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Congenital Ilea Duplication Cyst Lined By Columnar and Squamous Epithelium: A Rare Case Report

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Abstract: Enteric duplication cysts are a rare congenital anomaly. They can occur anywhere in the gastrointestinal tract. Their most common location is the small intestine, particularly the terminal ileum. We report a rare case of a neonatal male child with intestinal duplication cyst of the ileum lined by columnar and stratified squamous epithelium.
Keywords: cysts, congenital anomaly

INTRODUCTION

Enteric duplication cysts are a rare congenital anomaly. These lesions are found anywhere in the gastrointestinal tract but the most common site is small bowel [1]. Incidence of enteric duplication cysts are reported to be 1 in 4500 [2]. They may present in a variety of ways ranging from mild symptoms like vomiting, abdominal distension to lethal complications such as volvulus, intussusception and bowel obstruction [3, 4]. More than 80% of the cases present before 2 years of age. The most common presentation is intestinal obstruction [4].

CASE REPORT

A male baby born at term by normal vaginal delivery to a second gravida mother weighing 2800 grams had intermittent vomiting and poor feeding since birth. An antenatal ultrasound performed at 36 weeks gestation showed an abdominal cyst thought to be a duplication cyst. No resuscitation was required at birth. On day 10 of birth he presented with persistent bilious vomiting and distension of abdomen. On examination he was irritable; abdomen was distended but soft with a palpable mobile mass in the right lower quadrant. Plain X-ray abdomen showed air fluid levels. Abdominal ultrasound revealed a cyst in the right abdomen measuring 6.5 x 3.5 cm with appearances suggestive of duplication cyst.

Exploratory laparotomy was performed which revealed a fluctuant cystic lesion measuring 8.5 x 5.5 cm involving the ileal portion of the small bowel in the mesenteric part, approximately 8 cm from the ileo-cecal junction. The cyst was not in communication with the lumen of the ileum. The segment of ileum comprising the ileal duplication was resected and primary end to end anastomosis was performed. The excised specimen was sent for histopathological examination.

The resected segment of ileum grossly showed a globular cystic dilated segment measuring 9 x 5.5 cm. External surfaces was smooth and glistening (Fig-1). On cutting open mucosa was filled with grey brown fluid.

Fig-1: Resected segment of ileum

Microscopic examination of the sections prepared and stained from the specimen showed cyst wall with all the layers of the intestine lined by columnar epithelium and non-keratinizing stratified squamous epithelium at places (Fig-2). Consistent with duplication cyst. His postoperative period was uneventful and the patient thrived well.

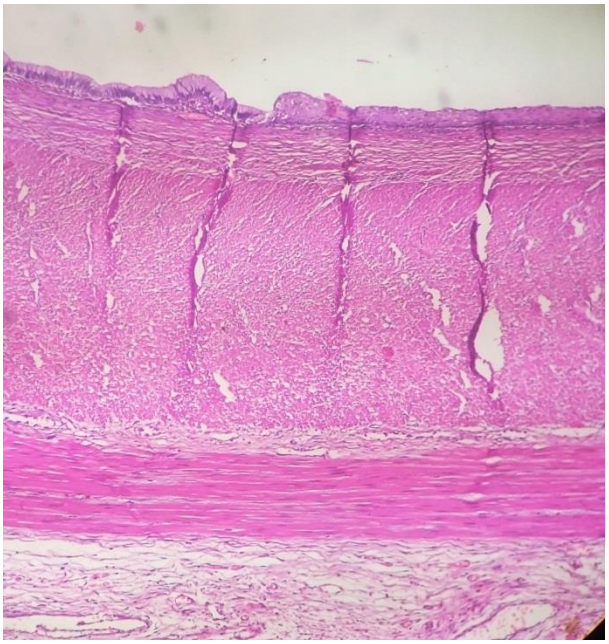

Fig-2: 40X view of the cyst wall showing transition from columnar epithelium to non keatinizing squamous epithelim

DISCUSSION

Enteric duplication cysts are epithelium lined cystic, spherical or tubular structures that are attached to wall of gastrointestinal tract. They may be in communication with the gastrointestinal tract or not [1]. The most common site of enteric duplication cyst is small intestine with about 60-70% occurring in the ileum [5]. Other sites in decreasing order of frequency are oesophagus, colon, jejunum, stomach and duodenum [6].

Enteric duplications are congenital and develop from disturbances in embryonic development [7]. Various theories have been proposed for its development. The most favoured theory is the one proposed by Brenner in [3]. He suggested that the cysts arise because of the fusion of longitudinal folds allowing a passage for sub mucosa and muscle layer at the second or third month of intrauterine life [7].

The cysts must have at least one outer muscular layer usually lined by mucosa of the native tissue [5]. Ectopic tissue may be present in 25-30 % of the cases. Gastric mucosa is the most common ectopic tissue seen in 50 % of cases, followed by pancreatic exocrine and endocrine tissue [2]. Some enteric duplication cysts lined by epithelial linings not resembling that of the native tissue such as cuboidal, respiratory lining epithelium, cartilage, stratified squamous epithelium have been reported rarely [8]. This case falls in the minority of reported duplication cysts. In this case the lining epithelium was composed of columnar epithelium with transition to non-keratinizing stratified squamous epithelium.

The presentation of enteric duplication cysts vary greatly with age. Infants and neonates present with abdominal pain, vomiting and abdominal mass[5]. It may even be asymptomatic for several months and even years and may present during adulthood[2]. Complications include perforation, obstruction volvulus etc. Rare cases of malignancy arising within duplication cysts have also been reported [3].

Ultrasonography is an excellent tool in the prenatal diagnosis of congenital enteric duplication cysts and the likelihood of detection is very high [9]. Postnataly and in adults abdominal ultrasonography may identify the cyst. It shows inner hyper echoic mucosal and outer hypo echoic muscular layer [3].

The diagnosis is best established with contrast enhanced computed tomography (CECT) of abdomen. It demonstrates a filling defect or luminal communication with the bowel [5]. Recently endoscopic ultrasound has been widely used as an evaluation and diagnosis of enteric duplication cysts since it can distinguish between solid and cystic lesions [10].

The treatment of choice for symptomatic enteric duplication cyst is surgical excision [3]. The treatment protocol for asymptomatic cysts is not clearly established though some authors favour resection in view of preventing complications [5].

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