# Fever of Unknown Origin for Six Years: Munchausen Syndrome by Proxy

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DR. PAMELA R. WOOD (*Assistant Professor of Pediatrics*): Munchausen syndrome by proxy is a puzzling and potentially life-threatening disorder in children. Cases as extreme as the one presented today are uncommon. Other examples along the spectrum of fabricated signs and symptoms are more common but often go unrecognized. As primary care physicians, it is our responsibility to be able to recognize and manage this disorder so that we do not contribute to ongoing abuse of such children.

## CASE PRESENTATION

DR. JOHN FOWLKES (*Senior Resident in Pediatrics*): A 7-year-old girl was admitted to the otorhinolaryngology service for placement of pressure equalization tubes. The patient had a chronically draining right ear despite seven months of treatment with many different antibiotics. An audiogram showed a 20-dB air-bone gap on the same side.

The patient was the product of a full-term pregnancy complicated by maternal preeclampsia. She was treated for multiple episodes of otitis media beginning at 4 months of age and required pressure equalization tubes at 14 months of age. The mother also reported admissions for bacterial meningitis and for pneumonia in the first year of life.

In the five years prior to the current admission, the patient was seen at least 126 times in different clinics and by a private physician and was treated for several disorders. At 1 year of age, she was admitted to the hospital for evaluation of fever. Soon after admission the patient began to have spiking fevers, with peaks occurring once or twice daily and subnormal temperatures in between. Cultures of blood, urine, stool, spinal fluid, and bone marrow were negative, and several sedimentation rates were normal. Findings of numerous diagnostic procedures, including cranial computerized tomography (CT), gallium scan, technetium bone scan, chest x-ray examination, oral cholecystogram, barium enema, echocardiogram, bone marrow aspirate, serum fungal serologic studies, febrile agglutinins, antistreptolysin O (ASO) titer, studies to evaluate humoral and cellular immunity, serum complement levels, and sweat chloride, were normal. The patient was discharged on aspirin for presumed systemic-onset juvenile rheumatoid arthritis.

Between 12 and 18 months of age, the patient did not grow well. Her mother reported daily fevers to 39 °C, diarrhea, frequent vomiting, and several episodes of hematemesis. The patient was usually afebrile when seen in the clinic. Stool guaiac tests were consistently negative, and two upper gastrointestinal series were normal. At 2 years of age, her growth rate returned to normal.

At 4 years of age, based on historical data supplied by the mother, the child was begun on medications for asthma and allergic rhinitis. By the age of 6 years, she was receiving five different asthma medications, including home nebulization treatments. Her diet was limited to ten foods based upon a history of food intolerance. Radioallergosorbent tests (RAST) to egg white, wheat, oat, and milk were elevated. Although the mother reported multiple emergency room visits for asthma, wheezing was never documented, and spirometry was normal. Repeated skin tests for inhalant allergens as well as multiple serum IgE levels were also normal.

Family history was positive for a maternal uncle who required weekly injections of gammaglobulin; for rheumatoid arthritis in the maternal grandmother, paternal grandmother, and maternal aunts; for an unknown intestinal disorder in the maternal grandfather; and for stomach cancer in the maternal grandmother.

The patient lived with her 30-year-old mother, a 12-year-old stepsister from her mother's prior marriage, her stepfather, and a 13-year-old stepbrother from her father's first marriage. Her stepfather was employed by the highway department, and her mother was completing training as an emergency medical technician (EMT). The patient

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was receiving home-bound instruction because frequent illness prevented her from attending school.

On physical examination, the patient was found to be a pleasant, healthy-appearing girl who was afebrile upon admission to the hospital. Her height was 110 cm (5th percentile for age) and her weight was 19 kg (25th percentile for age). The physical examination was normal except for the right ear, which contained granulation tissue and pus.

After placement of pressure equalization tubes, she developed a fever; mastoid x-ray films showed coalescent right mastoiditis. The patient underwent a right mastoidectomy. She continued to be febrile on intravenous cefazolin and gentamicin and subsequently underwent a left mastoidectomy. Cultures of blood, urine, mastoids, and middle ear secretions were negative. Culture of the left pressure equalization tube was positive for Pseudomonas aeruginosa, and the patient's antibiotics were changed to ceftazidime and tobramycin. Her fever persisted, and a retrograde cerebral venogram showed an area suggestive of right sigmoid sinus thrombosis. Subsequent exploration of the right sigmoid sinus revealed no clot. Postoperatively the patient was afebrile for almost 24 hours in the pediatric intensive care unit (PICU). After she was returned to the ward, she developed a fever of 39 °C orally and was transferred to the pediatric service for further evaluation.

Attempts were made to obtain reliable recordings of body and urine temperature. These efforts were hampered by the mother's reluctance to give up an active role in vital sign monitoring and medication administration. Two days after the patient's transfer, the mother reported that her daughter had a dilated left pupil. With the exception of left mydriasis, the child's physical examination was normal, as was a cranial CT scan. The anisocoria resolved over several hours, although the patient continued to have variable dilation of both pupils over the next two days. Nebulized atropine was one of the medications given by the mother. Despite persistent fever, the child was asymptomatic.

One week later, plans were made to discharge the child. The child then began to have severe diarrhea and vomiting and required intravenous fluid administration. After three days of persistent vomiting, a serum theophylline level was found to be 250 μmol/L (45 μg/mL; therapeutic range 55 to 110 μmol/L). The mother stated that she had not given the child any theophylline for three days.

The patient was immediately moved to the PICU. Children's Protective Services, legal counsel, the primary care physician, and the father were notified. An emergency protective custody order was obtained. The patient became afebrile upon transfer to the intensive care unit. The serum theophylline level gradually returned to normal, and the child's gastrointestinal symptoms resolved. Her theophylline elimination half-life was eight hours. Despite withdrawal of all asthma medications, she had no wheezing, and spirometry was normal. She was able to eat a regular diet without problems. She was discharged to the protective custody of the Department of Human Services and was subsequently placed in the custody of the maternal grandparents.

DR. WOOD: At the time of transfer from the otorhinolaryngology service, the ward physicians were suspicious about the nature of this child's illness, particularly the long-standing history of fever. Numerous attempts were made to obtain reliable temperature measurements so that a fever pattern could be documented. These efforts, however, were hampered by the mother's constant attendance at her daughter's bedside. Later, several nurses admitted having let the mother continue to take vital signs, especially temperatures. The mother had been allowed to dispense her daughter's asthma medications, keeping them at the bedside rather than on the medication cart. She stated that she was the only one who could convince her daughter to take the medications and agreed to keep a record of all medications given. She was unable to produce the charted information when asked.

## CLINICAL DISCUSSION

### Theophylline Toxicity

DR. DANIEL CASTO (*Clinical Assistant Professor in Pediatrics and Pharmacology*): The discovery that the child had a toxic serum level of theophylline, a medication being given by her mother, led to separation of the mother and child and an opportunity to obtain objective information about her illness. Laboratory error, nonlinear pharmacokinetics, coexistent medical conditions, and concurrent medications were considered as potential causes of an elevated serum theophylline level and were ruled out. Her elimination half-life of eight hours, although longer than would be anticipated for her age (average $t_{1/2}$ = 4 hours), was within the observed range for young children. More important, the half-life was not sufficiently long to result in the accumulation of theophylline to the measured toxic levels had the drug been administered according to the prescribed regimen. The toxic level, therefore, could be explained only on the basis of an inappropriate administration of the drug and was inconsistent with the mother's history that no theophylline had been given for three days.

### Medical Aspects

DR. WOOD: The patient, who reportedly had no temperature less than 39 °C for two weeks when measured by her mother, became afebrile immediately upon transfer to the PICU. She remained without symptoms of fever, asthma, or other allergic disease while in this unit.

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In 1977 Meadow[1] first described two children with a disorder that has become known as Munchausen syndrome by proxy. In this condition, one person (usually the mother) persistently fabricates symptoms and signs in another, thus causing that person to be regarded as ill. The average length of fabricated symptoms is about two years. Common signs and symptoms are fever, bleeding, neurological symptoms, and serum electrolyte abnormalities. One half of patients present with multisystem disease, as did this patient. Fifteen to 20 percent experience an episode of nonaccidental poisoning in addition to other health problems.[2] The disorder is a serious one, with a mortality rate of between 5 and 10 percent. The most common causes of death are suffocation and poisoning, and the greatest danger seems to be for very young children and infants.[2,3]

This patient and her family exhibited many of the so-called warning signs of this disorder: the patient's illness was prolonged and unusual, the signs and symptoms were often inappropriate or incongruous, and she had multiple "allergies."[4,5] Her mother fit the description of other mothers of children with Munchausen's syndrome by proxy.[5]

The patient's mother was intelligent, articulate, and overly attached to her daughter. As a result of her training as an emergency medical technician, she was familiar with medical terminology and hospital procedures. She was very comfortable with nursing personnel and seemed at ease in the inpatient setting. In contrast, the marital relationship seemed somewhat strained, and the patient's father was absent during the entire hospitalization.

The patient's illness was both prolonged and unusual. Approximately 9 percent of patients with greater than a one-year history of fever of unknown origin have factitious fever.[6] In the case of this patient, the long-standing history of fever without evidence of persistent active disease was highly suggestive of factitious fever. Her fever lacked the usual diurnal variation, and there was a striking discrepancy between observed skin temperature and recorded temperature readings. The child was usually afebrile when examined in the outpatient setting and when she was separated from her mother, such as for surgical procedures and postoperative care.

Other signs and symptoms also were incongruous or were present only in the mother's presence. Despite a six-year history of fever, severe asthma, middle ear disease, and multiple "allergies," the child looked well and had normal height and weight. Despite maternal reports of numerous emergency room visits for asthma that occurred when the family was "out of town," she was never wheezing when examined and never required hospitalization for asthma. Reported hospitalizations for bacterial meningitis and for pneumonia could not be documented.

Although a major portion of this child's illness was demonstrably factitious, there were significant obstacles to making the correct diagnosis. The first was the mother's affect and her apparent attentiveness to her daughter's care. It was difficult to question the behavior of a mother who appeared to be concerned and attentive, particularly since she did not seem to have any overt psychiatric disorder. Second, because of the vagaries of the alleged illnesses, physicians relied heavily upon historical information, which the mother was often able to amplify based on her medical expertise. Finally, it was difficult to distinguish between real and falsified illness. This child probably had true middle ear disease in addition to factitious illness.

The greatest obstacle to recognizing the true nature of this patient's illness was that her medical care involved many different health care providers who often failed to communicate with one another. Each provider focused upon only one aspect of this patient's illness and relied heavily upon historical information provided by the mother.

As primary care physicians, we must take an active role in the medical management of patients with complicated or undiagnosed conditions. We cannot allow their care to become fractionated among multiple subspecialists. The primary care physician is often in the best position to facilitate communication between health care providers and to recognize the warning signs of a disorder such as Munchausen syndrome by proxy. We must be suspicious when an illness is unusual, when there is a discrepancy between the child's apparent good health and reported signs and symptoms, or when the child's mother's behavior resembles that of the mother described in this case.

Appropriate management of children with suspected Munchausen's syndrome by proxy begins with separation of parent and child, so that signs and symptoms may be documented accurately. It is also important to check all details of previously recorded medical history by careful review of all medical records and other sources of information. As in this report, such a review will often reveal multiple inconsistencies and unsubstantiated claims. It is important to involve psychiatric consultants in the evaluation and management of such families. Once a diagnosis has been made, appropriate child protective agencies must be contacted, and the parent should be confronted in an honest, but noncondemnatory manner. The goal of management is to protect the child physically and psychologically and to restore an appropriate parent-child relationship.

## PSYCHIATRIC EVALUATION

DR. PATRICK HOLDEN (*Assistant Professor of Psychiatry*): During her hospitalization the patient usually

refused to talk with medical personnel and seemed unable to separate easily from her mother. Psychological testing showed that the child saw the world as a threatening place and felt unprotected and vulnerable, especially to parental figures, whom she perceived as unavailable. Under stress, she was likely to regress in her behavior to more immature levels of functioning and to use more somatization. The recommendations included individual psychotherapy.

The psychiatric evaluation and testing of the mother provided some clues to her bizarre behavior. The mother's early history was marked by abuse and neglect. She was born to alcoholic parents who fought openly in front of her and occasionally physically abused her. She was removed from the home at 5 years of age and placed in foster care, where she was sexually abused by one of the foster fathers. At the age of 6 years, she was adopted by a stable couple who became her psychological parents. At 18 years, she married her first husband, who physically abused her. After three years of marriage and one child, she divorced him and later married her present husband. Three months after the birth of her second child (the patient), she underwent gynecologic surgery, which left her sterile and feeling intermittently depressed. She devoted herself to the medical care of the patient. She also had concerns about her own health.

The initial psychiatric evaluation revealed a pleasant, cooperative woman who "couldn't understand why the doctors were suggesting that I would do those things." She was quietly persistent in her denial, explaining that the physicians' suppositions were due to a misunderstanding. She presented herself as reasonable and caring. As she talked about the patient's illness, however, she emphasized the dramatic aspects of the illness while maintaining an excited but unconcerned attitude. She spoke in broad sweeping terms about the illness but could not provide specific details when questioned.

At psychological testing, there was a striking discrepancy between her self-report and her unconscious thoughts and feelings. At a conscious level she described herself as optimistic, trusting, sensitive, at ease with others, and having no psychological conflicts or situational stresses. At an unconscious level there was evidence of early deprivation and humiliation and its sequelae. She was deeply resentful of parental figures and therefore mistrustful of adults in general. She was markedly insecure and fearful that others would criticize and derogate her. These limitations led her to utilize functional complaints and dramatization of her daughter's symptoms to get her needs for nurturance met.

There was no evidence of a formal thought disorder, a marked mood disturbance, or Munchausen syndrome in the mother herself. Diagnostically, it was felt that she had a somatization disorder.

She was treated in individual psychotherapy for seven months. Although she angrily denied responsibility for fabricated symptoms and signs, she gradually developed some appreciation for the connections between her early experiences and her present life circumstances. Repeat psychological testing at termination of therapy revealed modest improvement, particularly in her ability to resist her impulses.

## PSYCHOPATHOLOGY OF MUNCHAUSEN BY PROXY

The mother's psychosocial history is similar to previous reports describing abuse as a child, disturbed family relationships, deaths or other losses, a distant relationship with the husband, and enmeshment-overattachment with the identified patient.[3,7–9] These mothers often have recurrent functional complaints or documented medical problems including obstetrical complications or infertility.[3,7,9] Some have a previously diagnosed psychiatric disorder.[7–9]

Why did this mother fabricate signs and symptoms and then poison her child? Few of these mothers acknowledge their behavior even when confronted.[5,9–11] In most cases, the underlying motivation for this behavior must be inferred from the information available about the mother and her circumstances. The motivation is usually an attempt to (1) meet needs for affection, admiration, or attention, (2) combat serious underlying maternal depression, (3) deal with relationship problems, or (4) compensate for a seriously impaired identity.[5,7,10–12] Psychological testing suggested that this mother was unable to get her needs for nurturance and affection met through the usual means. She discovered that health care professionals were readily available and could unwittingly meet these needs. Furthermore, she did not see her daughter as a distinct and separate individual but rather as an extension of herself and as a means of getting her own needs met.

## MANAGEMENT OF MUNCHAUSEN SYNDROME BY PROXY

There has been some controversy about the value of psychiatric consultation for these patients. Most authors believe that child psychiatrists, especially those with experience with child abuse, can provide substantial help. It is recommended that the child psychiatrist be involved prior to confrontation to provide immediate psychiatric attention for those mothers who require it.[5,10,11]

Since these mothers can be quite convincing in their denial of responsibility, it is essential that the primary

care physician educate other physicians involved in the patient's care. The Children's Protective Services Department and the courts also need education about this disorder.[5,8,9] Specifically, these agencies should be made aware of the marked discrepancy between the mother's public attitude and her behavior in private with the child. In addition, the courts need to be aware of the potential for further harm to the child and the mother's resistance to voluntary psychiatric treatment.[7,9]

The long-term prognosis of children who are treated for Munchausen syndrome by proxy is unknown. Most authors recommend extended psychiatric treatment for children who have been severely abused.[7] If the disorder goes unrecognized, children often begin to participate in fabrication of illness and may eventually develop Munchausen syndrome themselves.[2]

## FOLLOW-UP

DR. WOOD: Following six months of court-mandated residence in the maternal grandparents' home, the child was returned to her own home. One year later, she is completely well, taking no medications, and is regularly attending school.

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