CASE REPORT

Munchausen Syndrome by Proxy: A Rare but Real Type of Abuse

Marta Ezequiel, Carlos Escobar, Pedro Nunes, Helena Cristina Loureiro, Maria Lurdes Torre, Helena Isabel Almeida

Port J Pediatr 2021;52:197-202
DOI: https://doi.org/10.25754/pjp.2021.20072

Abstract

Munchausen by proxy is a rare form of abuse but has a high morbidity and mortality risk. A 2-year-old boy was admitted for aphthous stomatitis, and during hospitalization, he started persistent vomiting that was extensively investigated and treated accordingly with transient resolution. Before the planned discharge, he had an unexplained event of vomiting, sleepiness, petechiae, and hematuria. An investigation revealed hemolysis, hemoglobinuria, increased creatinine, and ketonuria without acidosis. Days later, he had another sudden nocturnal episode of vomiting, miosis, sweating, and absence of reactivity. He maintained persistent hemoglobinuria that was thoroughly investigated with no etiological diagnosis. Due to the sudden and inexplicable onset of symptoms, the absence of an etiological explanation and maternal social history, Munchausen syndrome by proxy by poisoning was suspected. There was clinical resolution in the mother's absence and an extended toxicological screening of blood, urine, vomit, and food showed toxic levels of isopropanol (hand sanitizer). There are no cases described in the literature of intentional poisoning with isopropanol and the diagnosis of Munchausen syndrome by proxy on the first manifestation of the syndrome is rare.

Keywords: 2-Propanol/poisoning; Child Abuse/diagnosis; Child, Preschool; Munchausen Syndrome by Proxy/diagnosis

Introduction

Child abuse is defined as any intentional or unintentional act by a parent or other caregiver that results in actual or potential harm to the child physical or emotional health.[1] Munchausen syndrome by proxy, also known as medical child abuse or factitious disorder imposed on another, is a dangerous form of child maltreatment, which was first described in 1977 by a British pediatric nephrologist, Roy Meadow. Its hallmark involves the fabrication or induction of a medical condition, usually by the mother, resulting in a flawed evaluation and treatment plan that can include unnecessary and multiple medical procedures, and resolution of the symptoms when the child is separated from the caregiver.[2-4] Perpetrators often have medical knowledge, may suffer from psychiatric disorder, or intend to gain social attention or money.[5]

Because the literature tends to describe severe forms of Munchausen syndrome by proxy, the scale of this form of abuse remains unknown and probably underestimated. Diagnosis can be challenging as its spectrum of presentation is broad and might include dermatologic, endocrine, gastrointestinal, hematologic, neurologic, metabolic, infectious, or rheumatologic manifestations and fever.[5] In a meta-analysis of 796 cases, child death was reported in 7.4% of cases and long-term disability in 7.3%.[6]

We report a case of Munchausen syndrome by proxy in a 2-year-old boy perpetrated by his mother, with unexplained symptoms and diagnostic tests results that evolved during an initial hospitalization for aphthous stomatitis, until a final diagnosis was made by identifying isopropanol in the child's biological samples. Our aim is raising awareness of the disorder among professionals to allow a more rapid diagnosis of Munchausen syndrome by proxy, since it is more common than many of the diagnoses that are routinely excluded before considering the possibility of abuse.

Case Report

A 2-year-old boy, presented to the emergency department of a general hospital with fever, aphthous oral lesions, vomiting, diarrhea, and refusal to feed over the previous two days. Aphthous stomatitis was assumed, and the patient was admitted to the pediatric ward.

Children and Youth at Risk Hospital Unit, Departament of Pediatrics, Hospital Professor Doutor Fernando Fonseca, Amadora, Portugal
Corresponding Author
Marta Ezequiel
https://orcid.org/0000-0002-0013-7686
marta.ezequiel@gmail.com
Hospital Professor Doutor Fernando Fonseca, IC19, 2720-276 Amadora, Portugal
Received: 14/05/2020 | Accepted: 04/12/2020 | Published online: 02/07/2021 | Published: 02/07/2021

Fever, diarrhea, oral aphthous lesions, and refusal to eat resolved in the first days but the vomiting continued. On the fourth day of hospitalization, he started hematic vomiting. Mallory-Weiss was suspected, and he was started on omeprazole. Daily vomiting persisted, usually after every oral intake attempt, rarely with specks of blood. On day 16, pediatric gastroenterology consultation suggested an upper gastrointestinal series followed by upper digestive endoscopy. A mild reflux was diagnosed, and hemorrhage excluded. A gastric biopsy revealed mild gastritis. Hematemesis on day 18 urged a nasofibroscopy to be performed to exclude iatrogenic injury from endoscopy, which showed hemorrhage of the left adenoid. A perinasal sinus computed tomography scan was ordered to exclude a nasopharyngeal vascular malformation and a moderate sinusitis was diagnosed. He was treated accordingly with nasal corticoid and antibiotics, and clinical improvement was noticed with complete tolerance of oral intake from day 24.

On the night of the 28th day of hospitalization, before the planned discharge date, the child vomited after dinner and was somnolent but arousable. A couple of hours later, he presented with flushing (mainly in the face), sweating, tachycardia, associated with petechiae in the trunk, and gross hematuria. The laboratory results revealed altered coagulation tests. Activated partial thromboplastin time (aPTT) was twice the control value, international normalized ratio (INR) 1.6 s, hemolysis with hemoglobin (Hb) 10 g/dL, lactate dehydrogenase (LDH) 900 UI/L and haptoglobin 10 mg/dL, a creatinine increase in eight hours (0.6 to 0.94 mg/dL) without increased urea, mild hyperglycemia (294 mg/dL), ketonuria without acidosis and 4.3 mmol/L blood lactate, and 3+ for blood in urinalysis with abundant erythrocytes on urine sediment (Table 1).

He improved in 24 hours but maintained red colored urine due to hemoglobinuria that was exclusively nocturnal, associated with recurrent abdominal pain, refusal to eat and sporadic vomiting also of nocturnal frequency. Nephrology and hematology consultation suggested a thorough etiological investigation (Table 2), which was normal.

The child had an irrelevant prenatal and perinatal history. He lived with his parents, attended day-care, and the family had help from the paternal grandmother. He attended regular visits in primary care. Regarding his past medical history, he had a history of three previous hospitalizations (one for pyelonephritis and two due to bronchiolitis in the first year of life) and he was on macrogol due to constipation. His mother was an unemployed baker, had personal history of breast cancer, and two self-induced abortions and revealed being a victim of domestic violence. His father worked in construction and was healthy. He had a healthy 7 year old paternal half-brother. The mother accompanied the boy during his hospitalization. She appeared to be affectionate and caring toward the child, always showing concern about his clinical condition and never questioned the medical team about the need for further diagnostic exams or about the discharge home. Nonetheless, she appeared to be somehow untrusting of the clinical team interventions and whenever discharge planning was discussed she would always show insecurity about it, justifying the request for psychological counseling during the hospitalization period.

Considering the unlikeliness of the existence of an organic disease to explain the clinical picture, after a thorough and extensive investigation and multiple subspecialties consultations, Munchausen syndrome by proxy was suspected. The timing of symptoms with the need to extend hospitalization along with the mother's behavior (untrusting of the clinical team interventions, unwilling to share information with the father, and a negative attitude toward discharge revealing to be unprepared and asking to extend hospitalization) were also in favor of Munchausen syndrome by proxy. In addition, the mother's breast cancer history was discovered not to be true. The child was referred to the Children and Youth at Risk Hospital Unit (*Núcleo Hospitalar Apoio a Crianças e Jovens em Risco* - NHACJR) of the hospital for further evaluation and management. The child was placed under observation in an open space unit with permanent nurse presence, with supervised meals and medication intake. The refusal to eat improved and the vomiting stopped but the red colored urine persisted. At day 36 of the hospital stay, he had a nocturnal episode of vomiting, sudden and transient miosis, sweating, and a lack of responsiveness. The lab results showed hemolysis, alterations of coagulation tests, and ketonuria without acidosis. There were no alterations in the head computed tomography nor in the electroencephalogram (EEG), and he had a complete recovery in a few hours. Common central nervous system depressant drugs were ruled out (benzodiazepines, tricyclic antidepressants, barbiturates). Samples of the blood, urine, vomit, and food found near the child bed was then collected and sent to the Instituto de Medicina Legal for a more extensive toxicological screening. After this episode, a psychological and psychiatric evaluation of the mother revealed a relevant background (born in Angola, grew up without her mother and was placed in foster care at age 11 after her father's death), emotional suffering, and depressive humor with the fear of abandonment and rejection.

Table 1. Laboratory results by day of the hospital stay

Variable	Admission	Day 28 (night)	Day 28 + 8 hours	Day 31	Day 36 (night)	Day 45
Hemoglobin (g/dL)	11.9	11.2	10.1	10.8	8.0	9.4
Leucocyte count (cells/µL)	15,900	24,900	11,200	9,200	11,200	6,800
Platelet count (cells/µL) 392,000		401,000	354,000	392,000	315,000	30,200
Glucose (mg/dL)	90	259	116		121	
AST (U/L)	19	98			58	33
ALT (U/L)	15	21			79	24
LDH (U/L)	256	900		727		
Bilirubin total/ conjugated (mg/dL)		0.58 / 0.1		0.34 / 0.10	0.52 / 0.10	
Creatinine (mg/dL)	0.46	0.63	0.94	0.60	0.14	0.37
Urea (mg/dL)	23.0	14.0	16.0	28.0	18.0	16.0
Prothrombin time		18.6	11.0	12.4	22.3	
INR		1.6	1.0	1.1	1.9	
aPTT		53.4	27.1	34.4	40.1	
D-dimers (µg/L)		732.2	446.9	458.0	728	
Urine sample		Gross hematuria, hemoglobinuria, proteinuria, glycosuria, ketonuria		Gross hematuria, hemoglobinuria 3+, no ketonuria	(day 37) Gross hematuria, hemoglobinuria, ketonuria	Normal
Urine sediment		Abundant erythrocytes and rare leukocytes		Rare leukocytes	Abundant erythrocytes (some dysmorphic) and rare leukocytes	
Venous blood gas	pH 7.39	pH 7.33			pH 7.41	
pCO2 (mmHg)	pCO_2 49.9	pCO_2 44			pCO_2 42.5	
Bic (mEq/L)	Bic 29.5	Bic 22.7			Bic 26.6	
Lact (mg/dL)	Lact 1.2	Lact 4.19			Lact 2.68	
Other relevant laboratory results			Haptoglobin 10.7 mg/dL (low)	24-hour urine: hemoglobinuria, no erythrocytes or epithelial cells, myoglobinuria 1,568 µg/L (normal < 1,000 µg/L);		Reticulocyte count 120,000 cells/µL (high), haptoglobin 88.0 mg/dL (normal)

ALT - alanine aminotransferase; aPTT - activated partial thromboplastin time; AST - aspartate aminotransferase; Bic - bicarbonate; INR - international normalized ratio; Lact - lactate; LDH - lactate dehydrogenase; pCO_2 - partial pressure of carbon dioxide.

Table 2. Diagnostic hypothesis for hematuria, hemoglobinuria, and hemolytic anemia without abnormal results

Diagnostic hypothesis
Renal or bladder disease
Paroxysmal nocturnal hemoglobinuria
Immune hemolysis
Erythrocyte enzyme deficiency
Acute intermittent porphyria

At this stage, the parents were confronted with the possibility of intentional intoxication during hospitalization, which both denied. The father was then asked to accompany the child for a period during hospitalization, and during this time the child did not present any of the previous symptoms, including red colored urine. He kept asymptomatic, and hospitalization was maintained as a protective measure while the test results were pending. On the 61st day of hospitalization, the results of all the samples sent to the Instituto de Medicina Legal revealed toxic levels of isopropanol (> 1,000 mg/L) and the presence of acetone in the blood and urine. The case was referred to the local child protection services and public prosecution services.

The child was discharged to a temporary institution, after a court decision, on day 62 of hospitalization. The

mother was referred to psychiatric intervention that she refused. After two months, the child was placed under the paternal grandmother's responsibility with the prohibition of visits of the mother until further psychiatric treatment.

Discussion

Munchausen syndrome by proxy is a form of child abuse that can be associated with high morbidity, with an impact on child physical and mental health, and mortality, which has higher rates in cases of suffocation or poisoning.[7,8] The child health can be affected, on the one hand, by exposure to harmful experiences in order to get medical attention for a falsified pediatric condition, and consequent side effects of unneeded treatments and procedures.[3] On the other hand, by a caregiver with a psychiatric disorder who fabricates or induces these conditions.[6]

We describe a case of Munchausen syndrome by proxy by induction, namely by poisoning. The child was admitted to the hospital for a real clinical condition, and in the child medical history, there is no knowledge of previously unneeded medical seeking behavior. Therefore, the first manifestation of the syndrome appears to have occurred during this hospitalization. Red flags for Munchausen syndrome by proxy, both for the child and the mother, arose during hospitalization: an atypical presentation, normal etiological examinations for persistent symptoms (vomiting and red colored urine), absence of response to the initial treatment, unexpected complications, and new symptoms in key moments (before planned discharge), symptoms, and signs disappearance in the mother absence, caregiver acceptance of invasive diagnostic procedures without any doubts, unpreparedness for discharge, perpetrator history of foster care integration, psychiatric disorder, domestic violence, and manipulation of her medical history (false cancer diagnosis). Domestic abuse and recent self-inflicted abortion may have contributed to triggering the episode in a woman with a background of abandonment and abuse.

Although the diagnosis of Munchausen syndrome by proxy is challenging, and can rarely be proven, in our case, the isopropanol found in the patient's specimens proved non-accidental intoxication. Retrospectively, the clinical picture is compatible with recurrent intoxication that started after stomatitis resolution. In the pediatric wardroom, a more private environment, recurrent vomiting, abdominal pain, and refusal to eat could be explained by lower doses of isopropanol mixed with food, while a higher dose, before planned discharge, could trigger more serious symptoms and the need to extend the hospital stay. When moved to an open space infirmary, the symptoms were less frequent and always nocturnal, as it was the less supervised time of the day. Alcohol-based hand-rub solutions containing isopropanol are available in the hospital setting.

Isopropanol intoxication is relatively frequent because of its easy accessibility (alcohol-based hand rub, swabs, perfumes, paint thinners, among others).[9] It is normally associated with accidental intake in young children,[10] recreational use in teenagers or alcohol-addicted adults, or with suicidal intent.[11] It is rapidly absorbed following ingestion with a peak plasma concentration occurring within 30 minutes, and a half-life of approximately 2.5 to eight hours. Isopropanol is metabolized by alcohol dehydrogenase to acetone, acetol and methylglyoxal, propylene glycol, acetate, and formate.[11]

The symptoms mainly involve the gastrointestinal tract, the central nervous system, and the cardiovascular system at higher doses. Milder symptoms include flushing, headache, dizziness, nausea, and vomiting.[10] In a review of accidental isopropanol in children presenting to the emergency department, vomiting was the most frequent presenting symptom (92%), followed by lethargy and ataxia (20%). Symptoms developed 0.5-1 hour after ingestion and resolved within four hours when the ingestion was a small amount.[10] The major features of severe poisoning are due to the central nervous system and respiratory depression, shock, and circulatory collapse.

An elevated osmolar gap, ketonemia, and/or ketonuria without metabolic acidosis, along with a sweet breath odor and central nervous system depression support the diagnosis of isopropanol intoxication.[11] A normal anion gap and the absence of metabolic acidosis may help differentiating methanol and ethylene glycol poisoning. Although rare, rhabdomyolysis, hemolysis, and hepatic dysfunction can occur.[11]

The laboratory results in this case showed ketonuria without acidosis, and the high serum creatinine with a normal blood urea nitrogen (BUN) and pH are suggestive of a possibility of acetonemia due to isopropyl alcohol.

Isopropanol concentrations may not necessarily correlate with clinical status, but serum concentrations above 500 mg/L are associated with toxicity and > 1,500 mg/L with coma.[11]

A search of the scientific literature retrieved no results for Munchausen syndrome by proxy and isopropanol poisoning, thus to the best of our knowledge the case presented here is the first report in the literature. In comparison with the results of other studies, in this

Table 3. Red flags for Munchausen syndrome by proxy diagnosis in the perpetrator and in the victim

Perpetrator	Victim
Extensive and inconsistent description of the child's disease	Atypical presentation
Clinical picture uncorrelated with the medical history reported by the parent	Observation and tests are normal
Acceptance of invasive diagnostic or surgical procedures without any doubts	Absence of response to the treatment
Seeking attention and understanding from the medical personnel	Symptoms/signs occur in the presence of the caregiver and disappearing in his absence
Requests for further interventions and opinions of other specialists	Unexpected complications or new symptoms/signs when tests are normal
Having some medical knowledge or linkage to the medical sector	Multiple hospitalizations and surgeries
History of a psychiatric disorder	Multiple comorbidities
Family problems	Father absent in the child's life

Adapted from: Paturej A, Pogonowska M, Kalicki B. Munchausen syndrome by proxy: A case report. Pediatr Med Rodz 2019;15:93-6 and Faedda N, Baglioni V, Natalucci G, Ardizzone I, Camuffo M, Cerutti R, et al. Don't judge a book by its cover: Factitious disorder imposed on children-report on 2 cases. Front Pediatr 2018;6:110.

case, the diagnosis was established after two months, during the first hospitalization, while in most case series, the mean time from the onset of symptoms to the time of Munchausen syndrome by proxy diagnosis is around 15 months, being this syndrome is rarely considered early in the child's illness.[6] We also must not assume that Munchausen syndrome by proxy does not occur in the hospital setting and that hospitalization is an effective protective measure, as the perpetrator is able to successfully simulate clinical symptoms despite child hospitalization in 54% of all cases.[4]

Munchausen syndrome by proxy must be considered in the presence of a child with unexplained or rare symptoms, the need of multiple diagnostic interventions and multiple subspecialities consultations, a lack of response to treatments, without a clear clinical diagnosis, and a history of repeated assistance in different health institutions. Medical professionals play an important role regarding both the initiation and perpetuation of Munchausen syndrome by proxy and particular attention regarding the existence of red flags is necessary (Table 3).[4,12]

In doubtful cases, all of the laboratory tests should be done or repeated in the presence of health professionals. When there is a suspicion of poisoning or the administration of drugs by the caregiver, samples of the patient and environment should be collected, and toxicology tests should be performed.[4]

As any form of child abuse, Munchausen syndrome by proxy may have a negative impact on the child's well-being and future consequences that might not be only physical but also psychological, thus emphasizing the importance of warranting the proper follow-up of the child and the family. Child protection should always be a priority when dealing with Munchausen syndrome by proxy.

WHAT THIS CASE REPORT ADDS

- Munchausen syndrome by proxy should be considered in the differential diagnosis of any child with unexplained persistent symptoms that do not correlate with the laboratory findings when there is a lack of response to treatment or the recurrence of symptoms with similar or different characteristics.
- If professionals are aware of this diagnosis and have a high index of suspicion, unnecessary investigation, treatment, hospitalization, and ultimately further harm to the child can be prevented.
- It should not be assumed that all cases of Munchausen syndrome by proxy will have a history of extensive healthcare utilization.
- The diagnosis is confirmed by the resolution of symptoms after separation from the perpetrator.
- The case described is the first report of Munchausen syndrome by proxy by isopropanol poisoning in the literature.

Conflicts of Interest

The authors declare that there were no conflicts of interest in conducting this work.

Funding Sources

There were no external funding sources for the realization of this paper.

Provenance and peer review

Not commissioned; externally peer reviewed

Consent for publication

Consent for publication was obtained.

Confidentiality of data

The authors declare that they have followed the protocols of their work center on the publication of patient data.

References

1. World Health Organization, International Society for Prevention of Child Abuse and Neglect. Preventing child maltreatment: A guide to taking action and generating evidence. Geneva: WHO; 2006.

2. Ali-Panzarella AZ, Bryant TJ, Marcovitch H, Lewis JD.

Medical child abuse (Munchausen syndrome by proxy): Multidisciplinary approach from a pediatric gastroenterology perspective. Curr Gastroenterol Rep 2017;19:14. doi:10.1007/s11894-017-0553-1.
3. Stirling J Jr. Beyond Munchausen syndrome by proxy: Identification and treatment of child abuse in a medical setting. Pediatrics 2007;119:1026-30. doi: 10.1542/peds.2007-0563.
4. Paturej A, Pogonowska M, Kalicki B. Munchausen syndrome by proxy: A case report. Pediatr Med Rodz 2019;15:93-6. doi: 10.15557/PiMR.2019.0017.
5. Wittkowski H, Hinze C, Häfner-Harms S, Oji V, Masjosthusmann K, Monninger M, et al. Munchausen by proxy syndrome mimicking systemic autoinflammatory disease: Case report and review of the literature. Pediatr Rheumatol Online J 2017;15:19. doi:10.1186/s12969-017-0152-6
6. Yates G, Bass C. The perpetrators of medical child abuse (Munchausen syndrome by proxy). A systematic review of 796 cases. Child Abuse Negl 2017;72:45-53. doi: 10.1016/j.chiabu.2017.07.008.
7. Rosenberg DA. Web of deceit: A literature review of Munchausen syndrome by proxy. Child Abuse Negl 1987;11:547-63. doi: 10.1016/0145-2134(87)90081-0.
8. Shaw RJ, Dayal S, Hartman JK, DeMaso DR. Factitious disorder by proxy: Pediatric condition falsification. Harv Rev Psychiatry 2008;16:215-24. doi: 10.1080/10673220802277870.
9. Royal Society of Chemisty. The Merck index online: An encyclopedia of chemicals, drugs, and biologicals [accessed 30 April 2020]. Available at: https://www.rsc.org/Merck-Index/
10. Stremski E, Hennes H. Accidental isopropanol ingestion in children. Pediatr Emerg Care 2000;16:238-40. doi: 10.1097/00006565-200008000-00005.
11. Slaughter RJ, Mason RW, Beasley DM, Vale JA, Schep LJ. Isopropanol poisoning. Clin Toxicol 2014;52:470-8. doi: 10.3109/15563650.2014.914527.
12. Faedda N, Baglioni V, Natalucci G, Ardizzone I, Camuffo M, Cerutti R, et al. Don't judge a book by its cover: Factitious disorder imposed on children-report on 2 cases. Front Pediatr 2018;6:110. doi: 10.3389/fped.2018.00110.

Síndrome de Munchausen por Procuração: Um Tipo de Abuso Raro, mas Real

Resumo:
Munchausen por procuração é uma forma rara de abuso, mas apresenta um elevado risco de morbidade e mortalidade. Um menino de 2 anos foi internado por estomatite aftosa e durante o internamento iniciou vómitos persistentes que foram extensamente investigados e tratados. Antes da alta planeada teve um episódio inexplicado de sonolência, petéquias e hematúria. A investigação revelou hemólise, hemoglobinúria, aumento da creatinina e cetonúria, sem acidose. Dias mais tarde teve um episódio noturno súbito com vómitos, miose, sudorese e ausência de reatividade. Manteve hemoglobinúria persistente que foi cuidadosamente investigada, sem diagnóstico etiológico. Devido ao início súbito e inexplicável dos sintomas, ausência de explicação etiológica e história social materna, suspeitou-se de síndrome de Munchausen por procuração por envenenamento. Houve resolução clínica na ausência da mãe e uma extensa triagem toxicológica de sangue, urina, vómito e alimentos, mostrou níveis tóxicos de isopropanol (desinfetante para as mãos). Não há casos descritos na literatura de intoxicação intencional com isopropanol e o diagnóstico de síndrome de Munchausen por procuração na primeira manifestação da síndrome é raro.

Palavras-Chave: 2-Propanol/envenenamento; Abuso Sexual na Infância; Pré-Escolar; Síndrome de Munchausen Causada por Terceiro/diagnóstico