Two infant deaths linked to intussusception without peritonitis

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Abstract

We report two infant deaths attributable to intussusception, but without clear evidence of peritonitis. In the first instance, a 3-year-old girl had presented with abdominal pain, vomiting, and melena before her demise. Aspirated vomitus was subsequently ascertained as the immediate cause of death, due to intussusception-induced ileus. The other infant, a 2-month-old male, showed autopsy evidence of intussusception at two sites, with likely aspiration of gastric mucus. Since the circumstances surrounding his death were vague, timing of the intussusception was difficult to pinpoint. Thus, an inconsequential, agonal event could not be discounted. Taken together, however, death from intussusception, without peritonitis, is the most viable postmortem interpretation for both patients. The causes of death in such cases are established by comprehensive delineation of preceding clinical events, plus autopsy documentation of coexistent intussusception and vomitus aspiration.

Keywords: forensic pathology, intussusception, sudden infant death

Introduction

Infant intussusception is typically associated with abdominal pain, vomiting, and melena. It rarely leads to death, if treatment is initiated early [1]. Death may, however, ensue when treatment delay allows peritonitis to develop. Sometimes intussusception results from specific organic pathology, especially the enlarged Peyer's patches of viral infection [2,3] and surgery [4]. It also occurs idiopathically [5]. In general, it is viewed as a pediatric affliction; and while newborns are occasionally affected [5, 7, 8], the highest incidence is between the ages of 4 months and 1 year [6].

Patients with prolonged intussusception usually develop peritonitis. Autopsy proof of both events is therefore needed for pronouncement of death from aggravated intussusception. On the other hand, death prior to the onset of peritonitis is also feasible, given that frequent vomiting, secondary to ileus, may lead to aspiration and suffocation. Under these circumstances, it may be impossible to demonstrate peritonitis, even with confirmed intussusception. At least one published report [1] has suggested that intussusception, in some instances, may represent an agonal development, rather than a premortem disorder. It would then appear that a finding of peritonitis is critical, in order to definitively rule on the timing of intussusception.

We encountered two cases of infant intussusception, without signs of peritonitis. One of the patients presumably succumbed from aspiration of vomitus, triggered by intussusception before death. For the other patient, it was difficult to state whether the intussusception was premortem or agonal. The circumstances surrounding his death were unclear. Key points in differentiating the timing of intussusception are presented in this article, based on our experience with these two cases.

Case 1

The patient, a 3-year-old girl, initially presented with vomiting, but did not have abdominal pain. By evening of the following day, she finally complained of abdominal distress, and around midnight, her mother saw she vomited and aspirated vomitus. An ambulance was called, but before reaching a hospital, she underwent cardiopulmonary arrest. When she was intubated with endotracheal tube, food and debris flew out of the tube. Her rectal temperature on arrival was 41.3°C, and melena was noted in her diaper. Laboratory testing indicated a C-reactive protein (CRP) of less than 0.3 mg/dl, hypernatremia of 220mEq/l, and a leukocyte count of 14400/m³.

An autopsy was performed approximately two days postmortem. The child's body weight was 14.5 kg and her height was 94.5 cm. Ileoileal intussusception was found at one point, with intestinal swelling at the proximal, rather than distal, margin of the lesion. The length of the intussusception was 1.5 cm. Her airway was filled with light brown, viscous liquid, assumed to be gastric contents. There were no gross autopsy findings of peritonitis, such as intestinal or peritoneal discoloration and hyperemia, and no histologic evidence of peritonitis could be found. Specifically, inflammatory cell infiltration and necrosis were absent in the vicinity of the intussusception (Figure 1). There was no clot in the heart and fluidity of the cardiac blood was found. Petechial hemorrhages were found in the pericardium and were not in the conjunctiva. These findings indicated the possibility of asphyxia. Histology revealed congestion and oedema in lungs and no evident inflammatory disease in lungs, heart, kidneys, pancreas, liver and brain.

Case 2

This patient was a 2-month-old male infant left in the care of his father, while his mother, the usual caregiver, attended a wedding one day in winter. The father fed the baby 80 cc of milk around 11:00 AM, 2:30 PM, and 5:00 PM, for a total of 240 cc. He then drove to another location to socialize and exited the vehicle, leaving the infant on the back seat at around 5:00 PM. Upon returning to the car at 8:00 PM, the father noticed that the baby was not breathing. The infant was transported to the hospital, where his death was confirmed at 10:00 PM. The rectal temperature was 31.5 °C at 12:23 AM.

An autopsy was performed about one and one-half days postmortem. The infant weighed 6220 g and was 63 cm long. No trauma was found to suggest that the baby had been abused. Light brown, viscous liquid, not inconsistent with gastric contents, was observed

in the airway. Ileoileal intussusception was also visible at two distinct sites, and again, intestinal swelling involved the proximal, rather than distal, margins. Both the intestinal tract and peritoneum were devoid of discoloration or hyperemia, and there was no histologic evidence of peritonitis (Figure 2). Postmortem blood work returned CRP of less than 0.3 mg/dl. The ratio of carboxyhemoglobin to hemoglobin molecules was 1.5% in the cardiac blood,

Discussion

In Case 1, hypernatremia indicated that the patient was dehydrated due to vomiting. The high body temperature might be due to dehydration or inflammation in the early phase of either colitis or aspiration pneumonia, which did not cause the positive CRP. She presumably died without developing peritonitis. There was no gross or microscopic evidence of peritonitis at autopsy, and the CRP was negative. Furthermore, the fact that only several hours had elapsed from the time of intussusception until death implies that mortality from intussusception may not necessarily involve peritonitis—at least as far as autopsy findings permit. Demonstrable intussusception with aspiration of vomitus is hallmarks of the diagnosis.

This patient also experienced melena, vomiting, and abdominal pain in advance of death, so it was felt that her symptoms logically signaled a premortem occurrence of intussusception. An accurate assessment of such deaths, therefore, depends on reliable accounts of the patient's preceding course and symptoms, in conjunction with complete autopsy findings. The immediate cause of death in Case 1, however, was cited as suffocation from aspiration of vomitus, all related to intussusception with ileus.

Case 2 was marked by dual sites of intussusception. Interestingly, it has been reported that intussusception developing in the agonal state can, at times, be multifocal [1], and repeated bouts of intussusception during life have been described [5, 9]. For this particular infant, it was especially difficult to judge whether intussusception occurred as a premortem or an agonal event. Not only was autopsy evidence of peritonitis lacking, but also the infant's condition before death was not fully known. Still, the presence of probable gastric mucus in the airway was sufficient to support aspiration as the cause of death, similar to the Case 1 scenario.

Proof of patient death due to intussusception, before the onset of peritonitis, is dependent on the following: 1) confirmation of vomitus in the airway, and 2) compatible events and symptoms prior to death. When both requirements are satisfied, as in Case 1, the diagnosis can be made reliably. Unfortunately, when historical data is limited, as with Case 2, one is hard-pressed to differentiate premortem from agonal intussusception. A more rigorous investigation by authorities must then be conducted and integrated with autopsy results, so that cause of death can be better assessed. While intestinal dilatation, proximal to the intussusception, and intra-airway vomitus may serve as presumptive evidence, their validity in defining death from intussusception will require

added corroboration through case studies going forward. Likewise, the very concept of agonal intussusception merits closer scrutiny.

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Figure Legends

Fig.1

Case 1 - Gross and microscopic autopsy findings

A: Intestinal intussusception observable in abdomen, without visible peritonitis.

B: Airway filled with light brown mucus, likely aspirated gastric contents.

C. Inwardly telescoping intestinal segment of intussusception, manually returned to natural state and routinely stained. (Hematoxylin-Eosin 20X)

No peritoneal inflammation.

D: Outer intestinal segment of intussusception. (Hematoxylin-Eosin 20X)

No peritoneal inflammation.

Fig.2

Case 2 - Gross and microscopic autopsy findings

A: Intestinal intussusceptions (2) observable in abdomen, without visible peritonitis.

B: Light brown viscous liquid in bronchi bilaterally.

C: Proximal-most margin of intussusceptions, sampled and routinely stained (Hematoxylin-Eosin 20X). No peritoneal inflammation.

D: Distal-most margin of intussusceptions, sampled and routinely stained (Hematoxylin-Eosin 20X). No peritoneal inflammation.

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Figure(%)

Figure(2)