

Obsessive Compulsive Tic Disorder: appropriate diagnosis and treatment as key elements to improve health and rationalize use of resources

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ABSTRACT

Background: Obsessive Compulsive Tic Disorder (OCTD) has been described recently as an early onset and highly disabling endophenotype of Tic Disorder and Obsessive Compulsive Disorder. OCTD has a relevant but largely unknown clinical, social and economic burden for patients and their families. Our paper aimed to explore relevant aspects of the burden of OCTD, such as the etiological, clinical and epidemiological aspects of the condition, and implication of diagnosis and treatment on patients' health and socio-economic impact.

Methods: We conducted a literature review and a pilot study using retrospective demographic, clinical and economic data of patients with OCTD accessing the Galeazzi Hospital in Milan

Result: The literature review shows the absence of information on OCTD. From the pilot study we analysed data of 30 patients (80.0% male, 36.7% aged from 7 to 13 years, 63.3% aged from 15 to 48 years), 83.0% declared that obsessions and/or compulsions were the most important factors determining their social impairment. Adult patients refractory to drug treatment underwent Deep Brain Stimulation plus drugs. The mean clinical scores at the time of diagnosis indicated a severe condition for both tics and obsessive compulsive components. The mean time elapsed from symptoms onset to diagnosis of OCTD was 5.6 years, reaching up to 11 years in one case. Before reaching the correct diagnosis, different specialists visited the patients several times, 93.3% underwent diagnostic examinations and 86.7% took 2 or 3 different drugs. Ten patients were hospitalised and 8 had received psychotherapy.

Discussion: Albeit preliminary, these results show that attention is mandatory for establishing correct diagnosis and treatment guidelines to improve health and rationally spend resources for OCTD.

Key words: Obsessive Compulsive Disorder (OCD), Tic Disorder (TD), Obsessive Compulsive Tic Disorder (OCTD), Social impairment, Costs

INTRODUCTION

A multidisciplinary team of experts [1] has recently identified Obsessive Compulsive Tic Disorder (OCTD) as a specific chronic condition, which is identified as an endophenotype of Obsessive Compulsive Disorder (OCD) and Tic Disorder (TD), characterised by an early onset, relatively high prevalence, significant disability, presenting diagnostic and therapeutic difficulties. This could reflect negative impact on socioeconomic burden and Quality of Life of patients.

OCD is defined by the presence of obsessions (intrusive thoughts) and compulsions (repetitive behaviors) leading to adaptive malfunctioning and emotional maladjustment and, therefore, a high social impairment [2]. TDs constitute a group of common and complex neurodevelopmental disorders characterized by sudden, repetitive, not always fast and non rhythmic afebrile movement and/or sounds of vocalization [3]. Different types of TDs have been identified according to the duration and severity of tics, including transient tics, chronic tics and Tourette's syndrome (TS), with different phenomenology and epidemiology. More specifically, TS is a complex chronic tic disorder characterized by motor, vocal and phonetic tics [4]. In the scientific community and in clinical practice, Obsessive Compulsive Symptoms (OCS) and Behaviors (OCB) are frequently observed in patients with chronic TD, as happens for chronic tics in subjects with OCD [5,6]. Compresence of OCD, OCS or OCB with TD or tic symptoms is so frequent in specific subgroups of patients, that some experts in the field believe that OCD and TD are actually strictly interconnected and actually represent a specific subtype of disorder. In particular, some years ago, although not exhaustively yet experts of the London TS group [7] identified 5 phenotypes of TS: (1) minimally affected class, (2) chronic motor tics and OCD, (3) TS and OCD/OCB, (4) TS and OCD, and (5) TS and OCD and Attention Deficit Hyperactive Disorder (ADHD). Actually, no specific research has been conducted yet on OCTD, leaving to the clinician's expertise the responsibility of identifying, treating and following up across the years each patient, with the aim to reduce health impairment. Within the attempt of clarifying the most crucial issues of OCTD and related consequences, the objective of the present study was to explore relevant aspects of the burden of OCTD, such as the etiological, clinical and epidemiological aspects of the condition, and implication of diagnosis and treatment on patients' health and socio-economic impact.

METHODS

This study was conducted in two phases. A first phase consisted of a literature review aimed to understand what information is already available on the burden of OCTD in the scientific community. The second phase consisted

of a pilot study aimed to collect and analyse data from a sample of patients diagnosed with and treated for OCTD. The literature review was conducted through several databases and search engines, such as PubMed, Cochrane Library and Google Scholar, focusing on the epidemiologic, biological, clinical, economic, humanistic and other relevant issues related with OCTD, OCD and chronic TDs, including Tourette's Syndrome. Additionally, the lists of references included in the papers found in the engines specified above were checked to select other potentially relevant papers. To conduct the pilot study, we retrospectively retrieved from medical charts data on 30 patients diagnosed with OCTD and followed at the Tourette Centre of the Galeazzi Hospital in Milan (Italy): demographic and relevant clinical data, and information on the use of main medical resources for reasons attributable to their condition before and after the diagnosis of OCTD, such as the number and type of medical visits, diagnostic examinations, pharmacological treatments and other types of interventions. The medical charts were randomly selected among those of the patients followed at the Tourette Centre at the time of data collection. According with the purpose of this study, we decided to collect data from both patients treated pharmacologically and patients who underwent Deep Brain Stimulation (DBS), involving around half of the sample in either treatment option to be able to analyse the related data. Actually, only 4% of the patients at the Tourette Centre in Milano have been treated with DBS identified among adults chronically and severely impaired, and refractory to psychological and pharmacological treatments. Hence, we grouped the medical charts according with the treatment option pharmacological or DBS, and randomly selected those to be used in the study within each option.

These data were described using absolute and relative frequencies for the categorical variables, while continuous variables e.g. age and clinical measures were summarised by mean values along with minimum and maximum values as dispersion measures. We conducted a descriptive analysis using STATA SE 12 (Stata Corp, Texas, US) software. For the use and analyses of these data, ethical approval was not required since we used retrospective no patient identifiable data provided in the course of normal patient care.

RESULTS

Results of the Literature research

Etiology

TD/TS and OCD, together with other conditions like ADHD, are reported in the literature as characterised for being complex and often comorbid conditions. For instance, Robertson [8] has summarised a number of

studies and findings on co-occurrences that suggest a genetic linkage between these conditions. Grados and Mathews [9] also suggested a contribution of environmental factors in determining these conditions. Other researches have suggested that TD/TS and OCD, particularly early onset OCD, are familial and may represent alternative expressions of the same underlying etiological liability [7,10-14]. Family and genetic studies have recently shown TD/TS and OCD cluster in families, as being a possible reason of shared genetic liability [15].

From a recent study involving almost 22,000 adult twins from the general population [16], the authors found that tics, OCS and, less strongly, attention deficit/hyperactivity liabilities showed modest, but significant covariation. This finding is in line with previous family studies, which have shown a stronger familiarity between OCD and TD, than between TD and ADHD [9]. Finally, specific genetic studies support a substantial shared genetic etiology of these conditions [17-19].

Epidemiological and clinical impact

Information on prevalence and incidence of OCTD in the population is not available yet; however, clinical experience and the results of research on OCD and TD/TS suggest that this endophenotype is common, since it may be hypothesised that the lifetime prevalence of OCTD might reach 1% of the general population [1].

As regards OCD, this disorder can involve 1–2% of the general population [20], with pre adult onset occurring in more than half of the cases [21].

The prevalence of TS involves up to 20% of children and decrease to about 0.77–1% in the adults [22-24], while transient tic disorder has been found to reach a prevalence of 2.99%. Regarding OCD and TD/TS contemporarily present, results of a study published in 1992 show that the prevalence of OCD ranged from 11 to 80% of patients with TS [25]. OCS have been found in up to 90% of patients with TS [5, 6]. More recent research has identified tics in 53% of paediatric patients with OCD [26].

From a clinical point of view, an examination of patients presenting both TD and OCD reveals that compared to the single conditions, early onset, male sex, smoking, sensory phenomena, obsessions, aggressiveness, hoarding, exactness and sounds, impulsive behaviors and ADHD comorbidity, seem to be frequent and also determine a higher disability and a higher propensity of being refractory [5, 6, 27-30].

Health Related Quality of Life and Social Impairment

Clinical experience reveals that OCTD may be particularly challenging in terms of impaired Health Related

Quality of Life (HRQoL), since the association of OCD with TD results in a higher disability compared to the burden caused by each condition per se [5, 6, 27-29, 31]. Recent literature raises that the OC component is likely the most debilitating and disturbing aspect of the disease, foremost from a social impairment standpoint [32, 33], followed by sound tics and motor tics [34]. OCD has been ranked by the World Health Organization in the top 10 the most disabling illnesses by lost income and impaired [35]. A number of patients present serious problems with everyday life, above all in their family, social, studying and working life. Stigma, social isolation, exclusion, bullying, self injurious behaviors and suicide attempt, have been largely reported in OCD and TD patients, largely caused or rosined by misperceptions of the disorders by teachers, peers, and the wider community. It must be added that the burden of the condition, including the HRQoL impairment, goes beyond the individual patient and deeply affect families and caregivers [5, 6, 23, 27-29, 36]. Among the consequence on patients' health, Modafferi and colleagues [24] have underlined that sleep disorders in pediatric patients with TD are more frequent than in typically developing children, and the concomitant presence of anxiety, depression and OC symptoms. Among patients with OCD, some symptoms can have the strongest negative effect on the social domain: for instance, the hoarding behavior, consisting on an excessive acquisition and inability or unwillingness to discard large quantities of objects that cover the living areas of the home, sexual symptoms, consisting on living with intrusive sexual thoughts (e.g. fears to commit pedophilic acts without realising, fears of being depraved, fears of being punished for his own sins) and/or religious symptoms (e.g. blasphemous thoughts or excessive religious scrupulosity). Furthermore, an earlier age of OCD onset can account for academic and professional underachievement [5, 6, 23, 27-29, 36]. Patients with OCD often experience many limitations in the fulfillment of their fundamental human needs, such as: disturbed patients' functioning in the common life, in the ability to realise their goals, skills, potential, capacity to follow prescribed treatments, take medications, cooperate in addressing the root causes of their problems, reduce obsessive thoughts and compulsive behavior, as well as their willingness to realise exposures [37].

Economic aspects

If limited evidence is available about the socioeconomic burden and the efficiency of the different treatment options for OCD and TD/TS, no information is available OCTD. Table 1 summarises the literature on this topic, and in particular shows the high heterogeneity and non satisfactory informative value of the research conducted on OCD and TD/TS. However, looking at the available information on the single conditions, at the present it is

possible to hypothesise the high socioeconomic impact attributable to OCTD.

Among the studies conducted, we report here some details from the more informative of them on TS and OCD. As regards the burden of OCD, DuPont and colleagues [38] estimated that the total annual costs attributable to OCD corresponded to 5.7% of the estimated cost of all mental illness, which amounts to 147.8 billion \$, and 18.0% of the costs of all anxiety disorders, which amounts to 46.6 billion \$, annually. Furthermore, the highest contribute to the total costs was attributed to indirect costs (6.2 billion \$) for the lost productivity by individuals suffering from or dying from OCD. More recently, Hollander and colleagues [36] published a review on the costs of compulsivity, focusing on the several aspects that influence the high impact attributable to having OCD. In particular, they noticed the increasing annual costs attributable to OCD before (8.4 billion \$) and after the year 2008 (10.6 billion \$), which suggests a further increase that can also be expected in the future years.

As regards TS, the cost of illness was calculated from the point of view of healthcare and transfer payment providers including both direct and indirect costs [39]. Mean direct costs corresponded to 620 € per patient every 3 months, mainly attributable to drugs, (36%), hospitalisations (31%), and rehabilitations (16%). Indirect costs corresponded to 2,511 € per patient every 3 months for productivity loss due to incapability to work or early retirement, and 220 € per patient every 3 months for absenteeism.

Specific investigation is needed for the estimation of socioeconomic burden attributable to OCTD, and to conduct appropriate economic evaluations aimed to compare the cost effectiveness of the available treatments.

Diagnosis

Experience in clinical practice suggests that an anamnestic diagnosis of the disease, based on the identification of symptoms and occurrences during the previous life years, and on the identification of symptoms and behaviors among their relatives, is necessary to appropriately identify patients affected by OCTD. However, this approach is generally not adopted nowadays. Therefore, in many patients the condition is for years not correctly diagnosed. A wrong diagnosis leads to additional severity, reduced wellbeing, frustration, and compromised social and working relationship. In addition, patients continue to require visits to different medical specialists, adopting a doctor shopping attitude (i.e. patients change several different medical specialists to find a satisfactory treatment), several diagnostic examinations and diverse attempts of treatments, with the consequence of consuming high and unjustified resources.

Several instruments and scales [40] have been

realised and are used to diagnose patients with OCD and those with TS. Among the available scales, there are the Yale Global Tic Severity Scale (YGTSS), focusing on the severity of motor and sound tics [41, 42], and the Yale Brown Obsessive Compulsive Scale (YBOCS) [43], identifying the severity of OCD. A drawback of the YBOCS is the insufficient level of specificity, sensitivity and predictive value. Other instruments are the premonitory urge phenomenon in youths (PUTS) with TD [44], and the videotaping and the related scale (Modified Rush Videotape Rating Scale), used as a measure of treatments outcomes for tics [42]. However, the videotaping is a limited instrument since, at the time of the visit, the patients can suppress tics. Therefore, for the diagnosis of tics, obsessions and compulsions it is very important what do patients and their caregivers report on their everyday life at home [32]. Furthermore, none of these instruments is useful enough to identify the subjects who present the combined, and more complex, OCTD phenotype. All of the reasons presented above demonstrate why professional expertise is necessary for an accurate diagnosis.

Treatment

Patients diagnosed with OCTD are among the most severe and difficult to manage, actually they are more complex than the ones with OCD and TD/TS alone [1].

Overall, the treatment strategy for OCD and for TS/TD is supposed to need a symptom specific individualised approach, with the aim to alleviate suffering and social impairment according to the specific characteristics of each patient [1, 8, 19, 45-47]. In many cases, a multidisciplinary team of professionals with different expertise, e.g. psychiatrist/child psychiatrist, neurologist, and psychologist, can be necessary for the overall management of patients' symptoms and needs. The involvement of family members and caregivers is often needed within the therapeutic program.

Subsequent integrated approaches have been reported and recommended for patients' treatment. Different approaches and recommendations exist in the literature, however, they agree on some main steps [8, 47-51]: a psychological treatment or a prevention therapy (e.g., habit reversal) can be used initially, followed by a pharmacological treatment based generally on the use of one or combination of SRI (Serotonin Reuptake Inhibitors), SSRI (Selective Serotonin Reuptake Inhibitors), typical and atypical neuroleptics. Botulin toxin infiltration can be used in patients with TD/TS who do not respond to other drugs. More invasive procedures such as DBS can be used in severe refractory patients as a single or an add on therapy as well [52]. In particular, DBS is showing promising results in both OCD [53, 54] and in TS [1, 55]. Efficacy of DBS is being shown for OCD, with more than 50% responders in severely ill and treatment

TABLE 1. Studies on costs/burden of disease and economic evaluations

Reference	Condition(s)	Subjects and criteria	Type of study	Technique of analysis	Types of treatment	Types of costs and effectiveness (when applicable)	Indirect costs	Year of reference for costs analysis	Country
Bamelis et al. 2012 [56]	Personality disorders (including OCD)	Adults	Original research (ongoing)	Cost-effectiveness	ST vs COP vs TAU	Direct, indirect cost and QALYs	Productivity loss	2007	The Netherlands
Dodel et al. 2010 [39]	Tourette's syndrome	Adults	Original research	Analysis of costs/burden of disease	Pharmacological and psychological interventions	Direct and indirect cost	Productivity and income losses	2006-2007	Germany
DuPont et al. 1995 [38]	OCD	Adults	Original research	Analysis of costs/burden of disease	Pharmacological and psychological interventions	Direct and indirect cost	Productivity loss	1990	USA
Eaton et al. 2008 [57]	Mental disorders (including OCD)	Adults	Literature review	Analysis of costs/burden of disease	Pharmacological and psychological interventions	Direct and indirect costs	Productivity loss	Cost estimates drawn from the study by DuPont et al. 1995 [39]	USA
Gellatly et al. 2014 [58]	OCD	Adults	Original research (ongoing)	Cost-effectiveness	Self-managed CBT interventions (cCBT and GSH) vs CBT waiting list (3 and 6-month follow-up) Self-managed therapies (cCBT and GSH) + conventional CBT vs waiting list + conventional CBT (12-month follow-up)	Direct, indirect costs and QALYs	Productivity loss	Not reported	UK
Grover et al. 2011 [59]	OCD	Adults	Original research	Analysis of costs/burden of disease	Not specified	Intangible costs quality of life of caregivers	Depression, anxiety, embarrassment, disturbing behaviors and frustration of family members of patients with OCD	Not applicable since not monetary values were used	India
Kalenthaler et al. 2006 [60]	Depression and anxiety disorders (including OCD)	Adults	Literature review	Cost analysis and cost-effectiveness	CCBT vs TAU	Direct, indirect costs and QALYs	Productivity loss	Not reported	UK
Hankin et al. 2011 [61]	OCD and depression	Adults	Original research	Analysis of costs/burden of disease	Psychotropic medications	Direct costs	Not analyzed	Not reported	USA
Hollander et al. 2016 [36]	Compulsivity (including OCD)	Adults	Literature review	Analysis of costs/burden of disease	Pharmacological and psychological interventions	Direct and indirect costs	Productivity loss	Cost estimates drawn from the study by DuPont et al. 1995	USA, Europe
Lenhard et al. 2017 [62]	OCD	Adolescents	Original research	Cost-effectiveness	Therapist-guided ICBT intervention vs a wait list condition of equal duration	Direct, indirect costs and QALYs	Productivity loss and absenteeism	2016	Sweden
Mancebo et al. 2008 [63]	OCD	Adults (with primary DSM-IV diagnosis of OCD)	Original research	Analysis of costs/burden of disease	Psychotropic medications and cognitive-behavioral therapy	Occupational disability and related HRQoL impairment	Not analyzed	Not applicable since not monetary values were used	USA
Mc Crone et al. 2007 [64]	OCD	NR	Original research	Cost-effectiveness	BT Steps vs clinician-guided therapy	Direct costs and unit improvement in YBOCS	Not analyzed	Not reported	USA
Ooms et al. 2017 [65]	OCD	Adults	Original research	Cost-effectiveness	DBS vs TAU	Direct, indirect costs and QALYs	Productivity loss and travel costs.	2015	The Netherlands
Singleton et al. 2001 [65]	Psychiatric morbidity (including OCD)	Adults	Original research	Analysis of costs/burden of disease	Psychotropic medications and counselling	Treatment and service use by patients with psychiatric disorders	Not analyzed	Not applicable since not monetary values were used	UK
Skapinakis et al. 2016 [67]	OCD	Children, adolescents and adults	Original research	Cost-effectiveness	Pharmacotherapies vs psychological interventions and combinations of both	Direct costs and QALYs	Not analyzed	Not reported	UK
Tolin et al. 2011 [68]	OCD	Adults	Original research	Cost-effectiveness	Stepped care ERP vs standard ERP	Direct, indirect costs, YBOCS scores and patient satisfaction	Cost of travel and lost wages from time spent in treatment, Provider indirect and labor cost (for patients and providers)	2008-2010	USA
Torres et al. 2012 [69]	OCD	Adults	Original research	Analysis of costs/burden of disease	Psychiatric or psychological treatment	Burden in family caregivers	Not analyzed	Not applicable since not monetary values were used	Brazil

ST: Schema Therapy; COP: Clarification Oriented Psychotherapy; TAU: Treatment As Usual; CBT: Cognitive behavioural therapy; cCBT: computerised CBT; GSH: Low-Intensity Guided self-Help; CCBT: Computerized Cognitive Behaviour Therapy; ERP: Exposure and Response Prevention; QALYs: Quality Adjusted Life Years

refractory patients. The effects of DBS for OCD seem to go beyond its classically defined symptoms. DBS targeted at the ventral striatum improves HRQoL independently of OCS, and is also able to improve mood, anxiety, and addiction [52]. Many target areas have been identified for DBS for TS, probably due to its large phenotypic variability [34]. Thus, in order to successfully treat each patient, the target areas of DBS for TS have to be chosen on the basis of phenotypes. For example, patients with mainly tic symptomatology and little comorbidity might benefit from DBS of the thalamus, whereas patients with TS and symptoms of OCD may best be treated with ventral anterior limb of the internal capsule DBS.

It is important to clarify that DBS is nowadays being considered as a treatment option in severe refractory cases; however, the definition of treatment refractory is still under discussion for its complexity. At the Tourette Centre of the Galeazzi Hospital in Milan a patient was considered refractory because he fails to respond to 2 years of psychological therapy and shows inadequate clinical response with at least 2 drugs of these categories: traditional and/or innovative antipsychotics, catecholamine depletors, and SSRIs [32].

However, no specific research has been conducted on the treatment of OCTD, leaving to the personal expertise of the clinician the responsibility of correctly treating and following up the patient across the years. Since no guidelines or agreements on treatment of OCTD exist yet, treatments for OCTD are often administered off label, especially in young subjects with additional need of caution for treatments.

Results of the pilot study

We analysed the data of 30 patients, 43.3% who after the diagnosis of OCTD underwent DBS (all adults), and the other 56.7% treated with pharmacological treatment. Table 2 reports a description of the study sample: 80.0% of the patients were male and their mean age was 22.3 years at the time of data analysis: specifically 36.7% of them were aged from 7 to 13 years, and 63.3% were aged from 15 to 48 years, similar to their mean age at the first access to the Tourette Centre 0.9 years before.

Most of the patients (83.3%) specified that obsessions and/or compulsions constitute their main cause of social impairment, while only 16.7% attributed this to their tics. The patients accessed for the first time the Tourette Centre on average 0.9 years earlier, when they were aged 21.4 years and did not receive yet the diagnosis of OCTD. At the time of first visit at the Centre, the patients' had a mean YGTSS score of 65.8, and a mean YBOCS score of 23.3, indicating both a mean severe level of disease. The patients declared that the time passed from the symptoms onset until the diagnosis of OCTD was on average 5.6

years, with one patient declaring even 11 years to obtain the correct diagnosis.

Table 3 shows the main medical resources adsorbed by the patients before OCTD diagnosis. On average, they were visited about 4 times by 5 several different specialists, showing the typical doctor shopping attitude. Neurologists, psychologists, neuropsychiatrists and psychiatrists were the most frequently visited specialists. Most of the patients (93.3%) reported one or more diagnostic examinations, including blood tests, magnetic resonances, electroencephalograms and psychological tests among the most frequent.

As regards treatments, before the diagnosis of OCTD, 86.7% of the patients had taken one or more drugs belonging to different categories: 40.0% neuroleptics, 36.7% antidepressants, 26.7% tetrabenazine and/or tripride, and/or topiramate, 6.7% anxiolytics and 30.0% antibiotics (amoxicillin or penicillin), homeopathic compounds, vitamins and/or cortisone. In addition, 10 patients had been hospitalised for their severe condition, e.g. for their self injurious behaviors, and 8 subjects had followed a psychological therapy.

DISCUSSION

Although OCTD is recognised by some experts as a complex, relatively common and burdensome condition [1], literature shows that no work has been conducted yet to measure the clinical, epidemiological and socioeconomic burden of this condition, nor to elaborate the strategies that should be adopted to correctly and promptly diagnose and treat patients.

Hence, we conducted for the first time a pilot study investigating the burden attributable to OCTD, including consequences related to delayed diagnosis and inappropriate treatment. Interesting results, albeit preliminary and incomplete, arise from the analyses of retrospective data of 30 pediatric and adult patients diagnosed with OCTD. We found that before accessing Tourette Centre, they spent many years and many resources, both human and financial, in searching unsuccessfully a correct diagnosis and treatment. Unfortunately, because the retrospective data available for this study did not include information on patients' HRQoL and/or treatment satisfaction, we were not able to measure the current perceived health status and level of satisfaction, neither to compare their status before and after the diagnosis of OCTD. However, according to what patients reported during the visits, the neurologist and the psychologist who follow them have observed an improved health from a clinical point of view, i.e. in terms of reduction of tics, obsessions and/or compulsions, and consequently in terms of wellbeing in their everyday social life, since some patients declared to return or start a near normal life. Furthermore, we could not measure the type and amount of

TABLE 2. Description of patients

Variable description	N (%) or mean (min-max),
Male	24.0 (80.0%)
Current age	22.3 (7-48)
Pediatric patients (aged 7-13 years)	11 (36.7%)
Adult patients (aged 15-48 years)	19 (63.3%)
Current treatment approach:	
DBS	13 (43.3%)
Pharmacological treatment	17 (56.7%)
Social impairment mainly attributable to:	
Tics	5 (16.7%)
Obsessions and/or compulsions	25 (83.3%)
Age at first visit at Galeazzi hospital	21.4 (7-48)
Pediatric patients (aged 7-13 years)	12 (40.0%)
Adult patients (aged 15-48 years)	18 (60.0%)
Duration (years) of attendance Galeazzi hospital	0.9 (0-3)
Time passed from symptoms to diagnosis	5.6 (1-11)
YGSS at first visit at Galeazzi hospital	65.8 (50.0-94.0)
YBOCS at first visit at Galeazzi hospital	23.3 (12.0-40.0)

productivity loss by the patients and their family caregivers, who are generally the parents of both pediatric and adult patients, strictly involved in the management of their condition. However, from clinical experience with these patients it is understood that often they work inefficiently and with a lot of discomfort, and sometimes even miss the opportunity of having a normal job. Their caregivers too can have working problems due to the need of taking care of their diseased children. Hence, we consider the results of this pilot study as underestimated, however, they suggest that it is mandatory to establish correct diagnostic and treatment to improve patients' health and rationally spend the available resources.

Within the area of Health Technology Assessment, after identifying the burden of OCTD, ad hoc studies are necessary to estimate the benefits and economic value of the different treatment strategies to manage each patient according to his characteristics and responsiveness to treatments. Actually, together with the absence of research in this field, despite recent attention toward an improved classification approach, the most recent edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM 5) (American Psychiatric Association's (APA)) still does not recognise OCTD as a disorder. In particular, OCD has been allocated into a newly defined chapter of obsessive compulsive and related disorders, with the introduction of a new 'tic related' specifier. In this perspective, OCTD may represent the evolution and further elaboration of the DSM 5 OCD tic related specifier.

Patients with OCTD can have severe social impairment, with consequences on their working and social life. Nowadays families and associations are not sufficiently organised to take care of these patients. Clinical experts can contribute to timely diagnose and

TABLE 3. Types of main medical resources before the diagnosis

Variable description	N (%) or mean (min, max)
Medical visits	
N. patients	30
Types of specialists per patient	5.0 (1-10)
Specialists more frequently reported:	
Neurologist	22 (73.3)
Psychologist	22 (73.3)
Neuropsychiatrist	21 (70.0)
Psychiatrist	15 (50.0)
Pediatrician	6 (20.0)
Ophthalmologist	4 (13.3)
Neuropath	3 (10.0)
Dietitian	3 (10.0)
Dermatologist	3 (10.0)
Homeopath	1 (3.3)
Naturopath	1 (3.3)
Rheumatologist	1 (3.3)
Psychomotricist	1 (3.3)
Diagnostic examinations	
N. of patients reporting at least one examination	28 (93.3)
N. of patients reporting specific examinations:	
Blood tests	22 (73.3)
Resonance	17 (56.7)
EEG	15 (50.0)
Psychological tests	14 (46.7)
Throat swab	10 (33.3)
TAC	9 (30.0)
EKG	5 (16.7)
Allergy tests	3 (10.0)
Eye examination	3 (10.0)
Dermatological tests	2 (6.7)
Sessions of psychomotricity	1 (3.3)
Drug Treatment *	
N. of patients with at least one drug treatment, of which:	26 (86.7)
Neuroleptic antipsychotic	12 (40.0)
Antidepressants (Tricyclic/SSRI)	11 (36.7)
Tetrabenazine and/or Tripride, and/or	8 (26.7)
Topiramato	2 (6.7)
Anxiolytics	9 (30.0)
Others	
Other interventions of which:	
Hospitalizations	10 (33.3%)
Psychological therapy	8 (26.7)

*Anxiolytics: Benzodiazepines; Antidepressants (Tricyclic/SSRI): Clomipramine, Fluvoxamina, Paroxetine, Citalopram; Neuroleptic antipsychotics: Sulpiride, Risperidone, Haloperidol, Paliperidone, Primozide, Aripiprazolo; Other: Antibiotics, Homeopathic, Vitamins, Cortisone.

treat patients with OCTD, and to disseminate knowledge on the phenomenon in the scientific community and among individuals who are directly or indirectly involved, i.e. patients themselves, their families, and budget holders representing the healthcare system as a whole. However, only few clinicians have the necessary expertise to face this condition, which is consequence and cause of high but restricted expertise on this condition. For instance, 280 new patients per year from different Italian regions and European Countries routinely access Tourette Centre in Milan to receive the correct diagnosis and treatment, totaling about 1,500 patients with OCTD in care at that Centre. Among these patients, 61 (4% of all the patients) chronic adults, who were severely impaired and refractory

to psychological and pharmacological treatments, have been treated with DBS.

In conclusion, albeit underestimated and limited by the lack of specific assessments, the results of the present study constitute a first attempt to better characterise patients with OCTD from an epidemiologic, clinical and therapeutic perspective, with specific attention in terms of disability and social impairment. Taken as whole, reported results on the burden of OCTD suggest that it is mandatory to establish correct diagnostic and treatment pathways to improve patients' health and rationally spend the available resources.

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