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Ileal duplication cyst with gastric heterotopia and melanosis in a toddler: A rare case report

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Abstract

Enteric duplication cysts are rare congenital anomalies, with the ileum being the most commonly affected site. The presence of gastric heterotopia within these cysts is uncommon, and the coexistence of melanosis is exceedingly rare. We report a unique case of ileal duplication cyst with both gastric heterotopia and melanosis in a pediatric patient. A 2-year-old male presented with abdominal pain and discomfort lasting for one month. Imaging revealed a well-defined cystic lesion in the right lower abdomen suggestive of an enteric duplication cyst or mesenteric cyst. The child underwent exploratory laparotomy with resection and anastomosis. The resected specimen was sent for histopathological evaluation. Gross examination showed a segment of ileum with an attached cyst measuring 1.5 cm in diameter. The cyst contained serous fluid and pigmented areas. Microscopic examination revealed gastric mucosa with areas of ulceration and inflammation. Numerous melanophages containing coarse melanin pigment were seen in the submucosa and muscularis. The diagnosis of ileal duplication cyst with gastric heterotopia and melanosis was confirmed. This case highlights a rare and unusual histopathological presentation of an ileal duplication cyst. The coexistence of gastric heterotopia and melanosis is extremely uncommon and should be recognized as part of the differential diagnosis in pediatric gastrointestinal anomalies.

Introduction

Gastrointestinal duplication cysts are uncommon congenital anomalies that typically present in childhood. They are characterized by a well-formed muscular wall and mucosal lining that resembles segments of the gastrointestinal tract. The ileum is the most common location for duplication cysts. These cysts may contain ectopic mucosa, most commonly gastric or pancreatic, which can result in clinical complications such as ulceration, bleeding, or perforation.[1] Gastric heterotopia has been frequently described in enteric duplication cysts; however, melanosis — defined by the presence of melanin-laden macrophages (melanophages) — is almost never associated with small bowel duplications. [2] Melanosis coli, in contrast, is more commonly seen in the colon, typically associated with laxative use.[3] The coexistence of melanosis with gastric heterotopia in an ileal

duplication cyst is exceedingly rare and not well-documented in the literature. [4]

Case Report

A 2-year-old male presented with intermittent abdominal pain and discomfort lasting for one month. On examination, the child had a mildly distended and tender abdomen. Laboratory findings were within normal limits. Contrast-enhanced computed tomography (CECT) of the abdomen showed a well-defined cystic lesion in the right lower quadrant. The radiological differential diagnosis included enteric duplication cyst and mesenteric cyst. A clinical diagnosis of intestinal obstruction with mesenteric cyst was made.

The patient underwent exploratory laparotomy with resection and anastomosis. Intraoperative findings revealed intestinal obstruction and a gangrenous bowel segment at the ileocecal junction. The affected segment was resected and sent for histopathological evaluation.

Gross:- Specimen consisted of 5 cm segment of ileum with an attached cyst measuring 1.5 cm in diameter. External surface revealed a focal grey black area. On cut surface serous fluid drained, blackish brown pigment noted. Adjacent to it other segment showing blackish brown pigment attached to the thickened wall of the ileum.

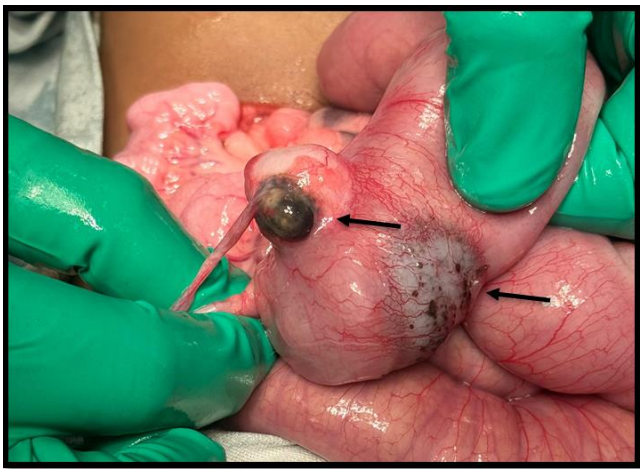

Figure 1: Arrows showing greyish brown pigment at the ileocecal junction. Intraoperatively.

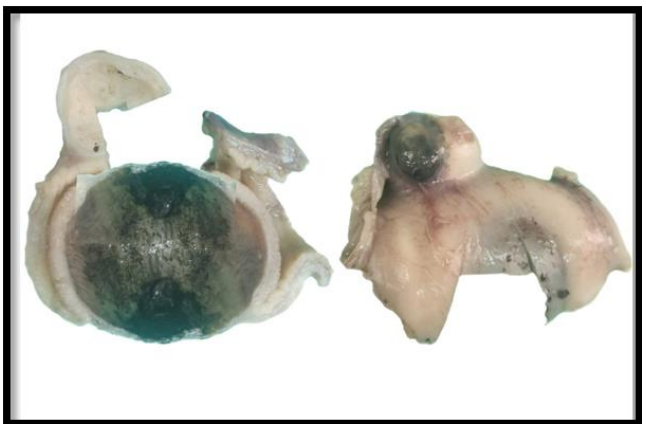

Figure 2: Formalin fixed specimen on cut surface showing blackish brown pigment.

Figure 3: Arrow showing ileal wall with attached cyst showing pigmentation.

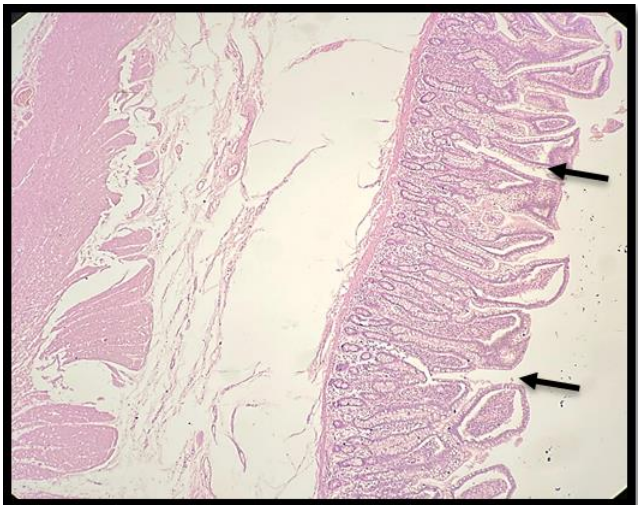

Figure 4: Arrows showing cystic wall lined by gastric mucosa (heterotopia) in the ileal wall

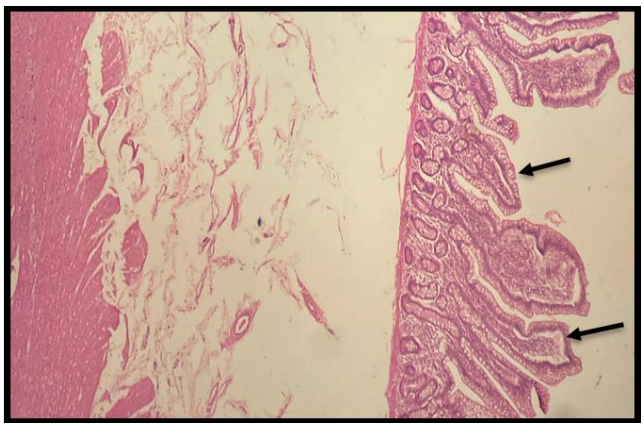

Figure 5: Arrows showing cystic wall lined by gastric mucosa (heterotopia) in the ileal wall.

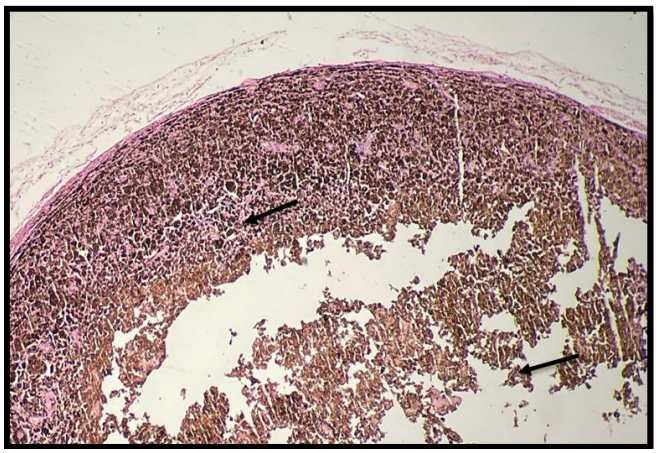

Figure 6: Arrows showing numerous melanophages containing coarse melanin pigment in the submucosa and muscularis

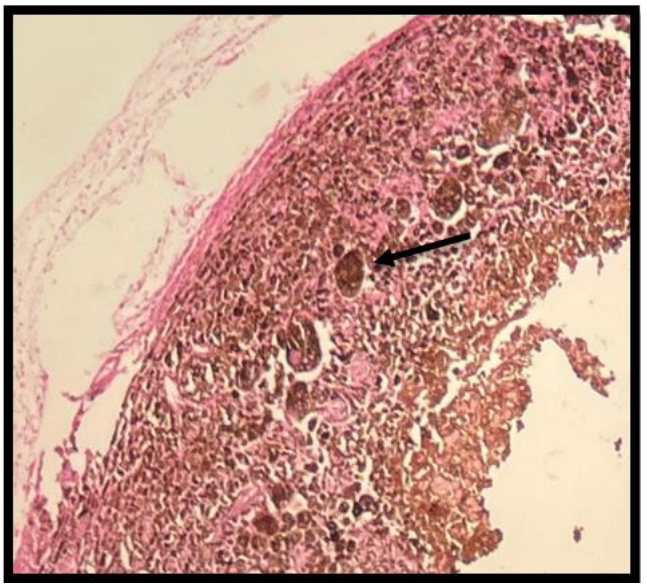

Figure 7: Arrows showing coarse melanin pigment.

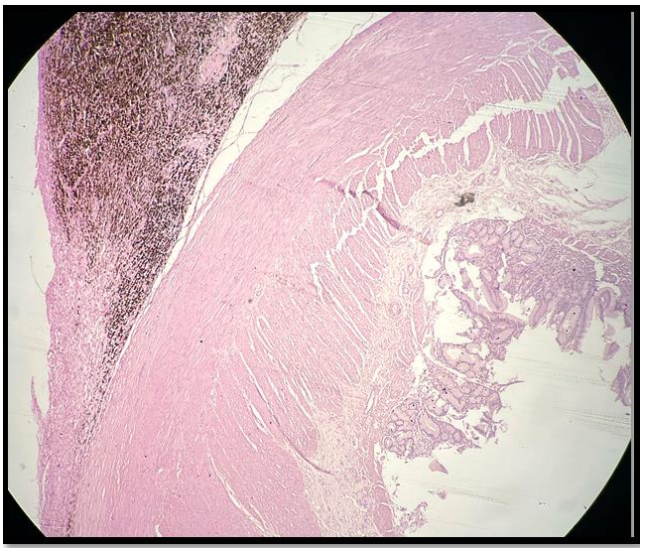

Figure 8:

Figure 6, 7 and 8 showing Numerous melanophages containing coarse melanin pigment in the submucosa and muscularis.

Microscopic examination: Hematoxylin and eosin-stained revealed a cystic wall lined with gastric-type mucosa and underlying muscle layers, consistent with gastric heterotopia. Focal ulceration and inflammatory infiltration were

observed. Numerous melanophages containing coarse melanin pigment were present in the submucosa and muscularis. The wall of the duplication cyst displayed ileal mucosa, confirming the diagnosis of an ileal duplication cyst with gastric heterotopia and melanosis.

Discussion

Enteric duplication cysts (EDCs) are rare congenital anomalies characterized by the presence of a well-developed smooth muscle wall and mucosal lining, typically located on the mesenteric side of the alimentary tract. They can occur anywhere along the gastrointestinal tract but are most commonly found in the ileum. EDCs may be cystic or tubular and often share a common wall with the adjacent bowel. The presence of heterotopic mucosa, particularly gastric or pancreatic tissue, is noted in approximately 17% to 36% of cases. [1]

The coexistence of melanosis within an EDC is exceedingly rare. Melanosis in the gastrointestinal tract is more commonly associated with the colon, often linked to chronic laxative use, and is characterized by the accumulation of melanin pigment within macrophages.[5] Its presence in the small intestine, particularly within duplication cysts, is not well-documented. The pathogenesis of melanosis in this context remains unclear but may involve chronic mucosal injury or developmental anomalies leading to pigment deposition.[2]

Clinically, EDCs can present with nonspecific symptoms such as abdominal pain, vomiting, or gastrointestinal bleeding, depending on their size, location, and mucosal lining. Imaging modalities like ultrasound and computed tomography (CT) scans are instrumental in diagnosing these cysts. Ultrasound may reveal the characteristic 'gut signature,' while CT scans can delineate the cyst's relationship with adjacent structures. In cases where ectopic gastric mucosa is suspected, a technetium-99m pertechnetate scan can be utilized, as it is taken up by gastric mucosa.[3]

Surgical resection remains the definitive treatment for EDCs, especially when symptomatic or complicated by bleeding or obstruction. Histopathological examination post-resection confirms the diagnosis and identifies the type of mucosal lining, which is crucial for understanding the potential for complications.[4,6] In our case, the identification of gastric heterotopia and melanosis within the ileal duplication cyst underscores the importance of thorough histological evaluation.

This case adds to the limited literature on the rare occurrence of melanosis within EDCs and highlights the need for awareness among clinicians and pathologists. Recognizing such unusual histopathological findings is vital for accurate diagnosis and appropriate management.[7,8]

Conclusion

This case illustrates an extremely rare pathological entity: ileal duplication cyst with gastric heterotopia and melanosis. Pediatric surgeons and pathologists should be aware of such rare combinations, especially when evaluating cystic lesions associated with intestinal obstruction. Recognition of these findings is essential

for guiding management and understanding potential complications.

Source of Funding

None.

Conflict of Interest

None.

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