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CASE REPORT

Caustic esophagitis in two siblings: Munchausen syndrome by proxy

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Abstract:
Background and aim: *Recurrent corrosive esophagitis is a rare condition among children and may be due to child abuse. The Munchausen Syndrome by Proxy (MSBP) is a special form of child abuse with variety of symptoms. It has challenging diagnostic process and needs a high clinical suspicion. When it is identified by physician, institutional child protection team must involve into the management of the case.*
Case report: *We present the two siblings with recurrent esopahigitis. One of them died due to severe gastrointestinal bleeding. Other one had severe esophageal stricture and oral lesions. Their mother fed both of them with caustic material at various time intervals. Both babies were treated in our hospital several times with the admission of their mother.*
Conclusion: *This is probably the first case report of Munchausen Syndrome by Proxy (MSBP) with caustic material ingestion of two siblings. Determining the exact cause of such cases is very difficult and physician must suspect child abuse in this situation.*

Key words*: reccurent caustic esophagitis, child abuse, Munchausen syndrome*
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Introduction

The Munchausen Syndrome by Proxy (MSBP) is a type of child abuse. The caregiver, mostly the mother with unmet psychological needs and any particular motivation exaggerates or fabricates symptoms and even induces signs of a nonexistent pediatric illness [1,2]. The child experiences unnecessary hospitalizations, diagnostic tests, medical and even surgical interventions. The physician has a significant difficulty in the exact diagnosis of the pathology and the child may receive an unnecessary and potentially harmful medical care [3].

In the current study, we represent two siblings with child abuse from their own mother.

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Case Reports

The mother fed both children with caustic material. She had led to the death of first child and caused a severe morbidity of the second. Both children were admitted to the hospital and they were treated several times. The mother seemed to be as worrying about the condition of her children. Until the last admission of her second child, we did not suspect and were not able to understand this pathological situation.

Case 1

A 6-day-old boy, the first live birth of an 18-year-old mother, was admitted to the pediatric

emergency room with dyspnea and hematemesis. His poor general condition and respiratory distress necessitated intubation and he was hospitalized in the Newborn Intensive Care Unit.

The diagnosis was neonatal sepsis and gastrointestinal bleeding. He responded to the palliative treatment and was extubated within 4 days. Esophagograms showed gastroesophageal reflux, stasis and decreased gastric capacity. Oral nourishment was started with poor swallowing. Esophagoscopic evaluation revealed two segments with strictures and a balloon dilatation was performed. The following esophagoscopy revealed eroded distal esophageal mucosa, multiple polipoid structures and ulcers within the gastric lumen. It was resembled the Zollinger-Ellison Syndrome but gastrin level was within normal limits [20 pg/ml (25-125)].

Laparoscopy was performed and there were many adhesions between the stomach and the abdominal wall. This finding aroused suspicion about an intrauterine or early perinatal intestinal perforation. Micro-stomach and near-total obstruction necessitated total gastrectomy, esophago-jejunostomy and jejunojejunostomy (Brown procedure). He was discharged at postoperative 15 days.

One week later, he was admitted to the emergency room and hospitalized again with the diagnosis of malnutrition, bronchopneumonia and urinary infection. Following the proper 7-day antibiotic treatment he was discharged again.

One day after discharge, He had hematemesis again with a significant anemia. He was hospitalized in the Intensive Care Unit. An explorative laparotomy, bilateral vagatomy and marginal ulcer resection was performed eventually. These interventions could not reverse the unpromising course and he died the next day of the last operation.

Case 2

After a year, a 25- day- old male baby of the same family who was the sibling of case 1 was admitted to the pediatric emergency room with symptoms of cyanosis and vomiting after meals. The pediatric gastroenterology department followed the patient with the diagnosis of gastroesophageal reflux.

He was admitted again to the emergency room with brownish vomiting when he was at 2 months of age. He had poor general condition and insufficient oral intake.

On follow up, his Hb level decreased down to 6,5 g/dl. Bone marrow sampling and analysis revealed no malignant cells but rise of eosinophils and myelocytic series. Erythrocyte suspension was given. His condition was thought to be related to antibiotics. Immunology consultation revealed immune deficiency. His axial hypotonicity was evaluated by the neurology department and follow-up was suggested. When the Hb level reached 13 g/dL, CRP decreased to 6 mg/L and his general condition improved, the antibiotic treatment was stopped and he was discharged.

One month later, when he was nearly 5-months-old, he was admitted to the emergency room following the sudden onset of vomiting and dyspnea about an hour after a meal. His mother explained feeding him with a bottle of infant formula.

His general condition was poor with dyspnea secondary to aspiration. Drooling and oropharyngeal desquamation and white plaques were present. These findings were associated with vomiting that resembles to a caustic agent pouring out of his mouth (Figure 1).

He had hematemesis and melena on the day of admission. He was consulted with plastic and reconstructive surgery department for the scalded facial skin and a daily debridment was recommended. Total parenteral nutrition was started. Afterwards salivation regressed.

Upper GI series taken after 3 weeks, revealed no esophageal stricture nor reflux but stasis at the distal esophagus suggested an impaired motility. Oral feeding was started. He could not suck the feeding bottle at the first days due to his oral lesions. Within a few days his oral intake improved but he started vomiting several times a day. Following a two-week clinical observation period, we repeated the oral contrast study which

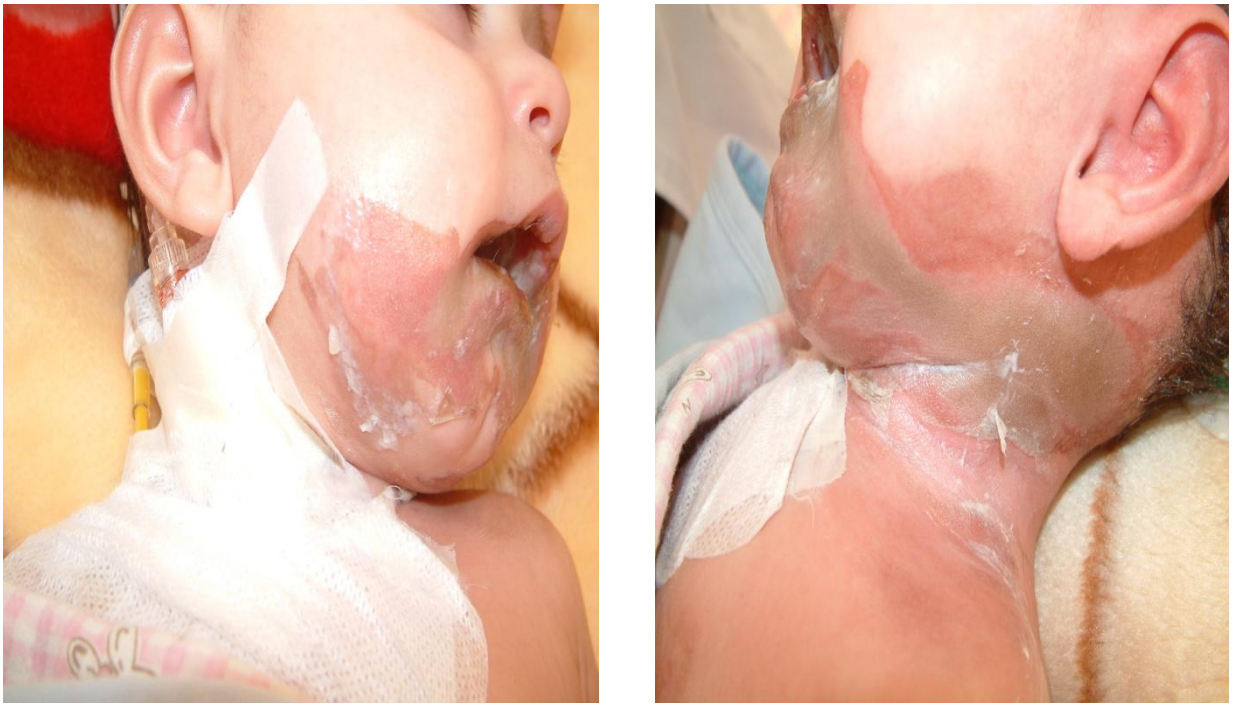

Figure 1. The perioral and neck lesions caused by caustic substance

showed no difference. Finally an endoscopy was performed. There was a stricture at the most proximal part of esophagus and hypopharynx. Stricture was dilated by balloon dilatation. Patient had started oral intake after 10 days of dilatation. Because of the perioral stretching lesions, perioral stent was placed and the patient was included into the regular dilatation program.

Meanwhile our instutional interdisciplinary child protection team investigated both cases and interviewed with all family members. They suspected from the mother of these babies for child abuse. The possible mechanism of the child abuse was an intentional feeding of the both babies with corrosive substances. The case was evaluated both psychiatrically and officially. The mother was accused of voluntary administration of caustic substance to her own children and leading to permanent physical damage and death.

Discussion

Caustic esophagitis in children is often caused by accidental ingestion. The appearance of mucosa and the final outcome depends on the quantity, duration and the nature of the ingested substance [4,5].

Strong alkaline compounds cause the most serious lesions and are responsible for the majority of deaths [6]. Massive ingestion causes diffuse esophageal necrosis.

In this report, the substance ingested by the second sibling was most probably acidic in nature although pH of the given formula was unclear.

Recurrent caustic esophagitis is rather rare and may indicate a child abuse. The diagnosis of MSBP is frequently overlooked [2, 3]. In our cases, very unusual stricture formation of esophagus and stomach in two siblings led us to consider a case of child abuse of a mother with MSBP.

In MSBP, death occurs most often in children aged under 6 years, and it is mainly caused by intoxication often due to overdose [7,8]. It is often difficult to accept the idea of a parent causing harm in his/her own child on purpose and also it is difficult for a clinician to prove this syndrome. Some papers have suggested the use of video-surveillance in hospital environment for confirmation of the suspected cases [9,10].

Understanding the motivation of the abusing caregiver is important. Every health center should have a multi-disciplinary child protective team where the primary clinician should consult. Once diagnosed, clinician has to report the case to legal authorities in order to protect the child before any irreversible damage occurs.

In conclusion, diagnosis of such a case might be a nightmare for the clinician in both medical and social aspects. Institutional child protection teams, special police department, courts specialized in children and social workers may help in management of this challenging problem.

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