' total scores on the behavioural intentions questionnaire (post-intervention).

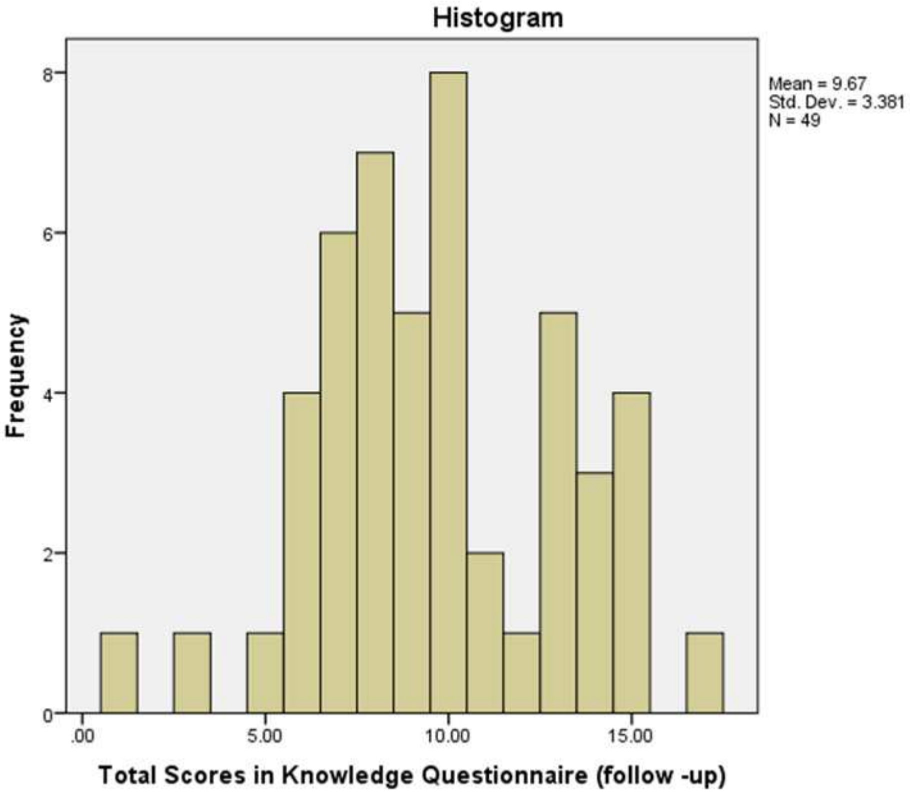


Figure 14 - Histogram displaying the participants' total scores on the knowledge questionnaire (follow-up).

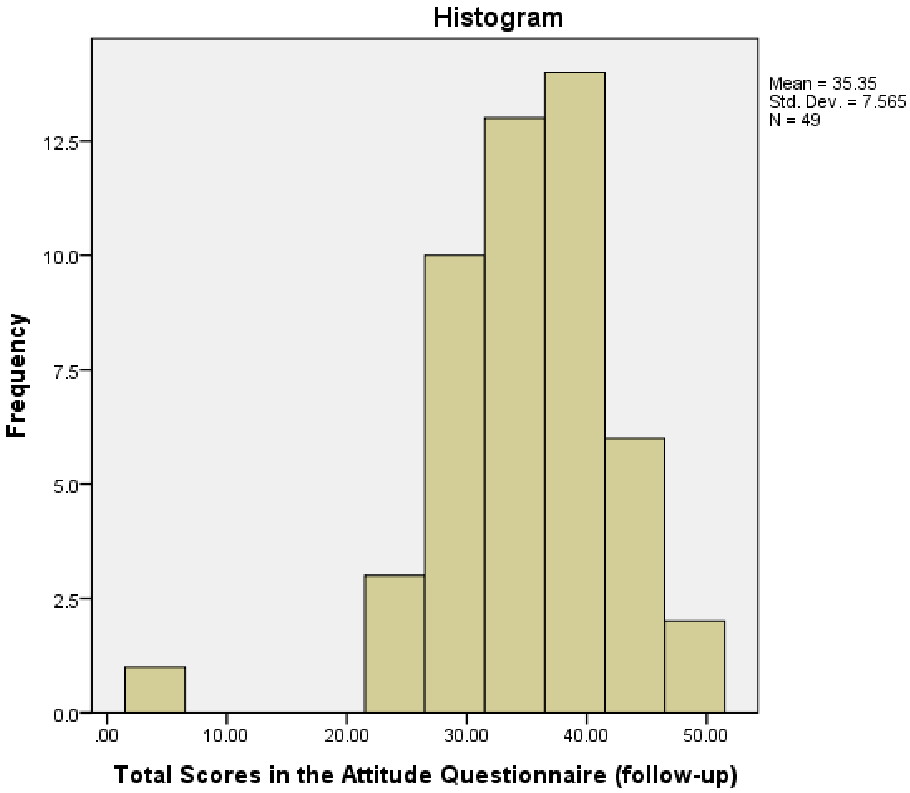


Figure 15 - Histogram displaying the participants' total scores on the attitude questionnaire (follow-up).

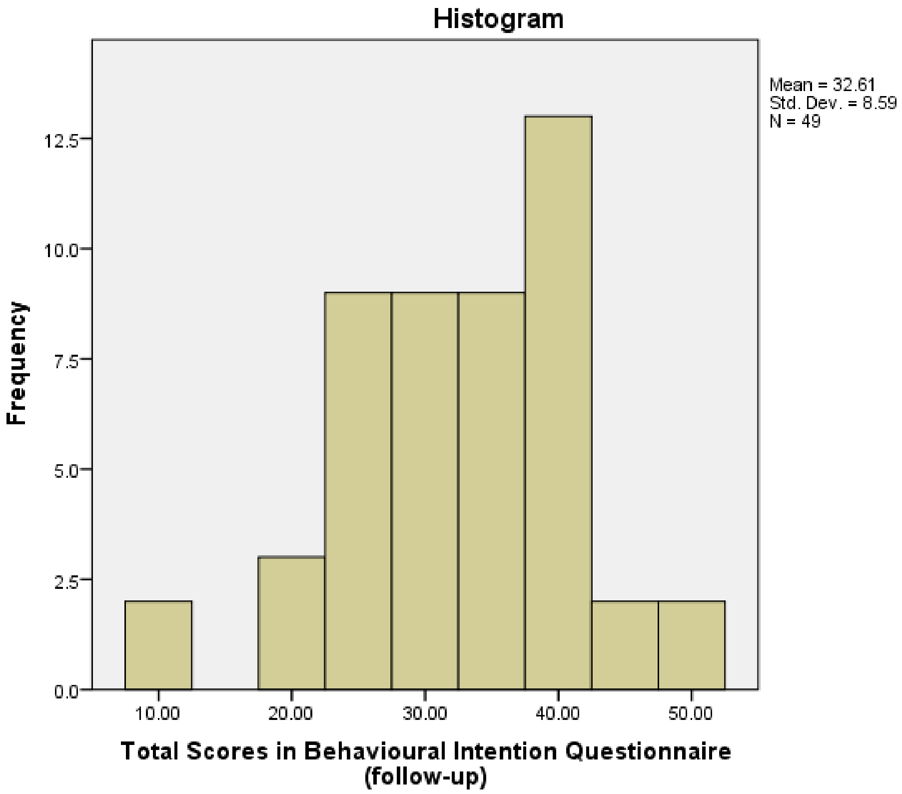


Figure 16 - Histogram displaying the participants' total scores on the behavioural intention questionnaire (follow-up).

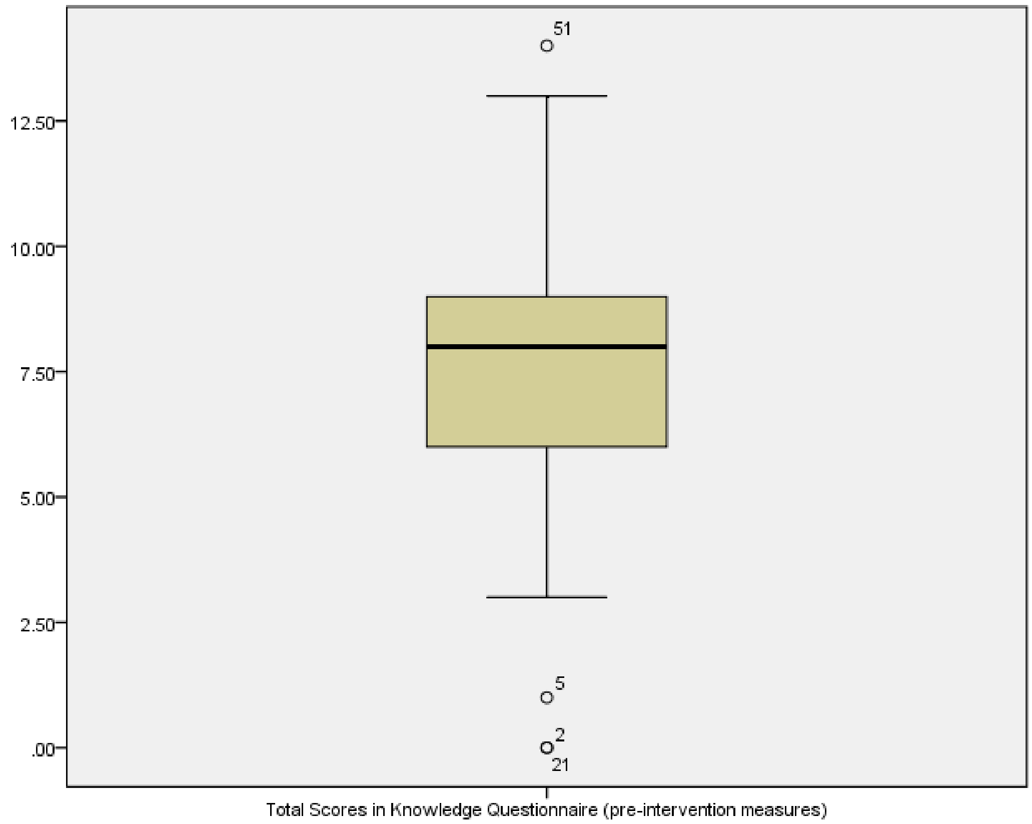


Figure 17 - Box plot for outliers on total scores on the knowledge questionnaire (pre-intervention).

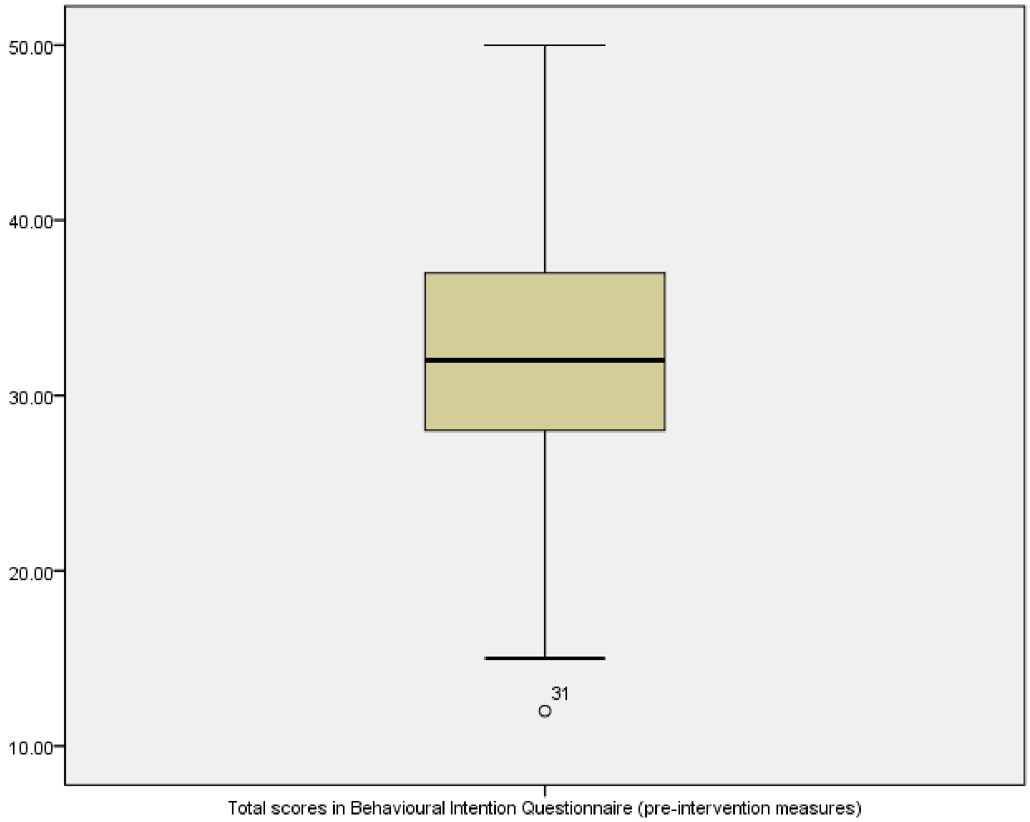
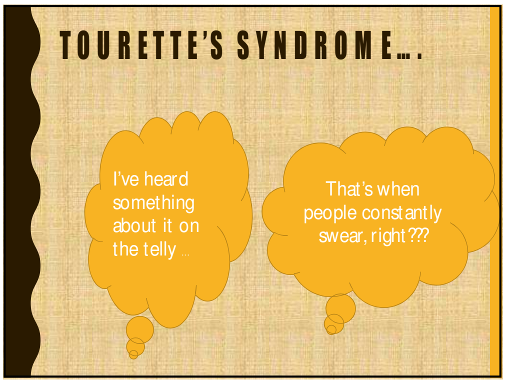


Figure 18 - Box plot of outliers on total scores on the behavioural intention questionnaire (pre-intervention).

Figure 19 - Box plot of outliers on total scores of the behavioural intention questionnaire (post-intervention measures).

Figure 20 - Box plot of outliers on total scores of the attitude questionnaire (post-intervention measures).

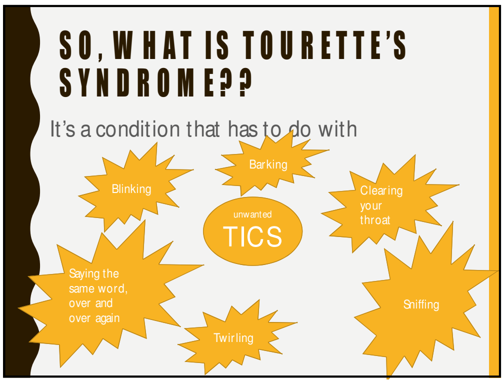
Appendix U



TOURETTE'S SYNDROME….

I've heard something about it on the telly …

That's when people constantly swear, right ???

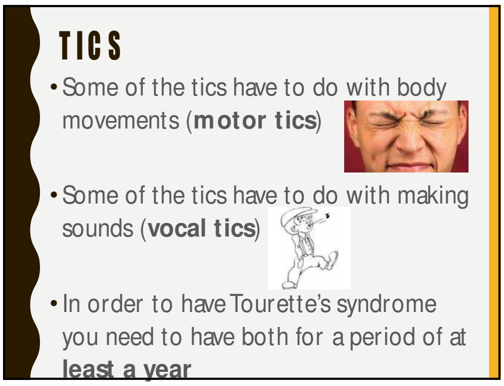


SO, WHAT IS TOURETTE'S SYNDROME??

It's a condition that has to do with

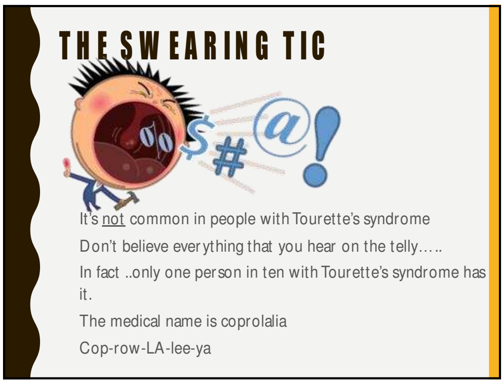
unwanted TICS

- Blinking
- Barking
- Clearing your throat
- Saying the same word, over and over again
- Twirling
- Sniffing



TICS

- Some of the tics have to do with body movements (**motor tics**)
- Some of the tics have to do with making sounds (**vocal tics**)
- In order to have Tourette's syndrome you need to have both for a period of at **least a year**



THE SWEARING TIC

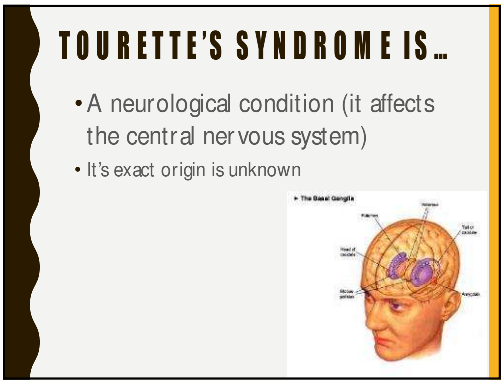
It's not common in people with Tourette's syndrome

Don't believe everything that you hear on the telly…..

In fact ..only one person in ten with Tourette's syndrome has it.

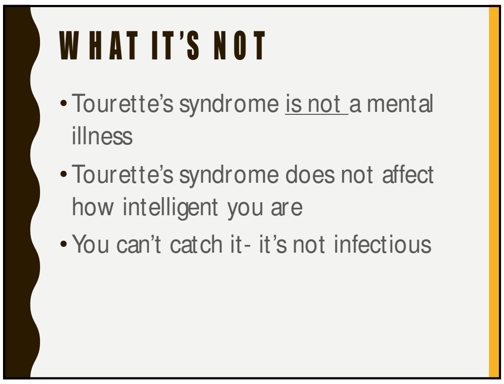
The medical name is coprolalia

Cop-row-LA-lee-ya



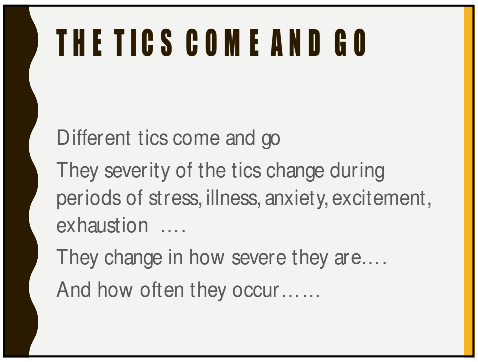
TOURETTE'S SYNDROME IS…

- A neurological condition (it affects the central nervous system)
- It's exact origin is unknown



WHAT IT'S NOT

- Tourette's syndrome is not a mental illness
- Tourette's syndrome does not affect how intelligent you are
- You can't catch it- it's not infectious



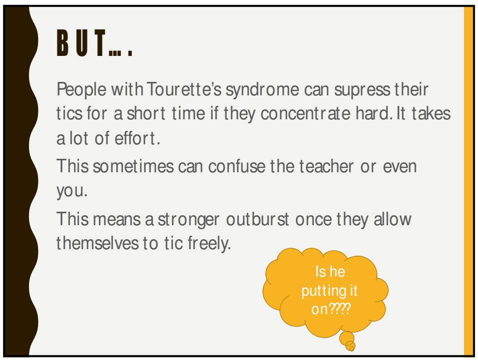
THE TICS COME AND GO

Different tics come and go

They severity of the tics change during periods of stress, illness, anxiety, excitement, exhaustion ….

They change in how severe they are….

And how often they occur……



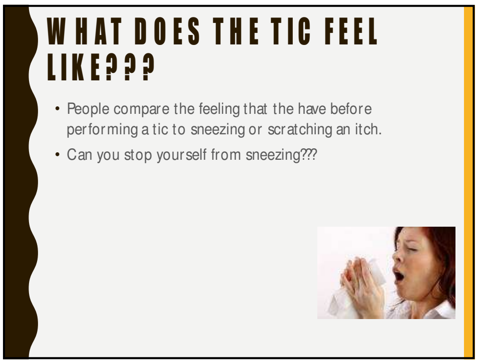
BUT….

People with Tourette's syndrome can supress their tics for a short time if they concentrate hard. It takes a lot of effort.

This sometimes can confuse the teacher or even you.

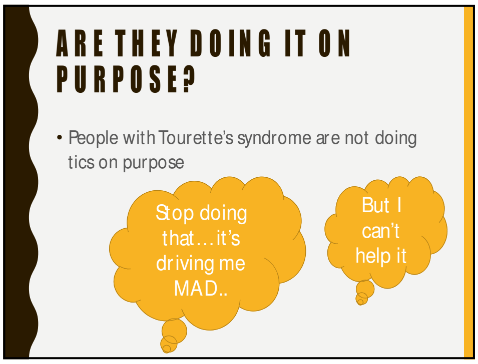
This means a stronger outburst once they allow themselves to tic freely.

Is he putting it on????



WHAT DOES THE TIC FEEL LIKE???

- People compare the feeling that the have before performing a tic to sneezing or scratching an itch.
- Can you stop yourself from sneezing???

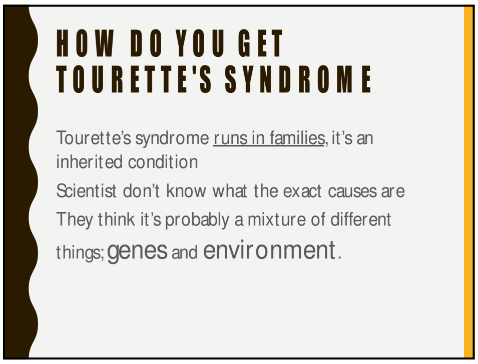


ARE THEY DOING IT ON PURPOSE?

- People with Tourette's syndrome are not doing tics on purpose

Stop doing that…it's driving me MAD..

But I can't help it

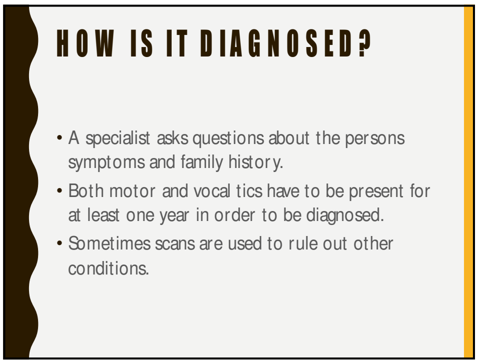


HOW DO YOU GET TOURETTE'S SYNDROME

Tourette's syndrome runs in families, it's an inherited condition

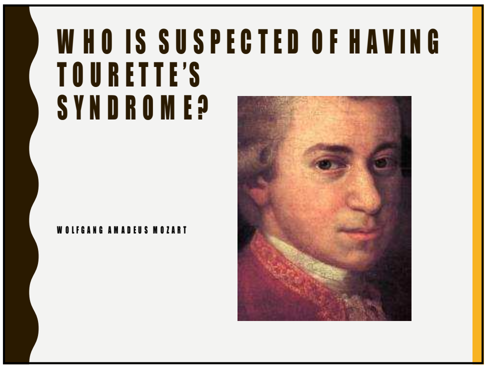
Scientist don't know what the exact causes are

They think it's probably a mixture of different things; genes and environment.



HOW IS IT DIAGNOSED?

- A specialist asks questions about the persons symptoms and family history.
- Both motor and vocal tics have to be present for at least one year in order to be diagnosed.
- Sometimes scans are used to rule out other conditions.

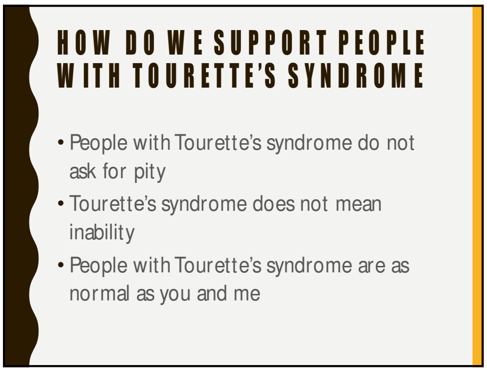


WHO IS SUSPECTED OF HAVING TOURETTE'S SYNDROME?

WOLFGANG AMADEUS MOZART

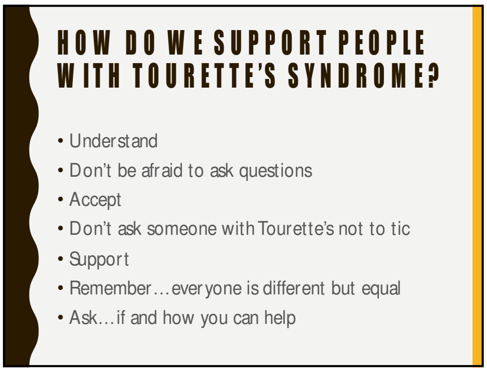
WHO IS SUSPECTED OF HAVING TOURETTE'S SYNDROME?

Amy Winehouse



HOW DO WE SUPPORT PEOPLE WITH TOURETTE'S SYNDROME

- People with Tourette's syndrome do not ask for pity
- Tourette's syndrome does not mean inability
- People with Tourette's syndrome are as normal as you and me



HOW DO WE SUPPORT PEOPLE WITH TOURETTE'S SYNDROME?

- Understand
- Don't be afraid to ask questions
- Accept
- Don't ask someone with Tourette's not to tic
- Support
- Remember…everyone is different but equal
- Ask…if and how you can help

THANK YOU FOR LISTENING

Appendix V

TIZARD
University of Kent

Tizard Ethics Feedback Form

Student Name:	Melina Malli
Supervisor:	Rachel Forrester Jones & Michelle McCarthy
Title:	"Adolescents' perception of their peers with Tourette's syndrome and their knowledge about the disorder: an exploratory study"

The Tizard Ethics Committee have considered the proposal and wish to make the following comments:

- This proposal is not subject to NHS approval so please amend front sheet to NO and question 3 has not been completed. The screening form needs to be revised so that it is only 6 pages long, references are not needed as part of this.
- Section 2 (b) refers to comprehensive schools in Kent. However Kent runs on a grammar/secondary system so the Researcher will need to consider different schools and amend this. Given the prevalence rates quoted it seems unlikely that any school would not have a child with Tourette's even if not formally diagnosed. Perhaps it would be better to require that no child has Tourette's in the relevant year group.
- Section 2 (d) This needs to be more explicit detailing how long these sessions will be
- Demographic information. It is felt that a free text response to gender should be allowed rather than stipulate male/female.
- Focus Group – the proposal doesn't note whether this will be recorded and if so mention should be made of this.

- Consent form for parents/Participants Consent – these need to have Supervisor contact detais and check for typos
- Information sheet for the Head Teacher. This needs to have Researcher and Supervisor contact details. The sheet notes that the Researchers are MM and RFJ this needs to be changed to show RFJ is the Supervisor.
- All these sheets need to confirm that Ethical approval has been given
- The photos are deemed not to be culturally diverse so this needs to be changed and to include some (clearly identifiable as) boys in the photos too.

Action: The Committee has approved the proposal with the above amendments to be made. These need to be discussed, agreed and signed off by the Supervisor.

Signed: J.Ruffels **Date: 19.01.15**

On behalf of Tizard Ethics Committee

Alterations approved by Supervisor	**Signature**	**Date**
Final approval On behalf of Tizard Ethics Committee	**Signature**	**Date 19.01.15**

15.02.15 The Committee confirm receipt of the amended proposal and confirm that this has been signed off by the Supervisor.

Appendix W

TIZARD
University of Kent

Tizard Ethics Feedback Form

Student Name:	Melina Malli
Supervisor:	Rachel Forrester Jones & Michelle McCarthy
Title:	"Adolescents' perception of their peers with Tourette's syndrome and their knowledge about the disorder: an exploratory study"

The Chair of the Ethics Committee confirms approval of the minor amendments submitted.
It is understood that the study will be conducted and carried out in [redacted] Academy, Margate which has involved minor changes to the consent form previously submitted and that the consent has been changed from opt in to opt out ie the parents will contact the school only if they disagree with their child taking part.

<table>
<tr><td colspan="3">Signed: J.Ruffels Date: 16.03.15
On behalf of Tizard Ethics Committee</td></tr>
<tr><td>Alterations approved by Supervisor</td><td>Signature</td><td>Date</td></tr>
<tr><td>Final approval On behalf of Tizard Ethics Committee</td><td>Peter McGill
Signature</td><td>Date 16.03.15</td></tr>
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