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Completely Isolated Enteric Duplication Cyst Presenting as an Inguinal Hernia

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Enteric duplication cysts are uncommon congenital anomalies whose embryogenesis remains unknown. We report here on an isolated enteric duplication cyst, that presents as an inguinal hernia. A 21-year-old woman was admitted with a month-long history of a palpable mass in the left groin. Radiologically, a computed tomography scan revealed a 3.5 × 2.5 cm sized cystic mass in subcutaneous layers of the left suprapubic area. Microscopically, the cystic wall resembled gut wall. The wall was composed of two distinct muscle layers with the presence of Auerbach's plexus. On examining the entire sections of the cyst wall very carefully, no epithelial lining was found on the inner surface. The submucosa was slightly fibrotic. The diagnosis was a completely isolated enteric duplication cyst.

Key Words : Duplication; Hernia, enteric

Enteric duplication cysts are uncommon congenital anomalies whose embryogenesis remains unknown. They can occur anywhere along the entire alimentary tract, and their most common location is the terminal ileum. Most enteric duplication cysts manifest during infancy or early childhood, although they can present at any age. They can present with variable clinical manifestations that depend on the location and size of the lesion, as well as the type of mucosal lining. Complications include bleeding, intestinal obstruction and perforation, intussusception, and rarely malignancy.

Herein, we present the first case of an isolated enteric duplication cyst, that presented as an inguinal hernia.

CASE REPORT

A 21-year-old woman was admitted with a month-long history of a palpable mass in left groin. She had no significant past medical or surgical history. On physical examination, there was a movable, tender mass on the left side of suprapubic area. A computed tomography scan revealed a 3.5 × 2.5 cm sized cystic mass in subcutaneous layer of the left suprapubic area (Fig. 1). The contour of the cystic mass was smooth. Radiologically, there was no communication between the cyst and the intestinal lumen. Laparotomy was done to remove the cystic mass. A skin incision of approximately 5 cm was made parallel and 2 cm superior to the inguinal crease. After subcutaneous dissection, a 3.5 × 2.5 cm sized bulging ovoid mass was detected. The hernia sac exited through the external inguinal ring directly. Complete detachment was possible except for the neck portion which seemed to be connected with structures within the peritoneal cavity (Fig. 2A). The cystic mass hung free on an isolated fibrous pedicle that emerged from the mesentery in the vicinity of the round ligament. On section, the cyst was uniocular and contained bloody fluid. The wall was relatively thickened and firm. Its inner surface was covered with blood clots (Fig. 2B). Microscopically, the cystic wall resembled the gut wall (Fig. 2C). The wall was composed of two distinct muscle

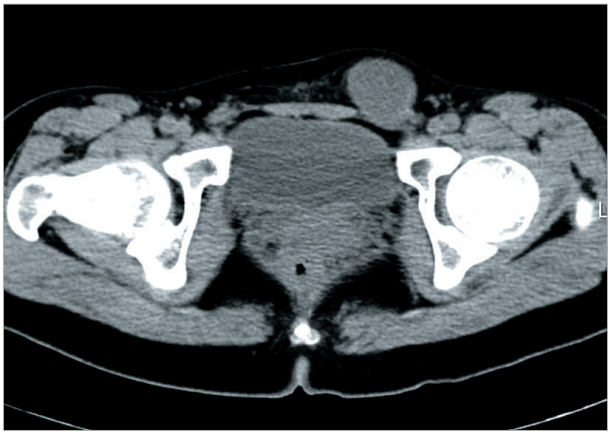

Fig. 1. Computed tomography scan showing a 3.5 × 2.5 cm sized cystic mass in the subcutaneous layer of the left suprapubic area.

layers with the presence of Auerbach's plexus (Fig. 2D). On examining the entire section of the cyst wall very carefully, no epithelial lining was found on the inner surface. The submucosa was slightly fibrotic. The diagnosis was a completely isolated enteric duplication cyst.

DISCUSSION

The diagnosis of an enteric duplication cyst requires fulfillment of the following histologic criteria: gastrointestinal mucosa, smooth muscle in the wall of the cyst, and attachment to the gastrointestinal tract.[1] The cyst can be stripped of its mucosal layer due to intracystic pressure or chemical ulceration. Ulcer-

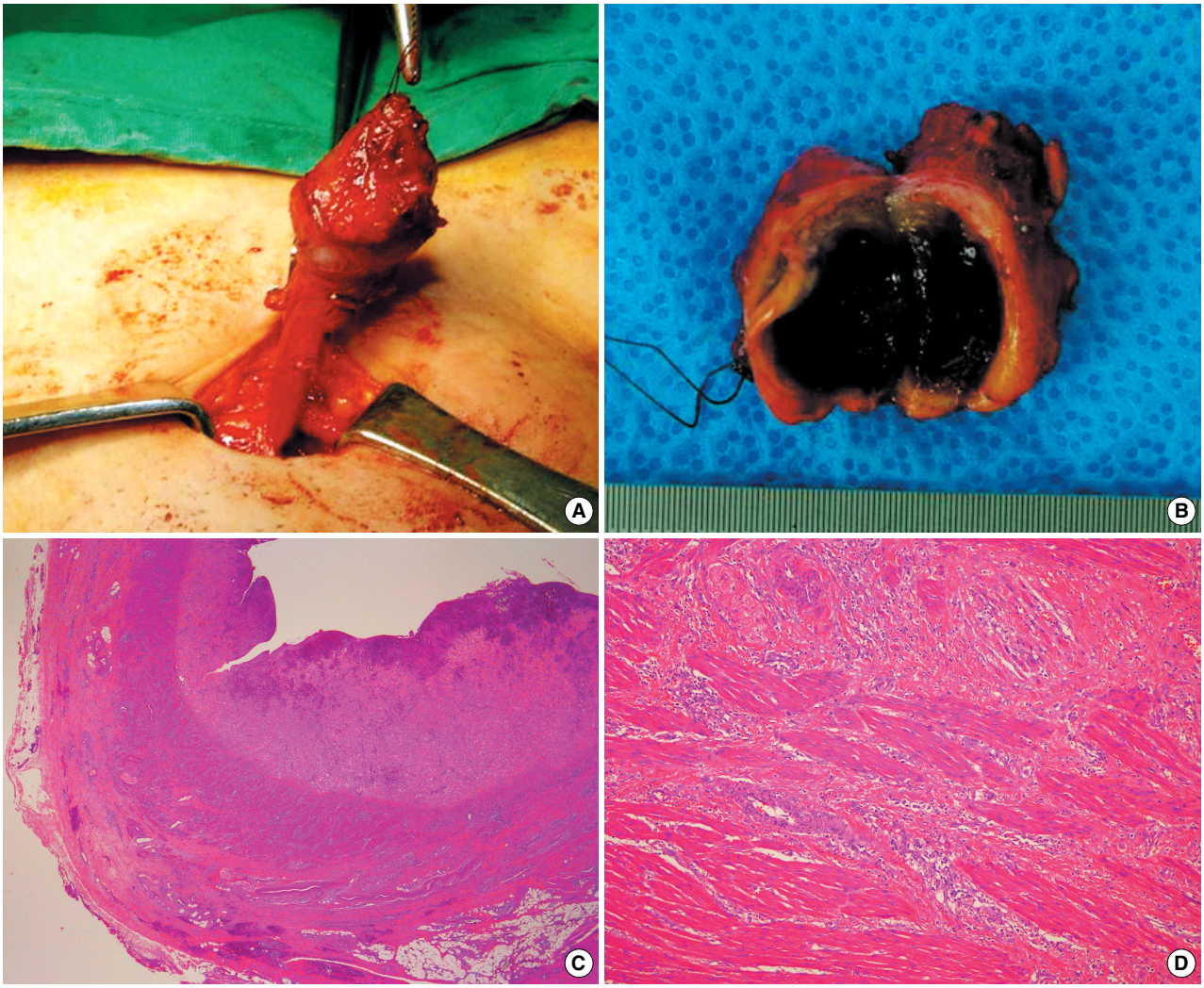

Fig. 2. (A) Intraoperative photograph of the cystic mass presenting a groin hernia. (B) Gross appearance of the specimen shows an intact cyst measuring 3.5 × 2.5 cm. On sectioning, the wall is found to be ulcerated and hemorrhagic. (C) The cyst shows that the wall comprises 2 distinct smooth muscle layers, submucosa, muscularis mucosa, and denuded mucosa. (D) The wall is composed of two distinct muscle layers with the presence of Auerbach's plexus.

ation, inflammation, and scarring are frequently reported in this anomaly.[1] Entirely denuded mucosa of enteric duplication cysts are rare, but there is the possibility of the entire mucosal layer being denuded.[1] In our case, ectopic gastric mucosa or pancreatic tissue was not found. But ectopic gastric mucosa or pancreatic tissue, which can be found in duplications, may cause ulceration, gastrointestinal bleeding or perforation. An enteric duplication cyst must be completely isolated from the adjacent alimentary tract. Hence, two distinct smooth muscle layers are a diagnostic clue for enteric duplication cyst. Barr *et al*.[2] suggest that when a double muscle layered wall is identified in a cystic structure, the diagnosis of enteric duplication cyst is confirmed.

Enteric duplication cysts are spherical or tubular structures, arising from some portion of the alimentary tract. They usually have a common blood supply with the adjacent segment of intestine, and an epithelial lining that resembles that of the adjacent alimentary tract. The Enteric duplication cyst completely isolated from the adjacent alimentary tract but the presence of typical histopathologic features of a duplication cyst would qualify for a diagnosis of a completely isolated enteric duplication cyst. Completely isolated enteric duplication cysts have independent blood supplies.

There are three main embryological theories for the development of enteric duplication cysts. According to the enteric bud theory proposed by Lewis and Thyng[3], buds of intestinal epithelium protrude into the lamina propria but maintain a connection with the bowel lumen. Bremer[4] postulated that the embryonic gastrointestinal tract outgrows the coelomic cavity by rapid enlargement followed by recanalization with intestinal epithelial cells. Ravitch[5] proposed that hindgut duplication represents partial twinning.

Isolated cystic duplications of the gastrointestinal tract are extremely rare. Steiner and Mogilner[6] proposed that the pathological events of a completely isolated enteric duplication cyst might have proceeded with torsion, or some vascular accident at the proximal end of the diverticulum. This event might have detached it from the intestine wall, and a separated duplication cyst had resulted.[6]

Although inguinal hernias commonly occur in males, they can be seen in females. Possible pathogenetic mechanisms in female patients are as follows. During embryologic development, the round ligament in women migrates from the retroperitoneum through the anterior abdominal wall to the inguinal canal along with a projection of processus vaginalis. The defect in the abdominal wall (internal inguinal ring) associated with this process represents an area of potential weakness through which an indirect inguinal hernia may form. Omentum, colon, small bowel, and bladder are the most common contents of groin hernias, although appendix, Meckel's diverticulum, fallopian tube, and ovary have been reported to herniate.

In 2006, Prada Arias *et al*.[7] reported that a neonatal male had an enteric duplication cyst resembling an umbilical cystic mass. But, an inguinal hernia as a manifestation of an enteric duplication cyst has never been reported.

In our case, an isolated enteric duplication cyst was passed through the internal inguinal ring and demonstrated that it can be a possible component of an inguinal hernia.

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