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CASE REPORT

Multiple Intussusceptions as the Cause of Death in Asymptomatic neonate: A rare case report

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Abstract:

Intussusception, a major cause of intestinal obstruction in childhood. The most common type is ileocolic, followed by ileo-ileal. Intussusception has a peak incidence between three to nine months (Pollack and Pender, 1991) and has a male predominance. Autopsy proof of both intussusception and peritonitis is needed for pronouncement of death from aggravated intussusception. Frequent vomiting, secondary to ileus, may lead to aspiration and suffocation when it may be impossible to demonstrate peritonitis, even with confirmed intussusception. If no pathological lead point, intussusception is likely to represent a primary rather than secondary phenomenon. The cause of intussusception is not known in most of the cases. Several association and possible causative factors have been identified like large peyer's patches, polyps, Meckel diverticula, cystic fibrosis, lymphoma, viral etiology particularly adenovirus, recent history of upper respiratory illness and heredity. It is important to clearly distinguish agonal intussusception from antemortem intussusception. Agonal intussusceptions are believed to occur terminally are an incidental finding, not a cause of death although designation of the intussusception as 'agonal' cannot be justified by some authors. We report a case of a 7 days old male child with history of not accepting feed for 1 day who was brought dead. Five ileo-ileal intussusceptions were evident grossly at autopsy and histopathological examination was diagnostic of cause of death. Multiple antemortem intussusception as cause of death is fairly uncommon and no ample literature was available for reference and hence reporting the interesting case.

Keywords: Sudden death; Infant; Intussusception.

Introduction:

Intussusception is the most common cause of intestinal obstruction in children younger than 2 years of age.[1,2] Although intussusception in childhood is usually a relatively benign entity, which is transient and resolve spontaneously, it may rarely be associated with unexpected death.[3] The most common type is ileocolic, followed by ileo-ileal. The cause of intussusception is idiopathic in most of the cases. Several association and possible causative factors as large Peyer patches, polyps, Meckel diverticula, cystic fibrosis, lymphoma, rotavirus vaccination, antibiotic use, recent history of upper respiratory tract illness, viral aetiology particularly adenovirus and heredity are identified.[1] Unexpected death may occur at any age, with intussusception at any level with no pathological lead point. It has been estimated that 13–20% of children with intussusception have so called 'painless intussusception' with no discomfort or colic.[4] Autopsy findings in sudden deaths in which no other gross abnormality other than intussusception are evident, it was difficult to ascertain if death is due to sequelae of intussusception alone or it is a postmortem agonal artefact.[2]

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The Case:

A 7-day old infant was found gasping during the morning hours and was declared brought dead in Trauma and Emergency in a tertiary level hospital of Central India with alleged history of lethargy and not taking breastfeeding in the night-time. Previous history of fever of two-day duration following vaccination was also present. The antenatal and postnatal period of the mother was uneventful. Since the death was out of the healthcare institution and was not correlating to any obvious pathology or injury a medicolegal autopsy was conducted to ascertain the cause of death. The weight of the neonate to be 2.1kg and length of 48cm. There were no external injuries to the neonate. All orifices were intact and devoid of any injury or pathology.

The remnants of the umbilical cord had fallen off with a healing umbilical area. No pus or pathology were evident at the umbilical area. On internal examination, the brain was 381gm in weight and was oedematous and congested on cut. All vital organs including both lungs, heart, liver and both kidneys were showing normal gross findings. Stomach contained 5 ml greenish brown mucoid fluid with no peculiar smell or congestion. On further exploration of intestinal loops, five ileo-ileal intussusceptions were evident grossly (Photo 1 and 2). At the level of intussusception, the proximal bowel segments were dilated, edematous & distal were dark reddish to dusky and collapsed, not easily reducible. There were no other gross significant findings in other organ systems. The affected portion was preserved in formalin for histopathological examination.

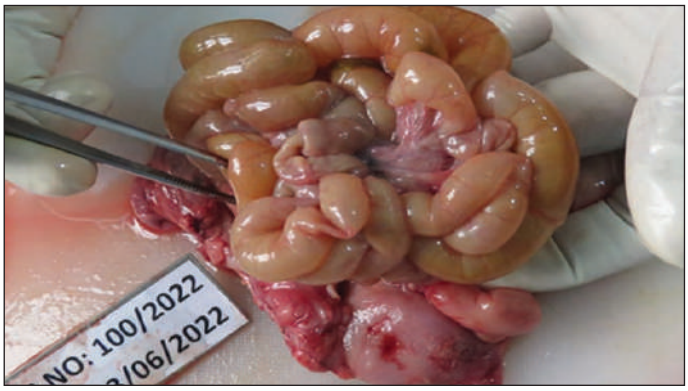

Photo 1. Multiple intussusceptions on gross.

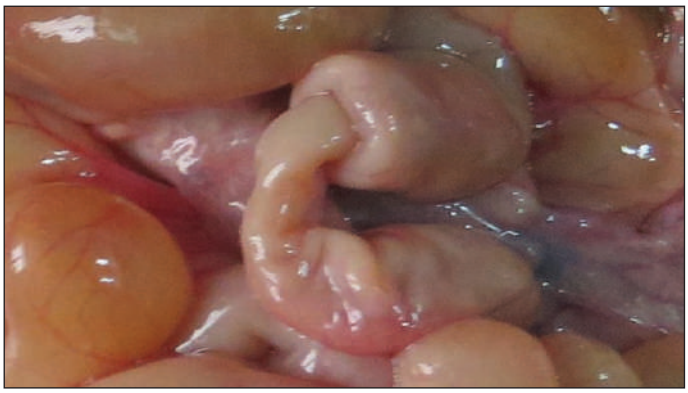

Photo 2. Single Intussusception site .

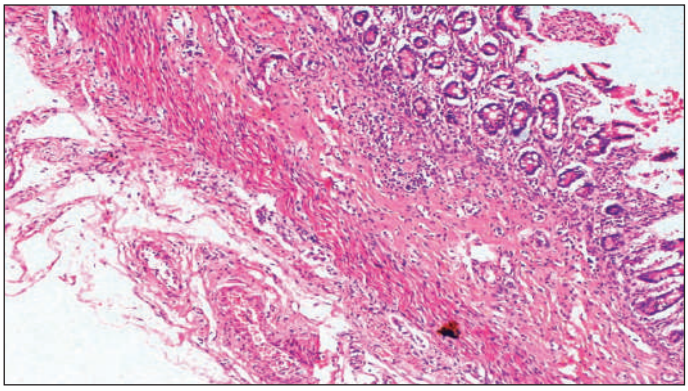

Photo 3. Inflammatory infiltrate in all layers, H&E, 10X.

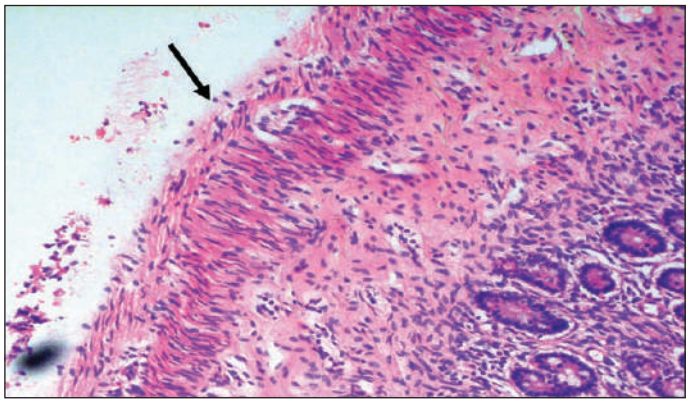

Photo 4. Inflammatory infiltrate upto serosa, H&E, 40X.

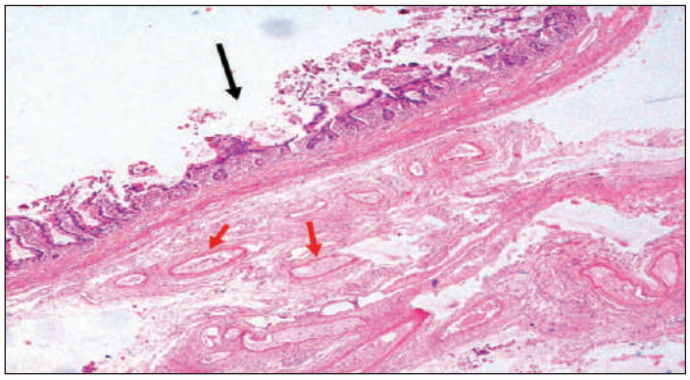

Photo 5. Mucosa showing focal ulceration (black arrow) and dilated lymphatics (red arrow), H&E, 10X.

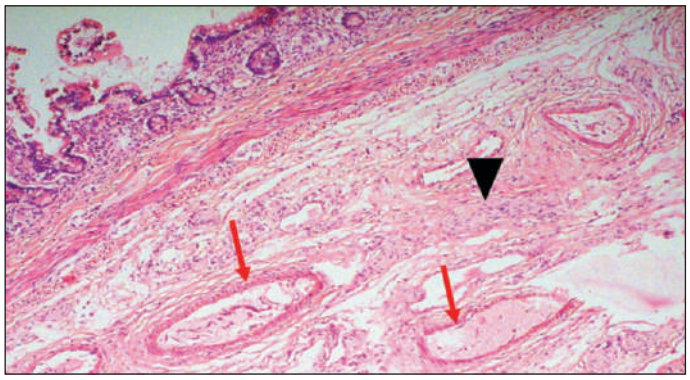

Photo 6. Hypertrophic nerve bundles (black arrowhead) and dilated lymphatics (red arrow), H&E, 40X.

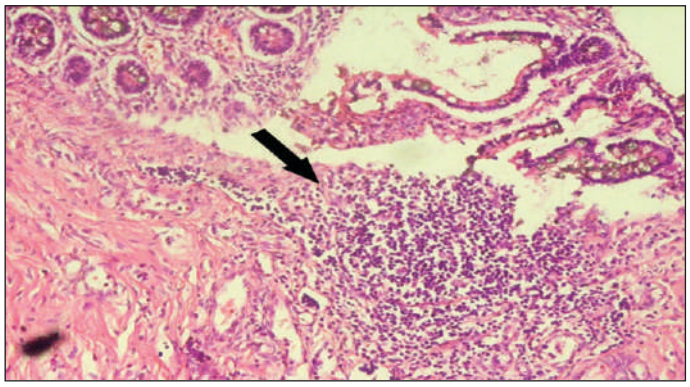

Photo 7. Hyperplasia of payer's patches, H&E, 20X.

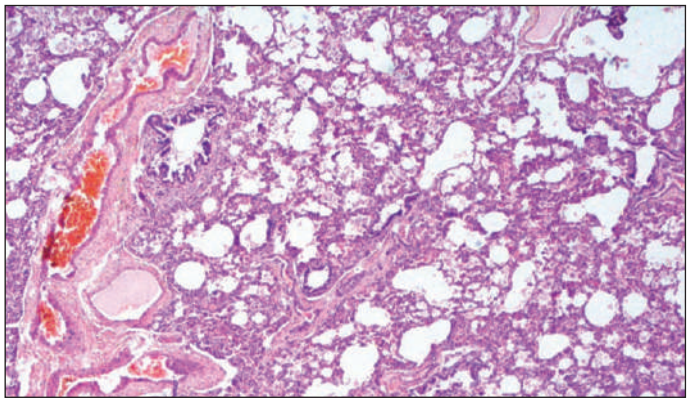

Photo 8. Focal alveolar damage, inflammatory infiltrates in septa and Congested blood vessel. H&E, 4X.

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Histopathological examination of the tissue specimens from the affected segment from intestine showed predominantly lymphocytic infiltration with few neutrophils present in all layers of the ileum (Photo 3 and 4). Also, focal ulceration of mucosa was seen along with dilated lymphatics (Photo 5). Hypertrophic nerve bundle was also evident in the myenteric plexus of intestine (Photo 6). Hyperplasia of payer's patches was also noted in one field on histopathology (Photo 7).

Other remarkable histopathological findings were present in tissue specimens of lungs which showed lymphocytic infiltration in alveolar septae, congestion of large blood vessels and focal alveolar damage at places (Photo 8).

Discussion:

Intussusception occurs when a segment of the intestine, constricted by a wave of peristalsis, telescopes into the immediately distal segment. Once trapped, the invaginated segment is propelled by peristalsis and pulls the mesentery along. Untreated, intussusception may progress to intestinal obstruction, compression of mesenteric vessels, and infarction. The most common type is ileocolic, followed by ileo-ileal, jejunojejunal and colocolic. The age range is four weeks to 13 years, with a male to female ratio of 1.8: 1. Some authors noted age range between 3 to 9 months and a male predominance. Our case was a 7 days old infant.[2] In a developing country like India, where there are health infrastructure constraints, a clinical diagnosis such as intussusception which requires imaging modalities especially in under-communicating neonates are not readily available and this may delay the prompt diagnosis and immediate management of the condition which could significantly contribute to infant mortality rates. The mortality with treatment is estimated to be about 1- 3%, but if untreated the condition is uniformly fatal in 2- 5 days.[1]

Symptoms and signs of intussusception are colicky abdominal pain, abdominal distension, fever, bloody mucoid stools, vomiting and a palpable abdominal mass but symptoms may either be absent or quite subtle.[2,3] Several association and possible causative factors have been identified. Among them are large peyer's patches, polyps, Meckel diverticula, cystic fibrosis, lymphoma, viral etiology particularly adenovirus and heredity. Recent history of upper respiratory illness was also a predisposing factor.[3,4] Death occurs in untreated intussusception from a combination of factors, including intestinal obstruction with resultant fluid and electrolyte imbalances, peritonitis, generalised sepsis and shock from intestinal infarction due to compromise of blood supply.[2,5] According to Iwase et al Demonstrable intussusception with aspiration of vomitus is hallmarks of the diagnosis.[6]

It is important to clearly distinguish agonal intussusception at autopsy from one which has genuinely caused antemortem functional disturbance. Agonal intussusceptions are believed to occur terminally, possibly related to intestinal dysmotility and peristaltic incoordination, and are an incidental finding, not a cause of death. They are easily reducible and macro and microscopic viability of the intussuscepted intestine is obvious.[2] Findings are corroborative grossly and microscopically in antemortem intussusception and not in post mortem agonal artefact.

Most common histological findings in cases of childhood intussusceptions are focal lymphoid hyperplasia, sometimes only edema which may be incidental finding.[3] Some studies reported ischemic necrosis of the intestine associated with vascular congestion and focal hyperplasia of the peyer's patches.[4] On histopathology of the affected intestinal segment, we noted inflammatory infiltrates from mucosa to muscle layer, ulceration of mucosa, dilated lymphatics, hypertrophic nerve bundle & hyperplasia of payer's patches. In lungs, on histopathological examination the findings were of focal alveolar damage, inflammatory infiltrates in septa & congested blood vessels.

Conclusion:

Present case exhibits an uncommon fatal occurrence due to intussusception and demonstrated the importance of forensic autopsy and histopathology in such unexpected infant deaths. It is important to clearly distinguish agonal intussusception at autopsy from one which has genuinely caused antemortem functional disturbance.

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