

OVERPROTECTIVE MOTHER OR MUNCHAUSEN SYNDROME BY PROXY? CHARACTERISTICS OF THE DISEASE BASED ON A LITERATURE REVIEW

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ABSTRACT

Background: Munchausen syndrome by proxy (MSBP), first described by Meadow in 1977, is a mental disorder in which the caregiver or parent (usually the mother) causes the child to become ill to attract attention. The disease most often affects women, with sufferers usually appearing very caring and involved in the treatment. However, they harm the child, leading to physical and mental damage and sometimes death. The caregiver fabricates disease symptoms and will report false symptoms in the child or directly cause the disease. Moreover, they agree to diagnostic procedures and numerous medical interventions and deny the cause of the disease, and after separation from the caregiver, the child's condition improves.

Aim of the study: The purpose of this review is to present a characterization of MSBP, taking into account its diagnostic difficulties, treatment methods, and consequences for the child.

Material and methods: All relevant publications were retrieved from the PubMed and Google Scholar databases, with keywords including “Munchausen syndrome by proxy” (PubMed – 780 results; Google Scholar – 10500 results), “Munchausen syndrome by proxy diagnosis” (PubMed – 612 results; Google Scholar – 7940 results), and “Munchausen syndrome by proxy treatment” (PubMed – 301 results; Google Scholar – 8210 results), with 53 papers selected. Half of the studies had been published in the last five years.

Results: The main MSBP diagnostic difficulties include the low age of the victims and the frequent changes of treatment centers, which often worsen the child's condition. According to some scientists, MSBP psychotherapy treatment may produce the expected results for the patient and child. Children can die or develop future mental and physical problems as a result of their parent's disorder.

Conclusions: MSBP is a disease that is usually severe and difficult to diagnose and treat, and to which children fall victim. In order to diagnose MSBP, attending physician vigilance is essential.

KEYWORDS: Munchausen syndrome by proxy, medical child abuse, diagnosis

BACKGROUND

Many caring mothers are often overly concerned with their children's safety, health, and happiness. However, worrying about the condition of their children sometimes causes anxiety - after all, every mother wants the best for her children. But where is the line drawn between a caring mother and a mother who can harm her child?

Munchausen syndrome by proxy (MSBP) is a mental disorder characterized by the caregiver or parent (most often the mother) causing a pathological state in a child, such as a mental or physical illness. The purpose of such behavior is to attract other people's attention or gain sympathy [1-3]. According to Flaherty and Macmillan, such activity focuses on the child and not the caregiver and is a form of child abuse [4]. MSBP is classified into sham disorders, in which the caregiver uses cunning to make the child a patient, often leading to health damage that may cause death [2].

The syndrome was named in honor of a German named Baron von Munchausen, who lived in the 18th century and became a symbol of "colorful blasphemy" due to giving false information about himself and exaggerating his achievements (there were even numerous fairy tales in his honor) [5]. MSBP was first described by Meadow in 1977 as a disease entity in which parents or caregivers, almost always the mother, lie about a child's illness and fabricate symptoms. However, Munchausen syndrome, from which MSBP originates, was already described in 1951 by Asher [6-9].

There are no accurate MSBP epidemiology data in the literature, which may be influenced by its diagnostic difficulty, with current evidence focused mainly on severe cases. Therefore, it is possible to underestimate the statistics of the disease [6,10,11]. In Poland, MSBP frequency is estimated at 3/100,000 children, with several cases reported annually (Barent et al., 2012) [12]. McClure et al. estimate the total incidence of MSBP victims under 16 at 0.5/100,000 and children under one at 2.8/100,000 [13-14]. According to Denny et al., MSBP has an incidence of 2/100,000 in New Zealand in children under 16, with a median age at diagnosis of 2.7 years. As such, the most common MSBP victims are preschool children [15]. Unal et al. reported a frequency of 0.4/100,000 in children under 16 and 2-2.8/100,000 in children under one [16]. Meanwhile, research by Bass et al. (2011) showed that children under five are the most common victims [17]. In a Turkish study by Ozdemir et al. (2015), victim age ranged from two months to 13 years, with a mean of nine months (diagnosis ranged from four months to 13 years, with an average of 46.5 months) [18]. According to Sheridan et al. (2003), the age of abused children ranges from a few weeks to adulthood (the mean age of victims was

four years, while the average diagnosis was made at 21.8 months) [19].

Research indicates that MSBP perpetrators are usually women (92-100%), young people (25-31), and married people (42-79%) [16-24]. Despite much research on MSBP, its etiology remains controversial, with most of the available research focusing on the profile of the victims, not the perpetrators [16,24]. It appears that women are more likely to abuse their children through medical means, with the same research suggesting that working in the healthcare profession is more common among MSBP perpetrators. However, many perpetrators fabricated their professional history to gain the trust of medical personnel [25].

It seems correct to say that, in many cases of MSBP, the perpetrator will have mental disorders. Indeed, personality, mood, and somatic disorders are often present, with borderline personality disorder being the most frequently reported. Dube et al. also described a high rate of depression and substance abuse, while Weierich and Nock demonstrated a relationship between self-harm and MSBP [26-27]. However, the absence of any psychopathology was reported in 4.59% of cases (Sirka et al., 2018) [21]. Mental stress can also trigger an obstetric event that can disrupt the mother-child relationship. Therefore, it would be wise to monitor women for MSBP by interviewing them in maternity wards [16].

MSBP has four essential characteristics, including the deliberate fabrication of disease or somatic symptoms by the caregiver, in which they present the child as sick, handicapped, disabled, or injured. The child then undergoes many diagnostic procedures, and the perpetrator denies the cause of the child's illness. The child's symptoms disappear after separation from the perpetrator during their hospital stay, although cases have been reported where the child is abused during hospitalization (Vennemann, 2005), and the child's condition deteriorates after discharge. The caregiver's action may be one-off or recurrent, with the latter occurring much more often [6,19,20,27]. A caregiver may fabricate a history of false symptoms or directly cause disease in the child through drugs, toxins, infectious agents, choking, changing laboratory samples, and falsifying body temperature, which leads to medical interventions and sometimes serious invasive procedures and surgery. These actions result in the child suffering physical and mental harm [28-29].

AIM OF THE STUDY

The article aimed to review reports on MSBP diagnostic difficulties, treatment methods, and long-term consequences for the child. In addition, based on the results, we also consider whether perpetrators are guilty of committing a crime.

MATERIAL AND METHODS

The inclusion criteria used in the review included publication date, compliance with the topic, and reliability.

Eligibility criteria

We analyzed studies published between 1951 and 2022 to present the history of the disease from its first description (1951). However, to enhance the study's credibility, the scientific evidence regarding diagnosis and treatment described in the results, and the epidemiological and etiology data in the introduction, are mainly based on works from the last ten years (35 references).

Sample: mothers with suspected MSBP presenting to health centers with children.

Phenomena of interest: disorder diagnosis, available treatment methods and their effectiveness, and the consequences of the disease for the child.

Evaluation: Any patient reporting will be evaluated.

Design: All types of observational studies, including cohort, case-control, and individual case studies.

Research type: qualitative, quantitative, and mixed studies.

Search strategy

The search was conducted using the PubMed and Google Scholar databases. The source texts were last reviewed on 25.10.2022. Keywords used included "Munchausen syndrome by proxy," "Munchausen syndrome by proxy diagnosis," and "Munchausen syndrome by proxy treatment."

Data collection process

Each of the three authors reviewed selected scientific articles for inclusion in the review, with each working independently. Abstracts were initially read, followed by full articles of the selected studies. The extracted data included the following information: recorded cases of MSBP, diagnostic possibilities and methods, treatment effectiveness, and consequences for the child. The risk of bias for each study was independently assessed by the same authors. The collected data are presented in text form to allow for a thorough understanding of the topic under discussion.

All relevant publications were retrieved from the PubMed and Google Scholar databases using the keywords: "Munchausen syndrome by proxy" (PubMed – 780 results; Google Scholar – 10500 results), "Mun-

chhausen syndrome by proxy diagnosis" (PubMed – 612 results; Google Scholar – 7940 results), and "Munchausen syndrome by proxy treatment" (PubMed – 301 results; Google Scholar – 8210 results), with 53 papers selected. More than half of them (35 results) were published in the last ten years.

RESULTS

Study selection

Of the articles found, 53 studies were ultimately qualified for review since some did not meet the inclusion criteria after reviewing the abstract, while some of those meeting the criteria were disqualified due to low credibility (insufficient evidence).

Discussion

MSBP mimics a variety of disease entities that range from mild to severe to possibly fatal. Ozdemir et al. (2015) showed a range of symptoms in MSBP victims, such as seizures, hypoglycemia, apnea, chronic abscesses, bleeding, hemiplegia, and sexual abuse [16]. Sirka et al. (2018) described a cutaneous form of MSBP in a 15-month-old child who was consulted from six months of age due to blisters and erosions on the skin (leaving leaf-shaped scars), which suggested burns (the smell of various oils and the flow of hot oil from the skin surface) [21].

Kuhne et al. (2019) described the case of a nine-year-old boy who was repeatedly hospitalized due to seizures with a disturbance of consciousness. The mother had a history of rash, photosensitivity, alopecia, arthralgia, hypertension, haematuria, seizures, and the presence of antinuclear antibodies (though laboratory tests in the hospital did not show any other antibodies or abnormalities). Previously, the child was in medical institutions with fever and hematuria, while on another occasion, he presented with fever, headache, vomiting, photophobia, phonophobia, and dizziness. Physical examination revealed agitation, confusion, atactic gait, slurred speech, horizontal nystagmus, painful facial expressions, tachycardia, and weight loss, but magnetic resonance imaging (MRI) and cerebral spinal fluid (CSF) samples indicated no abnormalities. MSBP mimicking systemic lupus erythematosus (SLE) was suspected, and phenytoin intoxication was confirmed (more than twice the normal concentration). The child was separated from the mother [30].

Sahin et al. (2020) presented a case report of a 15-month-old boy admitted to an infectious disease clinic with suspected acute gastroenteritis. The baby developed a high fever due to intravenous fluid ad-

ministration, and his blood showed the development of many microorganisms, which suggested MSBP [31]. Nogueira-de-Almeida et al. (2018) presented a rare case of MSBP in a 4.5-year-old boy who was consulted for obesity in a nutrition clinic 100 km from his place of residence. The child was examined previously in several other centers for severe obesity (48 consultations, during which numerous complaints were reported). The boy's mother, who appeared to be caring, provided a nutritional questionnaire in which she indicated an extremely low caloric intake and vigorous physical activity, inconsistent with the clinical description, and a BMI (body mass index) of 29.3kg/m². Laboratory test results were normal (as were hormonal, genetic, and neurological tests), and obesity treatment was unsuccessful. MSBP was diagnosed, though the mother refused psychiatric treatment and became aggressive [32].

Gupta et al. (2021) described the case of a 4-year-old girl brought to the emergency room by her mother with symptoms of polyuria, polydipsia, and weight loss. The mother reported high glucose and glycosylated hemoglobin (HbA1c) levels, which might suggest diabetes (there was a family history of diabetes, and the woman had gestational diabetes). Test results showed normal glucose and HbA1c levels, although home tests continued to indicate high glucose levels, and a "private" laboratory found that HbA1c levels were still elevated (both parameters were normal again in the clinic). During hospitalization, the child had normal parameters (extended diagnostics were carried out), and suspected MSBP was diagnosed [33].

According to Deaton et al. (2022), there were 181 exposures to superwarfarin in the USA in 2014, of which 142 were in children under the age of six, and 170 were found to be unaware. No major clinical episodes or deaths were reported [34].

The Diagnostic and Statistical Manual of Mental Disorders (DSM) describes that the perpetrator's goal is to maintain the diseased role by proxy. In cases of MSBP, a child is often affected. There must be no external motives for this disorder, such as financial benefits, avoiding legal problems, or free time from work. As such, MSBP is not included in the simulation [29,21,35]. According to Scheurman et al., the stimulus for such behavior is satisfaction with the tests and treatment of the child (victim). The mother attempts to make the child dependent on her by becoming involved in their care and the treatment process [21,35]. Moreover, the mother is overly protective and uses the child to attract attention (at the child's expense) in order to satisfy her desires [36].

A diagnostic difficulty in MSBP is the low age of the victims, which makes determining the cause and nature of the injuries challenging. Indeed, the most common victims are newborns, infants, and young children, with a mean age of 39.8 months, according to

Trendak et al. [36]. A doctor's vigilance and suspicion, and an interview with family members, are essential for an MSBP diagnosis. As such, pediatricians should be alert to discrepancies in the history of the child during physical examination, and attention is especially important when the child's condition improves while in the hospital and worsens after discharge [4,21]. The diagnostic and therapeutic problem also involves the patient's denial of the diagnosis, with mothers often raising reservations, becoming hostile, and changing to another facility, which leads to a vicious circle. In addition, the affected person agrees to have the child tested but ignores the pain and suffering associated with numerous medical interventions.

Frequent delays in MSBP diagnosis contribute to further deterioration of the child's condition [36-37]. Later in life, MSBP victims are often diagnosed with mental health disorders, including behavioral, attention, and sleep disorders, post-traumatic stress disorders (PTSD), and MSBP toward future children [36].

1. The perpetrator falsifies physical or psychological signs or symptoms or causes injury to another person (victim).
2. The perpetrator presents the person (victim) as sick, injured, or disabled.
3. The behavior occurs in the absence of external incentives.
4. Other mental or psychosomatic disorders are excluded, and the behavior cannot be explained by another disorder

Figure 1. The diagnostic criteria for Munchausen syndrome by proxy according to the Diagnostic and Statistical Manual of Mental Disorders 5 (DSM-5) [6, 38-39]

1. A history of repeated injuries (physical or mental), illnesses, and hospitalizations
2. Reports of physical and mental injuries that no one else can confirm
3. Symptoms disappear after the victim is isolated from the perpetrator
4. Symptoms worsen after discharge from the hospital
5. Inconsistent symptoms
6. Excessive involvement in medical matters
7. Increased relationships with patients and healthcare professionals
8. The perpetrator speaks for the child
9. Symptoms similar to other patients appear during hospitalization
10. Symptoms do not fit any disorder and do not match test results
11. Sustained response to treatment
12. Pathological lies (pseudologia fantastica)

Figure 2. Alarming symptoms [6,40].

The prognosis largely depends on the severity of the physical and psychological injuries sustained. Filho et al. claim that treating MSBP causes many difficulties for specialists. In their opinion, based on the reviews and empirical reports of clinicians, it can be concluded that no psychological therapy provides the intended effects for this disease [6]. However, Nicol and Eccles think there are cases in which therapy brings the intended effect, but only in specific circumstances, such as when the patient is well motivated to confront their difficulties, has at least an average level of intelligence, and has no significant family and social problems.

The most substantial problem for psychotherapists is the fact that they have to trust the patient's truthfulness [41]. Boyd et al. believe that the patient's greatest chance of recovery is to stay in a psychiatric hospital and receive supportive, analytical, or psychoanalysis psychotherapy. Unfortunately, there is no information on long-term prognosis since the patient may go to another hospital and start the process anew. Nonetheless, a study showed improvement after psychiatric therapy in one in four respondents [42]. In four cases reported by Lopez-Rico et al., psychotherapy brought about good results by helping to repair the toxic relationship between mother and child and reinstating the autonomy and individuality of the victim.

The best results are gained by therapy in which the child is treated without the mother present. As a result, the highest level of separation of the child from the mother is achieved, indicating that the caregiver and victim require psychological help. The first step to successful treatment involves establishing a good relationship between the mother and the therapist based on mutual trust. The task of the therapy is to develop the mother's self-esteem, empathy, and ability to cope with stressful situations. Additionally, fathers should receive treatment, as they are most often described as not being involved in family life. A perpetrator's psychotherapy is based on working through childhood traumatic experiences, the relationship with her mother, and building and repairing her own identity. Moreover, socio-behavioral therapy is useful as it helps improve social skills and introduces new ways of meeting emotional needs other than child abuse [43].

It is common for a patient to manipulate doctors and their children to meet their own psychological needs, so therapy should focus on searching for and working through past situations that may have led to such a state. Most often, this includes a history of trauma or illness during which the patient experienced concern from others [44]. Lin and Chan believe that MSBP may be associated with personality disorders, such as narcissistic personality disorder or borderline personality disorder. The authors

argue that psychotherapy may be effective in such cases, but there is no evidence that biological or psychological therapy is effective. Long-term supportive psychotherapy and, after some time, confrontational psychotherapy can increase awareness of their diagnosis and improve compliance with therapeutic recommendations [45]. Unfortunately, only 12% of patients consent to treatment.

Diagnosis and treatment of MSBP are extremely difficult. However, diagnosis of the disorder remains crucial. The only question remaining is, who is guilty of committing a crime, and who is the victim? Sigal et al. [46] and Glaser [47] describe the phenomenon of a triple relationship between the perpetrator, the victim, and the medical staff. In order to increase credibility, the caregiver often falsifies test results and exaggerates or invents the child's ailments. Doctors are often unaware of the caregiver's intentions and consent to tests, treatments, or other aids, such as wheelchairs and financial support. The task of doctors is to not overlook the child's healable ailments and prevent unintentional harm from excessive examinations and unnecessary treatment. However, the actions of caregivers and doctors lead to the general deterioration of the child's physical and mental health, though it is unintentional in the case of doctors. In summary, the mother's actions consist of deception, obtaining material help, and perpetuating misconceptions about the child's health. In order to meet their needs, the caregiver requires doctors to confirm the poor condition of the child. In response, doctors and health care professionals may inadvertently cause harm to a child. All of these activities cause the victim anxiety about their health condition, the development of mental illnesses, false beliefs about their condition, and social isolation [46,47].

The frequency of death and serious medical events associated with MSBP is not exactly known. The mortality rate has been estimated at 9-31% but is most often in the range of 9-10% [48]. Unal et al. report that it occurs in 6-10% of MSBP cases [9], while Zarankiewicz et al. indicate mortality levels of up to 33% [49]. As many as 8.5% of the siblings of victims are also abused by their parents [41], and according to a meta-analysis of 451 cases of MSBP, 61% of 210 siblings showed signs of abuse, and 25% died [18,48]. In the case of MSBP described, asphyxiation by the mother caused the female victim's death [48]. The woman confessed to abusing her other two children by closing their mouths and nose, though the cause of death was unknown due to the failure to perform an autopsy. However, this case is considered serial MSBP. When an infant has an unexplained death, physicians should pay attention to the presence of the disease in the remaining children [48-50]. Tozzo et al. emphasized the role of clinicians in appropriate patient care and making the best medical decisions,

as well as dealing with the ethical aspects of these activities [51].

According to Glazier, MSBP victims suffer from multiple physical and psychological ailments, such as bedwetting, developmental delay, and irreversible brain damage with consequent learning disabilities, social phobias, hypochondria, difficulty concentrating, nightmares, being prone to theft, and even tetraplegia [52]. Developmental delay may occur in children aged five, and more than half also suffer from another chronic disease [53]. Zarankiewicz et al. share Glazier's views on the consequences of MSBP victims later in life. They also describe a greater risk of these children developing depression, anxiety, PTSD, and growth failure, than the rest of society.

There is a hypothesis suggesting that victims may experience MSBP in the future. Moreover, MSBP victims may be reluctant to seek medical attention, even if they have an acute illness. In addition, They may have difficulties with the proper perception of their own body, making it challenging for them to recognize disease symptoms [49]. Bertulli et al. believe that MSBP victims could experience physical and psychological injuries or even die as a result of unnecessary and potentially harmful tests and treatments [1].

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Limitations

MSBP research is limited by the diagnostic difficulties hindering the accurate estimation of its incidence. In addition, the disorder is only suspected in many cases because the perpetrator often has excessive suspicion of healthcare providers and will regularly change treatment centers. Therefore, the credibility of studies decreases due to insufficient evidence of treatments. Perhaps increased suspicion and awareness of medical child abuse among healthcare professionals and improved recording could help to increase the base of verified cases and allow a robust assessment of therapy effectiveness.

CONCLUSIONS

MSBP is a disease that is usually severe and difficult to diagnose and treat, and to which a child falls victim. Attention is drawn to the child's suffering, and they may develop mental and physical problems in the future or even die. Therefore, the vigilance of pediatricians and family history is necessary for a diagnosis and the commencement of therapy as soon as possible for the mother and the child.

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