Case Report

Long segment jejuno-ileal duplication cyst with ectopic gastric mucosa detected on 99mTc-pertechnetate scintigraphy

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ABSTRACT

Enteric duplication cysts (EDC) are uncommon congenital anomalies that may occur anywhere along the digestive tract. Ectopic gastric mucosa (EGM), another rare condition, is usually present as short segments in the small intestine and may be associated with EDC. Abdominal scintigraphy with 99mTc pertechnetate may be useful in the diagnosis, since the radiotracer is concentrated by functioning gastric mucosa. In this case report, the authors describe a child with a 150 cm long jejuno-ileal duplication cyst containing EGM identified by intense 99mTc pertechnetate uptake on scintigraphy without any pharmacological intervention.

Keywords: Ectopic gastric mucosa, enteric duplication, pertechnetate, scintigraphy

INTRODUCTION

Enteric duplication cysts (EDC) are uncommon congenital anomalies that generally occur in the small intestine. Ultrasonography (USG) and computerized tomography are the usual imaging modalities employed to make a diagnosis. 99mTc pertechnetate is concentrated by ectopic gastric mucosa (EGM) that may occur in these cysts and scintigraphy using this radiotracer may be a useful modality in the pre-operative diagnosis of this rare anomaly.

CASE REPORT

A 6-year-old male child operated for exstrophy of the urinary bladder 4 years back presented with a 10 days history of fever, vomiting, abdominal pain and hematochezia. At presentation, hemoglobin was 10 g/100 ml and total leucocyte count 10,200/mm^3 with normal coagulation parameters. Suspecting a Meckel's diverticulum, dynamic abdominal scintigraphy was performed under a gamma camera for 20 min after intravenous injection of 37 MBq (1 mCi) of 99mTcO_4- (pertechnetate). Tracer uptake was seen in the central abdomen, corresponding to the shape of the small intestinal loops. The intensity was seen to increase simultaneously with that of the stomach, indicative of extensive ectopic functioning gastric mucosa [Figure 1]. USG showed a large well defined bean-shaped cystic lesion in the left lumbar region with a positive gut signature and internal debris and echoes, suggestive of an infected EDC. Exploratory laparotomy was performed and a 150 cm long tubular jejuno-ileal duplication cyst communicating with the ileal lumen was excised [Figures 2 and 3]. Presence of EGM in the cyst was confirmed on histopathology, which showed hyperplastic gastric lining epithelium, with viliform transformation at places [Figure 4] and marked edema and vascular congestion in the submucosa. However, the patient's condition deteriorated post-operatively, and the child died of septicemia 9 days after surgery.

DISCUSSION

EDC are uncommon congenital anomalies, may be single or multiple and may occur anywhere along the digestive tract on the mesenteric side. The small intestine accounting for about 50% of all lesions is the most commonly involved. Two thirds of the lesions are ileal in location and one-third jejunal.[1,2] The lumen of the duplication is usually not in continuity with the normal intestine; however, the two structures share a

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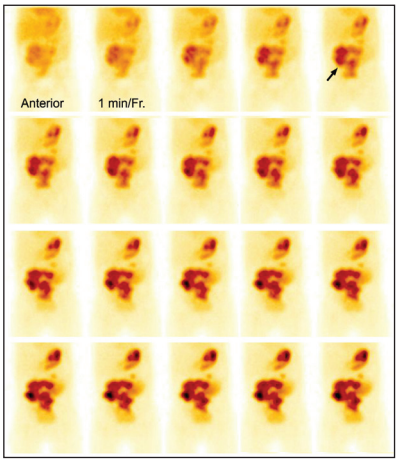

Figure 1: 99mTc pertechnetate scintigraphy showing increased tracer concentration in the central abdomen (arrow) corresponding to the small bowel. The intensity is seen to increase simultaneously with that of the stomach indicative of ectopic functioning gastric mucosa

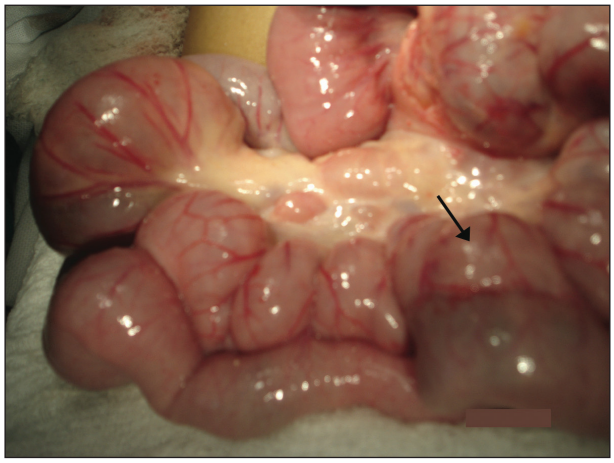

Figure 2: Operative photograph showing the duplicated small bowel (arrow)

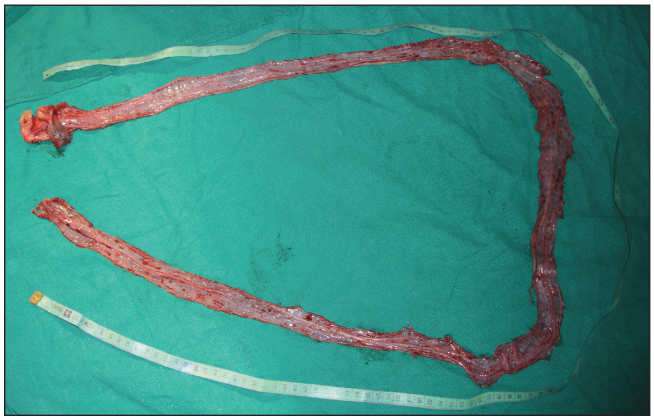

Figure 3: The entire excised duplication cyst measured post-operatively

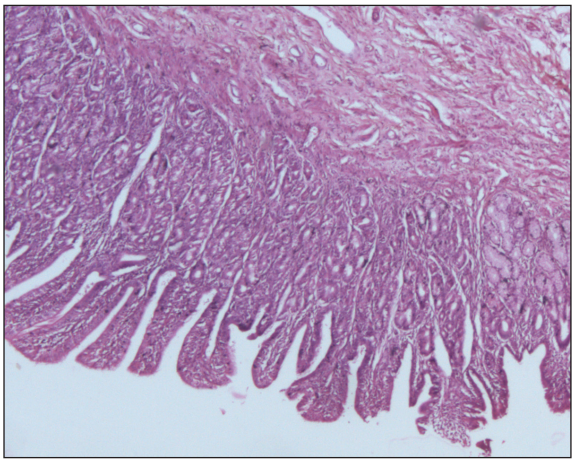

Figure 4: Histological section from the cystic duplication showing hyperplastic gastric lining epithelium, with viliform transformation with submucosal edema and vascular congestion

portion of the muscular coat and vascular supply.[3] Presenting symptoms include abdominal distention, vomiting, bleeding and a palpable abdominal mass, with complications like perforation, intussusception, bowel obstruction, volvulus and associated malignancy. Frequent micturition is a rare presentation.[4] This condition needs to be differentiated from other causes of lower gastrointestinal bleeding in children like polyps, clotting disorders, arteriovenous malformations and Crohn's disease.[3]

On USG, a double-layered wall (the combination of an echogenic inner mucosal layer and hypoechoic outer muscular layer) is found in over 50% of duplication cysts.[5] 99mTc pertechnetate is concentrated by EGM in a duplication cyst, similar to the uptake seen in gastric mucosa within a Meckel's diverticulum, and this phenomenon may be useful to diagnose this condition. However, the presence of gastric mucosa within duplication cysts is variable, and has been reported to be 17-36%.[6] Both Meckel's diverticulum and intestinal duplication should be included as differential diagnostic possibilities for a focus of increased tracer activity on 99mTc pertechnetate abdominal scintigraphy, because the two conditions cannot be distinguished based on the size or location of the EGM.[3] A normal 99mTc pertechnetate study should not show any focal tracer accumulation other than in the stomach and the urinary tract. The sensitivity of 99mTc pertechnetate imaging for detection of EDC is reported to be 75%.[7] False-positive uptake of this radiotracer may be seen in intussusception, focal hyperemia or bowel inflammation, gastrointestinal bleeding unrelated to EGM, uterine blush, retention of tracer in the urinary collecting system, vascular lesions such as hemangiomas and arteriovenous malformations.[3]

EGM in the alimentary tract is a rare condition, and the length of the involvement usually ranges from a few to several centimeters, with contiguous or composed of islands of EGM. It is usually diagnosed during laparotomy for complications of EGM, such as perforation or obstruction. While 99mTc pertechnetate scintigraphy in Meckel's diverticulum has been described in several studies, the pre-operative diagnosis of EGM in the small intestine using this technique is less common. The localized tracer activity in EGM shows a variable pattern, and tends to decrease during the course of imaging due to dilution with intestinal

secretions. Locally concentrated tracer also gets washed away by peristaltic activity.[8-10] The present case differs significantly from others previously described in the literature in the unusual length of the involved segment.

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