Int. j. adv. multidisc. res. stud. 2024; 4(4):166-168

Received: 21-05-2024
Accepted: 01-07-2024

International Journal of Advanced Multidisciplinary Research and Studies

ISSN: 2583-049X

Enteric Duplication Cyst Containing Respiratory Squamous Epithelium: A Case Report

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DOI: https://doi.org/10.62225/2583049X.2024.4.4.3019
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Abstract

Enteric duplication cyst of the alimentary tract is a rare congenital anomaly that can occur anywhere throughout the alimentary tract most commonly the small bowel. There is two types of gastrointestinal duplications exist: Tubular and cystic, however, studies have reported that foregut duplication cysts can have a respiratory epithelium lining. It is rare. We present a case of an enteric duplication cyst arising from the ileum lined with respiratory squamous epithelium, occurring in a 3years-old girl. The uncommon histology and unusual presentation make this case a unique of what is already a rare pathological entity.

Keywords: Respiratory Squamous Epithelium, Intestine, Duplication Cyst

Introduction

Duplications of the gastrointestinal tract are rare that predominantly present before the age of 2 years. The incidence has been reported to be 1/4500. It's located on the mesenteric border of the associated native bowel and varies in size, shape, location and symptoms, it could be cystic in 80% of cases or tubular in the remaining 20%, Gastric duplications account for only 3-4% of all gastrointestinal duplications, occur twice as often in females than males, and occur along the greater curvature of the stomach in over 50% of cases. Duodenal duplica-tions are slightly more common than gastric duplications representing 5-6% of the total seen. Colon duplications are the rarest reported. Rectal duplications are also exceptionally rare. Intestinal duplications account for 45-55% of all gastrointestinal duplications. Most occur at the terminal ileum [1], with or without other congenital anomalies like spinal cord defects, hernia, club foot and doubling of the genital or urinary organs [2, 4]. Although the cause is unknown there are three theories for duplication cyst formation: Split notochord mechanism, incomplete recanalization and incomplete twinning. Intestinal duplications may contain epithelium of the bowel to which they associated, or they may contain ectopic intestinal or respiratory epithelium [1]. Duplications most commonly come to clinical attention through the development of obstructive symptoms. Tubular duplications often contain ectopic gastric mucosa. Acid secretion can cause peptic ulcers in the adjacent bowel with which they communicate. Since duplications can occur anywhere along the gastrointestinal tract, bleeding may be of the upper type with hematemesis and melena, Peptic ulceration and perforation of adjacent bowel occurs by a mechanism similar to gastric peptic ulcer disease, Not infrequently, tubular duplications of the abdomen and chest are associated with neurological symptoms of muscle weakness or paralysis before the extra-spinal cysts have declared themselves [1]. Some are discovered as an asymptomatic mass. Vertebral anomalies such as bifid or hemi-vertebrae are important diagnostic pointers, particularly with foregut duplications. Other congenital malformations are found in about half of all patients. For example, intestinal malrotation or atresia may be associated with midgut duplications and genitourinary anomalies or bladder exstrophy with hindgut lesions. Radiographic studies are helpful to confirm the diagnosis. Abdominal ultrasonagraphy helps localize cystic lesions within the abdomen and can determine whether they are complex or simple, unilocular or multilocular, and single or multiple. A radioisotope technetium scan to detect heterotopic gastric mucosa, Upper gastrointestinal contrast studies and endoscopy may be necessary in some cases. Magnetic resonance imaging (MRI) and computerised tomography (CT)does not add much additional information but occasionally can exclude other abdominal organs as the source of the cystic and to enable evaluation of the cranial and caudal extent of thecyst [5].In older children, endoscopic retrograde cholangio pancreatography or magnetic resonance cholangiography are helpful in the assessment of pancreatic and selected duodenal duplications and magnetic resonance angiography may help to plan surgery for large

retroperitoneal lesions. Pelvic duplications are best imaged by CT or MRI in conjunction with a contrast enema, fistulogram, endoscopy, and urinary tract sonography [5-7]. Recurrence rate after resection is around 6-13%. Mortality is extremely rare and generally associated with cysts complicated by volvulus and ischemic bowel necrosis [1]. Here we present a very rare case of a small bowel duplication cyst containing respiratory squamous epithelium.

Presentation

3years old female came to emergency room complaining of right iliac fossa pain and vomiting the condition started 6 days prior to admission by central colicky abdominal pain, continuous right iliac fossa pain and vomiting once for which she sought medical advice at her nearby clinic and received oral medication and sent home. One day later her condition had worsened with increase pain intensity and frequency so she referred to hospital at pediatric department, and treated as gastroenteritis without improvement therefore then they decided to refer her to surgical department, at the time when she arrived our clinic, she was still complaining from the central and right iliac fossa pain. There was no fever, anorexia, or abdominal distention, no upper or lower GIT bleeding, she had normal bowel habits with normal color stool, she was born at full term by cesarean section due to oligohydramnios crying immediately, and passing meconium, There were no abnormal findings in the prenatal ultrasonography, her antenatal history was uneventful there was no history of recent acute illness or trauma. Also she had normal developmental milestones up to her age. There is no known consanguinity between parents. Systemic reviews showed.

No symptoms of central nervous system, musculoskeletal system involvement.

No symptoms of respiratory system involvement.

No symptoms of urinary system affection (urine of normal color amount and frequency).

On examination she was calm a febrile her vital sign was PR 98beat /min, RR 20cycle /min, 98%O2at room air BP110/60. Abdominal examination showed normal abdominal contour, hernia orifices were intact, there was mild tenderness at right iliac fossa with soft abdomen and no superficial masses or organomegaly. About her investigation CBC⁄RFT⁄VS⁄ they were normal with normal range of electrolyte Abdominal ultrasound showed normal review. The patient was still having the pain which become more severe there fore she planned for exploration, she underwent an emergency laparotomy under general anesthesia. A horizontal predominantly right-sided laparotomy, approximately 2 cm below the umbilicus, was performed. Laparotomy findings were a lump 8×5×2cm oval in shape about 50 cm proximal to ileocecal valve originating from the mesenteric side of the bowel compressing ileum lumen forming partial narrowing (Fig 2) firm in consistency with multiple enlarged mesenteric lymph nodes (Fig 1) primary resection and anastomosis done, there were no other gastrointestinal tract anomalies or diseases. The patient made an uneventful recovery and discharge on good condition.

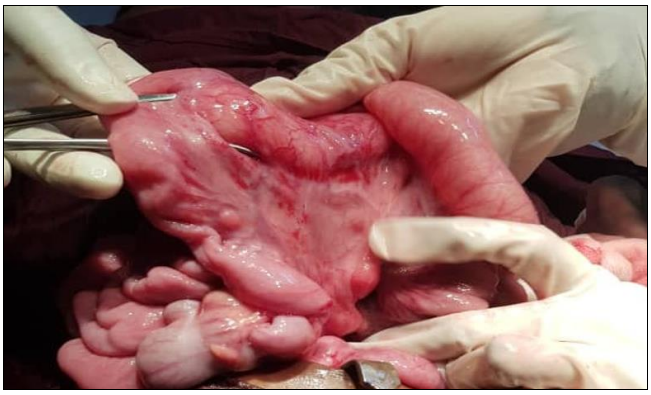

Fig 1: Duplication cyst with lymph node enlargement

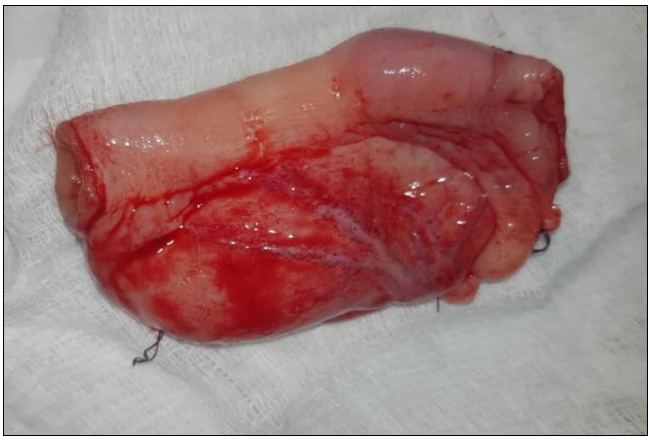

Fig 2: Excised part contain the cystic structure and part from the bowel

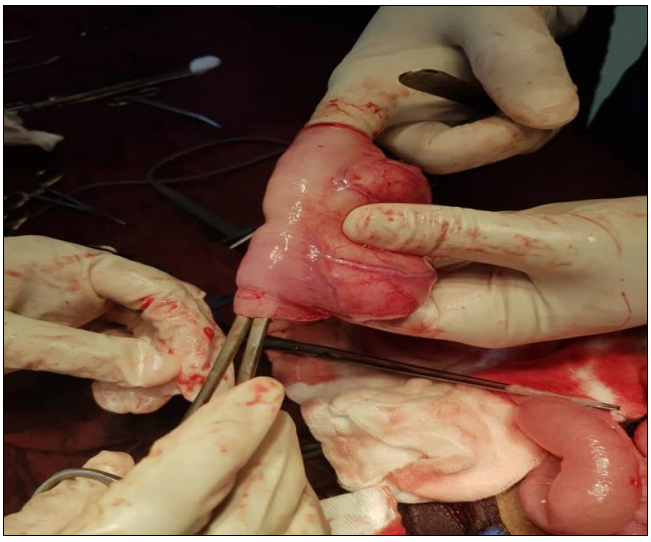

Fig 3: Partial ileum lumen narrowing at which the cyst originates from

Patient consent

Informed consent was obtained by both the parents of the patient for publication of this case.

Discussion

Enteric duplication cysts are rare congenital abnormalities that can occur anywhere along the digestive tract. They are

predominantly found in childhood with 67–80% presenting before the age of 2years. The type, size, and location of the cyst, as well as other factors like the presence of ectopic mucosa, communication with the adjacent bowel, or inflammation, are all related to the non-specific and variable presenting symptoms [8]. various treatments are conceivable depending on the size, location, blood supply and type of duplication. The treatment of choice is surgical excision of the cyst with preservation of the bowel and its blood supply [9]. If this is not possible, complete resection together with the normal bowel is mandatory and bowel continuity is restored with an end-to-end anastomosis [10]. However, our three years old girl present with intermittent central colicky and right iliac fossa pain and vomiting once. Although there was no anorexia, fever, abdominal distension, constipation or bloody stool. She was vitally stable and she had soft abdomen, her hernia orifice were intact, but there was mild tenderness at right iliac fossa, there were no any associated anomalies related to CNS, genital or urinary systems and she does not suffer from any foot or limbs abnormalities, she had normal blood test and normal abdominal scan by ultrasound, Preoperative diagnosis of Enteric duplication is uncommon with most being diagnosed post operatively. Because, it could be asymptomatic or present with non-specific and variable presenting symptoms with the complications that they can cause. With this limited scenario, preoperative diagnosis of Acute appendicitis, Meckel's diverticulitis and intussusceptions and abdominal tuberculosis were the differential diagnosis therefore, she underwent emergency laparotomy. A horizontal predominantly right-sided laparotomy, approximately 2 cm below the umbilicus, was performed. the intra operative diagnosis was made by founding small bowel mass 8×5×2cm oval in shape about 50cm from iliocecal valve arising from mesenteric border with multiple mesenteric lymph node enlargement (Figure 1, 2), that confirmed by histopathology latter on. Formal laparotomy revealed no other gastrointestinal tract anomalies or diseases. Other intra operative diagnosis was intestinal tuberculosis which was very common cause of abdominal pain in this country and this age. Since it was difficult to excise the mass and preserve the bowel because the mass was adherent to and compressing the wall of the adjacent ileal segment making mild to moderate narrowing to the lumen which explain the recurrent central colicky abdominal pain presentation (Fig 3) and this made excision of the cyst alone impractical. The surgical field protected by surrounding gauze swabs soaked in dilute aqueous povidone iodine. Localised lesions resected with the adjacent intestine after ligation and division of associated mesenteric vessels. Resection of 14cm (excision provided proximal and distal Free margins) of the ileum with end-to-end anastomosis is performed using a single layer of interrupted extra mucosal absorbable5/0 vicryl sutures. Excised duplications mass examined to assess the completeness of the excision and send for histopathology. The patient received a full work up following surgery to rule out any other related anomalies Histopathology result revealed small intestinal tissue with adherent cyst composed of muscularis mucosa its mainly lined by squamous epithelium with submucosal mucin secreting glands and respiratory epithelium confirming the diagnosis of an enteric duplication cyst containing respiratory squamous epithelium. Presently the patient is undergoing a semestral follow up with us. She is in a good condition does not experience any intermittent pain again.

In conclusion, GI duplication cysts are rare phenomena that present mainly in the pediatric population. Their presentation is reflective of the complications that they can cause. It is important to be aware and make a definitive diagnosis of this rare congenital anomaly as they can present in various clinical forms and can cause significant morbidity and even mortality if untreated by causing perforation obstruction, hemorrhage and a risk of late malignant degeneration, particularly with rectal and gastric duplications. Early resection avoids the additional operative difficulties arising from inflammation or perforation of the duplication cyst. The key steps in the successful surgical management of gastrointestinal duplications include: Understanding of the spectrum of these lesions, careful preoperative assessment of the cyst and its potential associated pathology, appropriate operative planning, complete excision where possible, and a full understanding of the alternative techniques if excision is deemed too hazardous.Also important to include them as a differential diagnosis of a vague intermittent recurrent abdominal pain in pediatric patients.

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