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AN INTERDISCIPLINARY EXPERIENCE WITH PEDIATRIC MUNCHAUSEN SYNDROME

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Abstract: Munchausen syndrome is a psychiatric condition likely encountered by all kinds of health professionals. In Munchausen syndrome patients injure themselves or fake their medical presentation features in order to assume the patient's role driven by the need for attention. Munchausen syndrome by proxy occurs when a child's caregiver injures or invents symptoms or signs of the child for the same reason.

We identified 4 cases of Munchausen syndrome assessed in two pediatric clinics in Bucharest. We bring them forward willing to draw attention over the assessment, diagnosis and their management and to emphasize the potential traps and subsequent treatment errors which may be easily made by the non-psychiatric clinicians.

Munchausen syndrome can be a challenging situation for clinicians since their mis-recognition may lead to pointless medical treatment, surgical procedures or dreadful complications.

Keywords: Munchausen syndrome, Munchausen syndrome by proxy, factitious disorder, pediatric.

INTRODUCTION

"Munchausen syndrome" (MS) describes "the patient who chronically fabricates or induces illness with the sole intention of assuming the patient role" [1]. Other authors state that it is "characterized by the intentional production or feigning of physical or psychological signs or symptoms, with a psychological need to assume the sick role" [2]. The term "Munchausen Syndrome" was first used in 1951 by doctor Richard Asher, who described patients who distorted their ailments and looked for medical assistance in multiple places for extended duration. The majority of dr. Asher's initial cases were represented by abdominal, hemorrhagic, or neurological emergencies [3]. Such reports of sickness, "dramatic and untruthful", were considered similar to the ones portrayed by the "Baron of Munchausen", Rudolf Erich Raspe's 1785 character based on the German baron Hieronymus Karl Friedrich Freiherr von Münchhausen, who was known for his overstated stories of unattainable accomplishments [4]. Nowadays, MS is considered compatible with the term "Factitious Disorder Imposed on Self" (FDIS). Some authors consider that a proper description of MS would be "a particularly severe and chronic presentation of FDIS" [5].

MS has been broadly studied and described in surgical patients. Different names - more or less euphemistic were associated with the condition of factitious disorder in surgery: laparatomphila migrans, surgery mania, scalpelophilia or mania operativa. Surgical patients with factitious disorders usually get great advantages from their condition, like receiving narcotics, immobilization or a permanent company for their disability [6].

Both MS and MSBP are challenging from a psychiatric clinician's and a non-psychiatric physician's perspective. Even more, these may represent extremely dangerous pitfalls and traps in pediatric practice with possible legal and ethical implications.

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MATERIAL AND METHODS

In the following lines, we will focus on four cases of this rare, but serious psychiatric disorder in children.

The collected cases are a result of a joint interdisciplinary experience between the Child and Adolescent Psychiatry Clinic in "Alexandru Obregia' Clinical Psychiatry Hospital' (first two cases) and Pediatric Surgery Clinic of "Marie S. Curie" Emergency Clinical Hospital for Children (3rd and 4th cases)- both from Bucharest, Romania, in the last 10 years.

We aim to draw attention over the assessment, diagnosis and management of MS/MSBP/FDIA through these cases and to emphasize the potential traps and subsequent treatment errors which may be easily made by the non-psychiatric clinicians. We consider that providing this information could be useful for other clinicians in recognising MS/ MSBP/ FDIA, while also helping to identify the adequate intervention and support services.

RESULTS

Case 1

A 5-year-old male child was referred for mental status assessment from a pediatric service where he was hospitalized for a state of obnubilation of unspecified etiology. The patient came from a rural area and he was not integrated in an educational structure. He lives with his parents, grandmother and 2 younger brothers. His father commuted daily for his job and his mother took care of the household. The patient's grandmother received treatment for epilepsy. The patient had multiple presentations in pediatric emergency and neurology services for altered consciousness with sudden onset in a healthy child with no significant history and with typical development.

The patient was repeatedly investigated in both pediatric and pediatric neurology services without identifying an organic etiology (pediatric, toxicological, traumatic, neurological or infectious cause) of altered consciousness. The intensity of the episodes fluctuated from numbness to coma, with complete recovery each time, after providing vital support in the pediatric service.

During the last hospitalization, the patient's status was fluctuating, alternating between periods of altered state of consciousness and periods of recovery. During this time, in the trash bin from the patient's room a foil of antiepileptic drugs was found, medication that was not part of the patient's treatment, nor of his mother. The concentration of the respective medication in the patient's blood was subsequently dosed, resulting in high values. The Child Protection Service was informed and the parents were confronted with this situation. The authorities were informed as well, given the suspicion that the alteration of the state of consciousness was due to the administration of antiepileptic medication without medical prescriptions, a suspicion that was later confirmed.

The family was referred for complex assessment of both the child's and the mother's mental status to the Pediatric Psychiatry and Psychiatry service respectively, in collaboration with Child Protection services, that supervised the family situation and provided counselling and support services the child and for the family.

We consider that this case exemplifies MSBP, as the mother's administering her child medication without prescription, without a declared purpose, determined the appearance of a range of symptoms that can have a significant, even lethal impact on the child's life.

Case 2

The second case we present is that of an adolescent girl coming from an urban area, with multiple hospitalizations in pediatric services, in several cities, for various symptoms in the gastric and urinary tract. We mention that the teenager has been taken care of by the same foster family since she was a 1-year old, a family with which he has a good relationship. No special pathological antecedents were detected and from a cognitive point of view the adolescent has very good school results.

From the patient's history we gathered that in the previous 2 years she was hospitalized on numerous occasions for various gastrointestinal symptoms (epigastralgia, nausea, vomiting, decreased appetite) for which no organic cause had been identified. Moreover, the adolescent was periodically hospitalized for complete urinary retention that persisted between 4 and 7 days and required urethral catheterization. No organic cause had been identified for the urinary retention either, and the teenager declared that "she will urinate only when she is ready" and that "she does not mind being catheterized". She managed to carry out her daily activities while being urinary-probed and went like this to school every day, where she won the sympathy of colleagues and teachers due to her condition. The adolescent also suffered multiple

accidents that she attributed to carelessness but they were often self-inflicted and resulted in trauma or orthopedic problems that required hospitalization.

For the symptoms she presented, the adolescent underwent multiple hospitalizations every time developing new symptoms whenever the discharge was announced, so that she obtained the extension of hospitalization and proceeded to new investigations. Most of the time, after completing the work-up, she was transferred to a pediatric psychiatric ward. She benefited from an individual therapeutic program, as well as family counselling, with fluctuating evolution of the symptomatology.

We consider that this case is suggestive for MS, as the adolescent presented various somatic symptoms unexplained by an obvious organic pathology, with frequent requests of pediatric services.

Case 3

This case is of a 16-years old female patient who was transferred to a tertiary pediatric surgery clinic for a suspicion of foreign body in a surgical wound (McBurney's incision for appendicitis). One month prior to this admission she was operated in another clinic for acute appendicitis and one week later she was readmitted in the same hospital declaring that she succeeded to pull out a needle fragment out of the incision space on her own. Therefore, at that moment the patient underwent wound exploration under general anesthesia without any particular findings.

She kept complaining that textile threads are coming out of her surgical wound, therefore she was referred to our unit. An abdominal X-ray didn't reveal any radio-opaque material around the surgical area, nor in the abdomen or pelvis and abdominal ultrasound of the soft tissue appeared to look normally. During the hospitalization on the surgical ward, the auxiliary medical staff had the chance to surprise her while she was trying to insert small textile threads into the scar. Therefore, psychiatric evaluation was recommended and MS diagnosis was made.

Unlike the first two cases presented, this situation could have led to a second wound/abdominal exploration under general anesthesia, setting the patient on a useless surgical and anesthesia risk.

Case 4

A 14-years old girl referred to "M.S.Curie" Outpatient Care Surgical Unit for a severe walking impairment syndrome associating chronic progressive severe limb pain. The patient was known as a high-level gymnast who suddenly interrupted her activity two years before. She has been known to our clinic for more than 16 months when she was presented for unsystematized recurrent ankle pain. At that time, the physical examination, radiological evaluation and blood work-up was unremarkable, therefore she was dismissed. Since then, the patient declared her status progressively worsened, with persistent pain in the lower limbs, at the present stage being wheelchair-dependent. During all this time, she was repeatedly re-evaluated by many different physicians (orthopedists, kinesiotherapists, neurologists etc.) without any diagnosis. Facing this impressive impairment of her lower limbs, a multidisciplinary board (neurologist, orthopedist, rheumatologist, surgeon) was united in our clinic and a comprehensive evaluation was conducted without identifying any cause for the impairment.

The costs of the evaluation were significantly high taking into account all the rare neurodegenerative and rheumatological diseases which were excluded, but justified by the family despair. Eventually, she was referred to the psychologist who presumed the diagnosis of MS and referred the adolescent to a psychiatric clinic. Her condition met a spectacular improvement after one month of psychotherapy.

DISCUSSION

The first written reports on Factitious disorders date back to the 19th century, with an official mentioning in the third edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM III) from 1986, where a very important distinction was made between the conscious and the unaware producing of a symptom (as it is the case in hysteria) [7]. In the last few years, the newer technological advancements and practices have determined the emergence of terms such as "Munchausen Syndrome by phone" or "Munchausen Syndrome by internet" [8, 9].

Looking in the latest edition of the DSM, the following FDIS diagnostic criteria can be found:

- "The patient feigns psychological and physical signs and symptoms, or induction of lesion or disease; factitious disorder;
- The individual presents him/herself to others as ill, impaired or injured;
- Fraudulent behavior is evident even in the absence of obvious external rewards;
- Individual's behavior is no longer well explained by a disorder, such as delirium or other psychotic condition" [10].

However, relying on these criteria alone is insufficient, as the diagnosis process is more complex, requires extended analysis of medical records, clinical interviews and both clinical and psychiatric evaluations, processes that can expand over large periods of time. The current literature provides various guides on identifying this disorder. Some steps that could be of aid in diagnosing and managing such a case are: performing an initial psychiatric examination and clinical interview, after which one could consider FDIS in the differential diagnosis; collecting thorough heredo-collateral and personal antecedents, as well as investigations history and medical records from previous professionals; conducting laboratory investigations for differential diagnoses; consulting with a psychiatrist, focusing on differential diagnosis as well on developing ethical treatment options; forming a multidisciplinary case team; putting together a cautious multi-approach therapeutic intervention that encompasses both inpatient and outpatient care; long-term monitoring and treatment-evaluation [11].

Obtaining exact prevalence data on FDs is quite difficult, as patients with such diagnosis provide unreliable history information and, in some cases, most often than not, when they gather that the medical staff is about to discover their feigning of symptoms they request their discharge against medical advice. Thus, it is quite difficult to collect accurate data for analysis and prevalence studies. We found two studies on adult inpatient groups that analyzed the incidence of FD in a clinical hospital. Sutherland *et al.* wrote that "0.8% of 1,361 patients referred over a 3-year period to a consultation–liaison service (CLS) for psychiatric evaluation in Canada were diagnosed with FD". A similar incidence (0.62%) was found by Kapfhammer and colleagues, after analyzing data from 15,000 patients from a German teaching hospital over an 18-year period [12-14].

While getting valuable prevalence estimates of FD in adult populations is difficult, obtaining such information in child and adolescent groups is almost impossible, as there is significantly less recognition that pediatric afflictions may also be purposely distorted and no findings have been shown to be pathognomonic [15, 16].

Of the narrow dependable prevalence data on child and adolescent FD available in current studies, we found that Ehrlich *et al.* reported in a retrospective study on 1,684 patients who were referred to a pediatric CLS (consultation–liaison service) over a 12-year period (Sample I) and 12,081 patients who were treated in a tertiary-care child health center from 2003 to 2005 (Sample II) that 0.7% of patients from Sample I had received a FD diagnosis and, respectively, 0.03% of the cases in Sample II [14].

A particular type of MS - "Munchausen syndrome by proxy" (MSBP), currently classified as Factitious disorder imposed on another (FDIA) since 2013, when the "5th edition of the American Psychiatric Association's Diagnostic and Statistical Manual" [9] was published, is characterized by "abusively and compulsively falsified physical, psychiatric or developmental disorders in a victim" (a child or an adult - more frequently a child) with an internal motivation, responding to certain psychological drives of the abuser, such as the need for attention [17, 18]. This new definition was proposed as it characterizes a set of behaviours rather than an underlying psychiatric disorder, thus being considered more accurate than MSBP [19]. Recent recommendations suggest that the use of MSBP is proper when referring to the abuse, while the perpetrator's psychopathology should be characterized as FDIA [4, 20].

The road leading to this nomenclature was not a smooth one. After Asher coined the term "Munchausen syndrome" in 1951, in 1977, dr. Roy Meadow described, for the first known time, cases in which caregivers deliberately determined or falsified different symptoms in their children, aspiring for appraisal for consequential dedicated care given to an ill child. He called this "Munchausen syndrome by proxy": "Here are described parents who, by falsification, caused their children innumerable harmful hospital procedures – a sort of Munchausen syndrome by proxy" [21, 22]. Even though health professionals such as dr. Donna Rosenberg [23] continued raising awareness about MSBP by publishing case report series of this type of child abuse, the "DSM-III" (1980) and the "DSM-III-R" (1987), although listing the term Munchausen Syndrome as a diagnosis, did not mention MSBP. The next editions of the "DSM" ("DSM-IV" and "DSM-IV-TR") proposed a classification of MSBP, and the DSM-5 distinctly described it as an individual disorder [19]. The current in-use edition of the "International Classification of Diseases by the World Health Organization (ICD 10)" describes Factitious Disorder (F68.1) and MS as synonyms, while not separately describing MSBP or FDIA. However, the ICD 11, which is momentarily only available online, underlines the discrepancies between "Factitious Disorder Imposed on Self" (6D50) and "Factitious Disorder Imposed on Another "(6D51) [24, 25]. According to DSM 5, the

following criteria must be met in order to make the diagnosis of FDIA:

- "The abuser engages in the deceptive falsification of physical or psychological signs or symptoms, or of induction of injury or disease in another;

- The abuser presents the victim to other as ill, impaired or injured;

- The deceptive behavior is evident even with absent external rewards;

- The behavior is not better accounted for by another mental disorder (e.g., psychotic or delusional disorder)" [10].

Nevertheless, as is the case with MS, diagnosing MSBP does not rely on DSM criteria alone, as it implicates an intricate process of putting together pieces of information that don't quite add up from the clinical interviews, clinical and psychiatric exams, patient history, and lab test results. In a literature review of 117 case studies of FDIA, Rosenberg identified a period of approximately 14.9 months necessary to identify a developing abuse [23]. There are several behaviours that a pediatrician should be aware of, like looking for various medical opinions, a resistance towards repeated confirmation of the child's health status or reporting unexplained symptoms [26]. Additionally, conducting an extensive review of medical records could be of help when trying to determine fabrication of illness, the goal being to assess if intentional deceit has haooened and, more than that, if it is expected to recur. One should see as a warning sign the caregiver's failure to provide these records or reporting them as lost [17].

Estimating an accurate prevalence of MSBP is a difficult process, mostly because it is under-diagnosed and thus under-reported, as it is the case with the majority of child abuse and neglect forms. The American Academy of Pediatrics (AAP) estimates an incidence ranging "from 0.5 to 2.0 per 100,000 children younger than 16 years" [20]. MSBP has been described in more than 20 countries, with aspects such as culture, race, or socioeconomic status not constricting it [27]. Studies conducted in specialized clinical settings provide higher estimates of 1%–13% [28, 29].

In most cases, the affected parties are children, but they can also be other categories of defenseless beings, such as the elderly or even animals. In a review published by Abdurrachid *et al.* after analyzing 108 articles on MSBP (81 case reports) published between 2004 and 2019, it was found that 51% of the victims were male and 43% - female (6% - not reported). Among the children victims, the mean age was 62,3 months [4].

A need for attention was considered by some authors as a valid potential motivation behind the parent's behaviours when imposing falsified ailments on their children[30]. Researchers that focused their attention on perpetrators profiles suggest that in most cases these people are "young (25–31.43 years), female, married, and the mother of the victim, with many having been sexually or physically abused" [29]. Personality disorders (frequently Antisocial, Borderline, Histrionic or Narcissistic), mood disorders, and somatoform disorders are common, as are aspects of FDIS. These mothers are usually familiar with medical terminology, as they spend a lot of time in the hospital or they "may have had prior training in the medical field" [4]. It is important to mention that "Hypochondria by Proxy" cannot be classified as FDIA as it does not involve deception and it is caused by "pathological anxiety about a child's health" [29]. In similar cases, parents with Asperger Syndrome can be excessively worried in regards to the possibility of rare diseases that could be ignored in their children [27, 29].

The victims can be harmed both directly and indirectly by the abusers. Directly - by the abuser's falsifications, and indirectly by being submitted to multiple, strenuous, redundant medical evaluations, procedures and interventions. In the case of children, depriving them of education during this process or making them miss developmental opportunities can also be part of the abuse [31].

Although usually cases of MSBP have a chronic evolution, they can be frequently exacerbated, presenting acute, diverse, and inexplicable symptoms [4]. In the reviews found in literature, there are listed various behaviors of the parents that can result in affecting the child's health. The most common are medication administration without medical recommendation, complaining of symptoms that cannot be clinically objectified (eg fatigue, vomiting, frequent urination) or requesting numerous medical opinions. The parent provides false information about the child's health status most often being supported by the child victim. The false version about the child's health is invoked not only in the medical office but also in family life, school or network of friends, the child being unable to carry out his usual activities [31, 32].

The existing guidelines in the literature identify different patterns of abuse: in some cases all the children in the care of the abuser are involved, other times only one child (for example the youngest or the one who raises the most problems for the parent). The range of exaggerated symptoms or induced medical disorders is

wide - from somatic disorders (allergies, gastrointestinal disorders, infections) to mental disorders (attention deficit, school difficulties, behavioral disorders) [22, 31, 33, 34].

The risks for the medical system consist in carrying out investigations and administering unnecessary treatments that, in the long run, although well-intentioned, contribute to affecting the child's health [11, 32, 35].

After formulating that MSBP diagnosis, the future development and progress of the respective child are highly impacted by the methods used during the case management process, as MSBP can cause disability, physical illness, social and emotional problems, and even death. The purpose of the therapeutic intervention is to develop the ability to function according to the chronological age, to increase the capacity to correctly identify the state of health and, last but not least, to increase self-esteem. Maintaining functioning in the school environment and maintaining social interaction with the group of friends are also objectives of the intervention and indicators of well-being. Regarding the abuser, the intervention should focus on increasing his/her capacity to recognize the abuse, to empathize with the victim and to improve his/her care capacity [17].

Dreadful situations for a surgeon have been previously reported in the late diagnosed MS context: persistent entero-cutaneous fistulas following appendectomy due to self-introducing of faeces in the wound and requiring 7 surgical interventions until the factitious disorder was assessed [36] or chronic complications of plastics surgery leading to limb amputations [37]. When it comes to suspected MS patients in the surgical area a set of self-injury clues may be pointed out: long history of unexplained multiple surgeries, strange course in wound healing contrary to the surgeon's clinical and work-up observations, a remarkable desire from the patient to undergo surgery, atypical signs, symptoms or hospital stay length (contrary to the one required for a certain operation), any evidence of paraphernalia. Clinical criteria for self-injurious behaviour in surgical patients include most of the times joint stiffness, dislocation or fractures, wound-healing distress, chronic lymphedema and superficial wounds (ulcers, scratches) for which the patient develops an atypical interest [6].

In conclusion, for physicians and mental health practitioners who may experience this potentially lethal type of psychopathology, MS/ FDIS and MSBP/ FDIA pose specific challenges. It can often take years before someone suspects MS/ MSBP due to elusive facts, disinformation, and deceptive yet actual disorders. A multidisciplinary team represents the best chance to observe these situations, decide whether abuse actually exists, record the abuse, and concentrate on the patient's health and welfare. Among the experts in these teams, mental health professionals could provide useful input by identifying suspected MS/ MSBP, adequately approaching expected denial, participating in the development and implementation of an exhaustive assessment, counselling the team on appropriate evaluation procedures. As follow-up data on such cases is almost non-existent, it is of utmost importance that this type of studies is performed in the future, thus providing vital information for efficient therapeutic interventions.

Non-psychiatric clinicians should always rely earlier on psychological or psychiatric expertise, when puzzle pieces of the diagnosis process do not fit well easily and constantly. This may lead to early diagnosis of MS/MSBP and avoidance of unwanted, unpleasant events with dramatic legal consequences or medical complications.

Conflict of interest

The authors declare that they have no conflict of interest.

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