Neonatal Intramural Calcification in Jejunal Atresia: Case Report of a Rare Phenomenon

Abstract

Intramural calcification in intestinal atresia is a rare type of intra-abdominal calcification. The exact etiology of intramural calcification remains obscure. A 1-day-old newborn male baby presented with signs of intestinal obstruction and was diagnosed to have jejunal atresia. The newborn underwent laparotomy with resection of atretic and dilated part of the small bowel. Histology of atretic part of jejunum and adjacent area revealed intramural calcification with extensive foreign-body giant cell reaction. This appears to be the first time that intramural calcification has been documented in association with extensive foreign-body giant cell reaction in a case of jejunal atresia. It can be hypothesized that vascular insult is the initiating event. The further consequences could be multifactorial. This could be the reason for the variation in the site of calcific deposits. Intramural calcification with extensive foreign-body giant cell reaction is a rare phenomenon and calls for focused studies aiming at elucidating the exact etiopathogenesis of intramural calcification.

Keywords: Foreign body, intramural calcification, jejunal atresia

Introduction

Neonatal intra-abdominal calcification is a relatively rare phenomenon. Intra-abdominal calcification may occur as meconium peritonitis, intraluminal calcification, and intramural calcification. Intramural intestinal calcification is least common type among the three types of calcification. It is associated with intestinal atresia, meconium ileus, or intrauterine volvulus. [2]

Jejunal atresia is an antenatally acquired common cause of intestinal obstruction in a newborn with an incidence rate of 1:5000.^[3] Only a few cases of intramural calcification associated with jejunal atresias have been reported.

Case Report

A 1-day-old newborn male baby presented with bilious vomiting. The baby cried immediately after birth and weighed 2400 g (<3rd centile). The baby was born by cesarean section. The indication for cesarean section was fetal distress. Apgar score was eight at 1 min and nine at the end of 5 min. Third-trimester antenatal ultrasound findings revealed significant distension of gut loops, which suggested high bowel obstruction with moderate polyhydramnios.

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The newborn had one episode of bilious vomiting. Postnatal X-ray was done. The newborn was diagnosed to have jejunal atresia. On the 2nd day, laparotomy was done with resection of atretic and dilated part. Distal small bowel was suspected to have a perforation. Morphologically, it appeared to be type I atresia (Grosfeld modification of Louw classification). The resected ends of intestine were anastomosed. Peroperatively, plenty of interloop adhesions were noted, with minimal fluid in the peritoneal cavity.

Two segments of small intestine were received. The longer segment measured 15 cm, and shorter segment measured 5 cm. The longer segment showed narrowed [atretic] area near the distal resected end, measuring two centimeter [Figure 1]. The atretic segment was 2 cm from the distal resected end. The proximal dilated small bowel measured 11 cm from the atretic segment. The rest of mucosa of the small bowel segments appeared normal. There was no obvious evidence of perforation in the resected specimen.

Sections from atretic segment and the adjacent area showed intramural calcification, [Figure 2] chronic inflammatory cell infiltration and extensive foreign-body type of giant cell reaction

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in the vicinity of calcification [Figure 3]. The overlying mucosa showed focal areas of congestion and erosion at



Figure 1: Gross photograph of segment of jejunum showing atresia (white arrow)

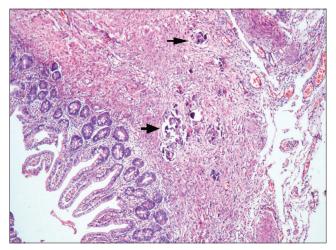


Figure 2: Tissue section of atretic segment of jejunum showing intramural calcification (black arrows) (H and E, $\times 100$)

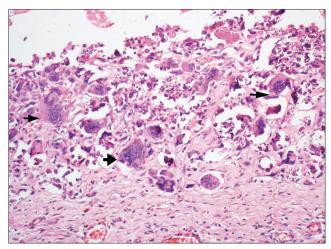


Figure 3: Tissue section of atretic segment of jejunum displaying extensive foreign-body type of giant cell reaction in the muscular layer (black arrows) (H and E, ×400)

places. The sections from rest of the intestine and resected surgical margins showed no significant pathology. The baby was discharged in a stable condition after a stay of 16 days.

Discussion

Neonatal intra-abdominal calcification usually accompanies bowel obstruction due to atresia or meconium ileus in cases of cystic fibrosis.^[1,4] The present case is that of intramural calcification in jejunal atresia. Intramural calcification has been documented in small intestine with and without atresia.^[1,2,4-6] In the present case, the patient was a 1-day-old male baby presenting with jejunal atresia.

Intramural calcification has been studied in a newborn of <3 days associated with atresia. [1,7,8] Contrastingly, intramural calcification in nonatretic bowel has been documented after 3 weeks. [2] Intramural calcification in a case of ileal atresia, associated with ileal duplication cyst has also been reported. [5] Subbarayan *et al.* [9] observed calcification in the mucosal layer in six cases of intestinal atresia.

In the present case, minimal peritoneal fluid and inter-loop adhesions of bowel loops were seen at surgery which raised the suspicion of perforation. However, no evidence of perforation was seen both grossly and microscopically. This is suggestive of a perforation which might have sealed off before birth. Brodribb^[7] had documented similar observation.

Cases have been documented demonstrating calcification in the mucosa and submucosa.^[1,2,7] Reports of calcific deposits with giant cell reaction in the submucosa of ileum has been described.^[5] In contrast, in the present case, the calcification was predominantly in the muscularis propria and was accompanied by the extensive foreign-body type of giant cell reaction. Mucosa and submucosa showed congestion and focal erosion. Tyagi *et al.*^[10] documented fibrotic changes in muscular layer in the preatretic part of most of the cases of jejunal atresia.

The calcification of meconium peritonitis is usually linear or plaque-like on the serosal surface of the bowel and abdominal wall. Meconium peritonitis is a result of meconium spillage through perforation of the obstructed bowel wall. This causes sterile peritonitis with calcium deposition composed of cornified epithelial cells, which is a part of extravasated meconium. Intraluminal calcification in case of bowel obstruction is amorphous or punctuate. It is postulated that interaction of urine and meconium might produce intraluminal calcification. [1] In 1915, Rudnow observed that meconium in the peritoneal cavity can calcify within a matter of few days. Forshall, Hall, and Rickham hypothesized that calcification in bowel wall was probably the result of intrauterine perforation of intestine. [11]

Both bowel atresia and intramural calcification may be due to ischemia.^[1] The alterations in the bowel wall depend on

the extent, severity, and duration of ischemic event. The mucosa and submucosa are more susceptible to damage than muscularis.^[2] Dystrophic calcification is known to occur in necrotic tissue. Interference with intestinal blood supply can cause atresia in uterus.^[1]

The peculiarity of the present case is that calcific deposits were predominantly in the muscularis propria with relative sparing of submucosa and mucosa. Steinfeld *et al.*^[6] observed that calcium in the atretic segments appear to lie in the dilated lymphatics within the bowel wall. The authors suggested that lymphatic stasis occurred in tissue which had become transformed as the result of vascular compromise. Aharon *et al.*^[1] opined that whether the calcifications are due to ischemia or are the result of introduction of meconium into the bowel wall through the ulcerated mucosa remains conjectural.

It can be hypothesized that vascular insult is the initiating event. The further consequences could be multifactorial. This could be the reason for the variation in the site of calcific deposits. Factors like pH of fetal environment and maternal lifestyle during pregnancy may have some influence. Foreign-body giant cell reaction could be secondary to calcific deposits.

Conclusion

Intramural calcification in a case of jejunal atresia is a rare phenomenon. This appears to be the first time that intramural calcification has been documented in association with extensive foreign-body giant cell reaction in a case of jejunal atresia. It can be hypothesized that vascular insult is the initiating event for calcium deposition. The further consequences could be multifactorial. This could be the reason for variation in the site of calcific deposits. The exact etiology remains conjectural and calls for focused studies aimed at elucidating the pathogenesis of calcification in the atretic bowel.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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