

CASE REPORT

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Munchausen syndrome by proxy and intra-alveolar haemosiderin

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Abstract Munchausen syndrome by proxy is characterised by the invention of a false history and/or the deliberate inducement of a factitious illness by parents in their child. First reported as a disorder of mothers, this syndrome is now recognised to have male perpetrators. One of the most common characteristic presentations is with the child allegedly suffering repeated apnoeic attacks. These are in fact deliberately induced episodes of upper airway obstruction. The children of these parents are at great risk of serious harm or death if not recognised. The identification of smothering at autopsy is fraught with difficulty and the presence of intra-alveolar haemosiderin has been claimed to be a marker of previous smothering. A case of Munchausen syndrome by proxy is presented where there were deliberate acts of repeated partial smothering and where the finding of intra-alveolar haemosiderin at autopsy provided additional supportive evidence of smothering.

Key words Factitious illness · Autopsy · Haemosiderin · Lungs

Introduction

In 1977 Professor Sir Roy Meadow reported the cases of two children in whom their mothers had deliberately induced factitious illnesses [1]. He labelled this disorder Munchausen syndrome by proxy and subtitled his paper the hinterland of child abuse. This subtitle was apt as these cases remain difficult to identify with the victims at great risk of harm and cases still end in the death of the child. A characteristic presentation involves the child's mother inducing apnoea by deliberate upper airway obstruction, the child presenting to hospital with so-called

apparent life threatening events (ALTEs), although other manifestations such as fictitious epilepsy or poisoning also occur [2–17]. The identification of these cases is fraught with difficulty and the findings at postmortem examination may be minimal or absent.

The presence of haemosiderin-laden macrophages (siderophages) in the lungs of young children has recently been proposed as a marker of deliberate upper airway obstruction [18]. This report presents a case of a child with multiple admissions for so-called ALTEs. The case had many features associated with Munchausen syndrome by proxy. Haemosiderin-laden macrophages were an important finding and supported the contention that the episodes of ALTE were deliberate episodes of smothering. Unusually the perpetrator was the father of the child.

Case history

A 21-month-old white female child was found collapsed in her cot by her father at approximately 0430 hours. The paramedics attended and found the child in cardio-respiratory arrest. Cardio-pulmonary resuscitation was started by the paramedics who were able to ventilate the lungs. The girl was taken to the local Accident and Emergency Department, where a balloon was found in the oropharynx. Despite vigorous attempts at resuscitation no cardiac output could be achieved and the child was certified dead.

The child was born at full term by vaginal delivery without complications. She had multiple attendances at hospital with inappropriate or inadequate explanations starting at the age of 14 days. Many were for trivial injuries or ailments that would not be expected to cause significant concern or warrant attendance at hospital. Furthermore the father often requested that inappropriate investigations be performed on his child, such as endoscopy. In addition there were five attendances when the girl was alleged to have choked on food or stopped breathing. The first episode was when she was 14 days old and was said to have choked on food. She re-attended aged 4 months when she again was said to have choked on food. She had three apparent episodes of stopping breathing, two at the age of 7 months and one at 10 months. Bleeding from the mouth and nose was reported to have occurred in some of these episodes. The last admission was 12 days before death when she was said to have been found in her cot with noisy breathing and the father alleged she had swallowed a lapel badge. He had tried to recover it but was unsuccessful. He stated she stopped breathing at one point for 20 s. He gave her mouth to

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mouth resuscitation. An examination in hospital revealed no injury or badge. Whilst the girl was awaiting radiological examination the father was observed to place his fingers in the child's mouth, an act he was told to stop, whereupon he produced a badge and said he had just retrieved it from his daughter's mouth. Subsequent examination revealed a small laceration on the mucosal surface of the lip.

On each occasion when the child had breathing problems the father said he carried out resuscitation, but not on the last occasion when she died. On each admission the child was fully investigated but no abnormality was found, other than radiological changes consistent with inhalational pneumonia on one early admission.

Postmortem examination revealed a well nourished white female child. No injuries were identified. No "classical" signs of asphyxia such as petechiae were present. Microscopic examination of the lungs revealed the presence of focal intra-alveolar haemorrhage, with easily identifiable haemosiderin-laden macrophages in the alveoli seen on Perls stained sections of lung tissue. These siderophages were seen in every block of lung and were predominantly peripherally located. No interstitial haemosiderin was present. The cause of death could not be fully ascertained but the presence of intra-alveolar haemosiderin-laden macrophages was felt to be supporting evidence that previous episodes of ALTE were in fact induced upper airway obstruction.

Subsequent forensic examination revealed semen on oral swabs and the balloon, which was DNA typed to her father. An examination of two other balloons subsequently retrieved from the home also revealed the presence of the father's semen.

Overall the findings and history strongly supported the opinion that this child had been subjected to multiple episodes of deliberate partial smothering.

Whilst the investigations were proceeding the father publicised his daughter's death through the national media as an accident, warning other parents about the dangers of children playing with balloons.

An examination of the father's medical records revealed multiple attendances at hospital, again many being for trivial matters. In addition the father had made unsubstantiated reports of burglary at his home and one admission to hospital was for smoke inhalation following a fire. He had been abusive to ambulance staff and to hospital staff. After the death, he was also abusive to staff in the coroner's office and to mortuary staff.

Although a diagnosis of Munchausen syndrome by proxy was being considered clinically, a final decision on diagnosis and management had not been made at the time of the child's death.

The father was subsequently charged with the murder of the child on the basis that death was not an accident due to choking on the balloon but a deliberate act of upper airway obstruction, against a background of repeated smothering. He was also charged with one count of child cruelty in respect of the alleged episode of the girl swallowing the badge.

The prosecution at the trial presented the case as one of repeated deliberate acts of upper airway obstruction and child cruelty. The semen on the balloons, it was stated, was evidence that the defendant was a sexual deviant with a balloon fetish.

At the criminal trial the father's defence was that death was an accident. No psychiatric evidence was presented by the defence and the father did not admit to harming his daughter. He was convicted of manslaughter and sentenced to 10 years imprisonment with a concurrent sentence of 5 years for child cruelty.

Discussion

In 1977 Meadow published his seminal work on Munchausen syndrome by proxy [1]. Since the original report, further reports have appeared characterising this syndrome, which is nearly always perpetrated by the mother [2–17]. Characteristically the child presents with a factitious disorder, the symptoms and signs of which diminish when separated from the perpetrator. The disorder is often

reported as an apnoea attack, shaking, fitting or similar symptomatology. It is not always clear whether the reported incidents are deliberate acts by the parent, or a deliberately invented false history, or both. Resuscitation is often performed and described by the perpetrator. Other forms of abuse include the deliberate administration of salt, or the inducement of bleeding. In one of the first cases described by Meadow, the mother tampered with urine samples to make it appear that her daughter had urinary tract infections.

In 1998, Meadow published a series of 15 cases in which the father was the inflictor of abuse [17]. This case shows many similar features. In his series of 15 cases, 10 of the victims were boys and 5 girls and the ages varied from new-born to 9 years. The commonest presentation was with the child allegedly suffering recurrent apnoea attacks and being resuscitated by the father. In 9 out of the 15 cases the final diagnosis was smothering, in 2 cases the children were repetitively poisoned and in three cases there was no direct evidence of harm, but the children underwent unnecessary investigations because of the father's false stories. Two children had evidence of physical abuse. In two cases the father was seen to smother their child by hospital staff and two were identified via covert video surveillance. Three admitted injuring their child during criminal investigations. Of the 15 index children, 3 died and there were a further 8 deaths of siblings. Of the fathers, five were diagnosed as having Munchausen syndrome and five had a somatising disorder. In this case the repeated attendances at hospital certainly raised the possibility that the father had a somatising disorder if not Munchausen syndrome. Meadow described the men as memorable because of the stories they told. They were frequent fantasists who reported remarkable achievements. They often reported factitious medical illnesses, sometimes bizarre. In four cases strange fires had occurred in the homes of the men, who claimed they were caused by strangers or neighbours, but were believed by the authorities to have been caused by the fathers themselves. Many reported robberies and burglaries. In four homes the factitious illness was reported in pets.

The fathers' relationships with hospital staff were often poor. They were characteristically described as overbearing, overdemanding and unreasonable. The father in this case had similar problems with hospital staff.

Overall male perpetrators of Munchausen syndrome by proxy are rare. An analysis of records of Munchausen syndrome by proxy in the United Kingdom and Ireland between 1992–1994 identified 128 cases [19]. In six cases the perpetrator was male, with only one case characterised by the inducement of factitious illness, the others being poisoning or smothering not associated with a repetitive or persistent factitious illness.

The identification of deliberate repetitive partial smothering of a child is difficult. Sometimes an act of smothering may be observed whilst the child is on the ward, but more sophisticated investigation may be required. Covert video surveillance (CVS) has been employed as a method of detection. [4, 6, 8, 10, 12, 17, 20–

22]. An important observation of Southall and his colleagues in respect of this case was that 11 out of 38 children they surveyed had bleeding from the nose and/or mouth [22].

Intra-alveolar haemorrhage has been reported as a common finding in deaths due to compression of the neck and in "sudden infant death syndrome" (SIDS) [23, 24]. Intra-alveolar haemosiderin-laden macrophages have been reported in the lungs of drug addicts, where previous hypoxia and resultant haemorrhage have been proposed as the mechanism of production [25]. An analysis of infant deaths from Sheffield has shown significant intra-alveolar haemorrhage in cases where death was due to "overlaying" and in cases where there was a strong suspicion of smothering [26]. These reports support the hypothesis that previous hypoxia, however induced, causes intra-alveolar haemorrhage, with haemosiderin produced as a repair mechanism. The presence of intra-alveolar haemosiderin therefore, raises the possibility of previous episodes of hypoxia in an infant. Becroft and Lockett [18] identified haemosiderin-laden macrophages in two pairs of siblings who had previous admission for ALTEs and in whom subsequent criminal proceedings resulted in convictions of murder and manslaughter, respectively. In one child with no history of ALTEs, but who died in the care of the mother of one pair of siblings who had admissions for ALTEs, no haemosiderin was found. In three children who had been cared for by this woman, and who survived but had episodes of an ALTE in her care, bleeding from the nose or mouth was observed, as it was during 6 out of 10 episodes of ALTEs in the fatal cases.

An examination of 143 infants who had died and been classified as SIDS revealed intra-alveolar haemosiderin in 7 cases [18]. In four of these cases, the infants had been treated in a neonatal intensive care unit, and two had been ventilated. None of these children had a recorded history of ALTEs. Fifteen of these infants did give a history of ALTEs, with only one case having intra-alveolar haemosiderin. Interstitial, as opposed to intra-alveolar haemorrhage has been reported as a common finding in cases classified as SIDS, although a second study found it less frequently, but did find it more frequently in SIDS cases with a history of ALTEs [27, 28]. In the four index cases of Becroft and Lockett, no interstitial haemosiderin was seen, nor was it identified in this case [18]. Becroft and Lockett suggested that the pathogenesis of interstitial haemosiderin may be different from intra-alveolar haemosiderin. Natural disease processes such as cardiac disease and bleeding disorders need to be excluded. Syndromes such as Goodpasture's and idiopathic pulmonary haemosiderosis have characteristic features, are rare, and should be able to be discounted [29]. Haemosiderin is easily identified in Perls stained slides, but is equally easily missed in haematoxylin and eosin stained sections. It is now our practice to examine the lungs of every child for stainable iron.

It is now well recognised that a proportion of SIDS deaths may be homicides. [30, 31] The percentage of SIDS that are homicides may be disputed, and may de-

pend on local factors and campaigns to prevent cot deaths, but undoubtedly occur, but are not recognised. Cases previously described as SIDS have been reviewed and declared to be homicides, some cases resulting in convictions [32]. A report describing a series of cases presented as prolonged apnoea followed by death, linked apnoea attacks to SIDS. It has been suggested that in reality these cases may well have been Munchausen syndrome by proxy [33, 34].

The investigation of deaths in early childhood where abuse is suspected are difficult as signs may be minimal or absent. Where a child has previous admissions for ALTEs, the possibility that these episodes are part of a factitious illness must be strongly considered. The absence of "classical" signs of asphyxia is more common than not. The finding of haemosiderin-laden macrophages in the alveoli of these children requires an explanation and provides supportive evidence of upper airway obstruction, but is not diagnostic. A history of later onset ALTEs and bleeding from the mouth or nose should raise the suspicion of deliberately imposed upper airway obstruction. The combination of background history, medical records and postmortem investigations will often identify deliberate abuse. A re-examination of any deaths of siblings should be performed. Failure to identify cases of such subtle abuse may jeopardise the safety of siblings and future children.

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