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Case Report
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Laparoscopic Resection of a Duodenal Duplication Cyst

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Abstract

Background: Duodenal duplications is rare disease especially in adult patients. The recommended treatment is complete surgical resection. We present a review of the current literature on duodenal duplications and focus on a case of duodenal duplication in a 33-year-old woman who was treated laparoscopically.

Methods: A PubMed search using the keywords "duodenal" and "duplication" was performed.

Results: Sixteen cases were included in our review; 56% were in men, with an average age at diagnosis of 44 years. All the patients presented with vomiting, 94% had abdominal pain, and only 37% reported weight loss. A palpable abdominal mass was found in one patient. The patients were examined using, on average, 3 different imaging techniques (63% received a computerized tomography scan; 56% underwent endoscopy; and 44% underwent an upper gastrointestinal series). The median cyst size was 7 cm, 63% were in the second duodenum, and 87% did not communicate with the lumen of the bowel. Eighty-one percent of the patients were treated via a laparotomy, 13% via endoscopy, and 6% via laparoscopy. Malignant transformation was observed in one patient.

Conclusion: Because of the risk of malignant transformation, complete resection of the duodenal duplication cyst is recommended. Based on a case treated at our institution, we show that laparoscopy is a promising alternative to a laparotomy, if anatomically possible.

Keywords: Duodenal duplication cyst; CT; MRI

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Background

Duodenal duplications are the least common type of enteric duplication. They represent 2% to 12% of all duplications of the gastrointestinal tract, and only 1 out of 100,000 births are affected by this rare congenital malformation [1]. Most duodenal duplications are found in the second duodenum, but they can also be found rarely in the first and third duodenum. A lack of knowledge of specific symptoms, associated with the scarcity of patients, can make diagnosis particularly challenging. Duodenal duplications are usually detected during childhood, and rare in adult patients [2]. The current recommended treatment is complete surgical resection of the duodenal duplication cyst. Nevertheless, surgical resection is occasionally impossible because of particular anatomical difficulties. In these situations, such cysts can be treated endoscopically. However, the optimal approach to the management of duodenal duplications remains unclear. We report an adult woman with duplication in the third duodenum who was treated laparoscopically, together with a review of the current literature, to clarify the optimal method for the diagnosis and optimal management of duodenal duplication.

Case Presentation

We report the case of a 33-year-old woman who had been diagnosed as having multiple sclerosis and she had persistent abdominal pain. The pain was localized in the right upper quadrant of the abdomen and radiated to both iliac fossa. The patient experienced post-prandial nausea and repeated episodes of vomiting. A palpable solid mass was found in the right upper quadrant of the abdomen. The patient weighed 60 kg, was 162 cm tall, and had a BMI of 22.86 kg/m^2. The laboratory tests were all within the normal limits. An abdominal ultrasound was performed which revealed a heterogeneous mass of 4.5 cm in diameter that was anechoic, had a well-defined wall, and was situated in the mesentery. An abdominal-pelvic Computerized Tomography (CT) scan was performed for further examination. The mass was well identified in the arterial phase with a significantly enhanced lesion wall and inflammatory infiltration of the surrounding adipose tissue. In the venous phase, the interior of the mass was enhanced, and the same considerable enhancement of the parietal structure around the lesion was observed (Figure 1.1: CT scan, venous phase). Complementary CT imaging was also performed after the administration of an

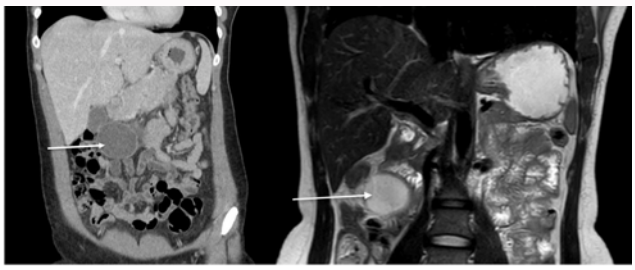

Figure 1-1: Computed tomography image showing a large extraluminal cyst (size: 46 mm × 38 mm × 39 mm) in the third part of the duodenum (arrow).
Figure 1-2: Magnetic resonance image of a large cystic lesion measuring 46 mm × 38 mm × 39 mm (arrow).

oral contrast agent. No evidence of a communication between the mass and the duodenum was seen. Based on the CT scan and the ultrasound results, two possible diagnoses were suspected: a non-communicating duplication cyst of the duodenum, or a mesenteric cyst. A mesenteric cyst was less likely, considering the atypically thick wall surrounding the mass. An abdominal Magnetic Resonance Imaging (MRI) examination was also performed to investigate the detailed characteristics of the cyst. The lesion measured an impressive 46 mm × 38 mm × 39 mm and was entirely composed of fluid. The wall of the lesion was thick. The origin of the cyst could not be identified (Figure 1.2). The patient then underwent an Endoscopic Ultrasound (EUS) examination that revealed a partially cystic, hyperechoic lesion 44 mm × 46 mm in size. The lesion had a regular outline that was similar to that of the intestinal wall. The mass was in close contact with the third duodenum but was extra-duodenal. The EUS examination did not show any signs of the compression of the second or third duodenum. Because of the persistent and invalidating symptoms, the laparoscopic removal of the cyst was suggested. The possibility of conversion to a medial laparotomy was also explained to the patient. She agreed to undergo the intervention and signed the informed consent. We performed a laparoscopic intervention that confirmed the presence of a large non-communicating duplication cyst in the medial wall of the third duodenum. The mass did not obstruct the pancreatic duct or the common bile duct. The sheaths surrounding the cyst were carefully detached. Once the dissection of the surrounding tissue was completed, the cyst was cleaved from the duodenal wall using an EndoGIA™ stapler and was entirely excised from the duodenal wall.

The patient's post-operative course was uneventful. The patient was discharged 7 days postoperatively. Figure 2.1 shows the excised lesion, which consisted of a circular cystic mass measuring 7.2 cm × 6.5 cm × 5.5 cm in size and was covered with a smooth serous layer. The cyst was filled with a partly mucoid and viscous beige liquid. The wall of the lesion measured 0.6 cm in thickness on average, as shown in Figure 2.2. Microscopically, as shown in Figure 2.3, the inner lining of the cyst was composed of a single layer of cubic or prismatic cells with a ciliated aspect in some areas. In some places, the coating was abraded and was replaced with foamy macrophages. The wall of the lesion resembled that of an intestinal wall and was composed of a muscularis mucosa, a submucosa, and a layer of smooth muscle. No signs of malignancy were detected. The diagnosis of a duplication of the duodenum was consequently confirmed (Figure 2.3).

Literature Review

A review of the current literature was undertaken in February

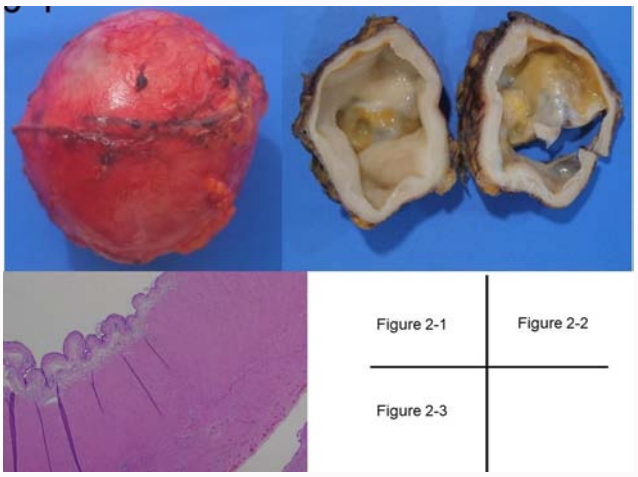

Figure 2-1: Macroscopic photograph of the duodenal duplication cyst (7.2 cm × 6.5 cm × 5.5 cm).
Figure 2-2: Macroscopic image: The wall was flexible and up to 0.6-cm thick. The inner coating of the cyst was smooth and white, with localized polypoid deposits on the mucosal aspect.
Figure 2-3: Microscopic image: The previously described macroscopic structure (Figure 2-2) was composed of a thin intestinal wall. The inner layer consisted of a thin sheet of connective tissue covered by a simple epithelium containing cuboidal and prismatic cells that were occasionally ciliated.

2018 by searching for articles on PubMed using the key words "duodenal" and "duplication". This search yielded 486 articles. All the articles that were not written in English were excluded. Papers that were not accessible on the Internet were also excluded. Cases of duplications of the duodenum in patients under the age of 18 years were excluded. Furthermore, all the patients who were included in this study had been described as having symptoms of gastric outlet obstruction. We found 16 cases of duplications of the duodenum that met all the above criteria. For each of the cases described in these articles, we noted the age and the sex of the patient and the symptoms caused by the duodenal duplication. Data found during the clinical examination as well as the types of imaging that were used to diagnose and characterize the duplication were also recorded. The size of the cyst, the part of the duodenum to which the cyst was attached, and whether the cyst was in communication with the lumen of the duodenum were also determined. Particular attention was given to the type of treatment that was performed for the removal of the duodenal duplications. Finally, findings concerning histology, malignancy and surgical outcomes were also described. The collected data are summarized in Table1, S1-S5. Of the 16 patients who were included in the study, were 56% were male, and the median age was 44 years (range: 21-73 years) (Table S1 and S2).

Symptoms

All the patients had episodes of vomiting (Table S1). Other symptoms were abdominal pain (94%), nausea (62%), loss of weight (37%), and early satiety (19%). Only 31% of the cohort manifested abdominal pain, nausea, weight loss and vomiting. One patient also reported gastro-esophageal reflux, and another patient complained of malodorous burping and thoracic pain. One duodenal duplication caused a peptic ulcer (Table S3). Upon clinical examination, the majority of patients expressed upper abdominal tenderness (56%). One patient presented with a soft palpable mass, and another had diminished abdominal sounds. Fifty percent of the patients had no findings upon clinical examination (Table S2).

Table 1: Treatment and histological findings of duodenal duplication cysts.

Author	Treatment	Post-op	Histology	Malignancy
Dave et al. [8]	Endoscopic polypectomy	sp	Duodenal duplication cyst Gastric pyloric mucosa	no
Fried et al. [15]	Laparotomy	sp	Duodenal duplication cyst	no
Gjeorgjievski et al. [12]	Endoscopic deroofing	sp	No histology	no
Jiménez et al. [23]	Laparoscopic duodenotomy	sp	Duodenal duplication cyst	no
Jo et al. [4]	Laparotomy with pylorus preserving duodenopancreatectomy	sp	Duodenal duplication cyst	no
Kovolinka et al. [24]	Laparotomy with cystoduodenotomy	sp	Duodenal duplication cyst	no
Mc Ardle et al. [25]	Laparotomy with duodenotomy	sp	Duodenal duplication cyst Containing blood and bile Lined with histiocytes and no epithelial lining	no
Rotondo et al. [17]	Laparotomy with cystoduodenotomy	sp	Duodenal duplication cyst	no
Salemis et al. [26]	Laparatomy with excision of the anterior wall and marsupilization of the posterior wall	sp	Duodenal duplication cyst	no
Sefa et al. [27]	Laparotomy with duodenotomy	sp	Duodenal duplication cyst Ectopic pancreatic mucosa and metaplastic changes	yes
Tang et al. [28]	Laparotomy with pylorus preserving duodenopancreatectomy	sp	Duodenal duplication cyst	no
Taura et al. [29]	Laparotomy with longitudinal duodenotomy	sp	Duodenal duplication cyst stomach and duodenal epithelium Gastric and duodenal epithelium	no
Kirtley et al. [16]	Laparotomy with longitudinal duodenotomy	sp	Duodenal duplication cyst	no
Uzun et al. [20]	Laparotomy with cystoduodenotomy	sp	Duodenal duplication cyst	no
Bar-Ziv et al. [14]	Laparotomy with duodenotomy	sp	Duodenal duplication cyst	no
Thompson et al. [10]	Laparotomy with cyst incision	sp	Duodenal duplication cyst	no

Imaging

The patients were examined using 3 different imaging techniques, on average. A CT scan (63%), endoscopy (56%), upper gastro-intestinal series (44%), abdominal ultrasound (44%) and MRI examination (25%) were performed in the majority of the patients. Nineteen percent of the cohort had undergone a barium meal test and another 19% had undergone a EUS examination. Two patients underwent Endoscopic Retrograde Cholangiopancreatography (ERCP), and one patient underwent a Magnetic Resonance Cholangiopancreatography (MRCP) (Table S4).

Characteristics and locations of duodenal duplication

The median size of the cysts was 7 cm (range: 1 cm to 15 cm). The cysts were mainly located in the first and second duodenum (87%). Some of the duodenal duplications were located in more than one part of the duodenum. Most of the patients had duodenal duplication cysts that did not communicate with the duodenal lumen: 87% had non-communicating cysts, and only 13% had cysts that were in communication with the duodenal lumen (Table S5).

Surgical treatment

Thirteen patients (81%) were treated via laparotomy, and 9 patients (56%) underwent a cystoduodenostomy with laparotomy. A pylorus-preserving pancreatico-duodenectomy was performed *via* a laparotomy in two patients. One of the cysts was approached via a laparotomy and was incised but was not entirely removed. In another patient who underwent a laparotomy, the anterior wall of the cyst was excised and the posterior wall of the cyst was subjected to marsupialization. Only one patient underwent a laparoscopic duodenectomy (Table 1).

Endoscopic treatment

Only two duodenal duplications were treated endoscopically. One of them was removed by deroofing the cyst, and the other was removed via a polypectomy (Table 1).

Post-operative course

All the patients had an uneventful post-operative recovery and reported a complete resolution of their symptoms following the treatment of their duplication cysts.

Pathological findings

A histological diagnosis of a duodenal duplication cyst was made in 15 patients (94%), and the presence of a duodenal mucosa, a layer of smooth muscle and a submucosa was confirmed in these patients. Seventy-five percent of the cysts did not contain any ectopic tissue of another nature. However, the presence of an ectopic gastric pyloric mucosa was confirmed in 2 cases and an ectopic pancreatic mucosa was found in 1 case. Only one patient exhibited metaplastic changes in their duodenal duplication cyst (Table 1).

Discussion

We report a rare case of an adult patient who was diagnosed as having a duodenal duplication after experiencing symptoms of gastric outlet obstruction. The cyst was resected using a laparoscopic procedure. To our knowledge, this is the first report of the resection of a duodenal duplication using a laparoscopic technique. Duplications of the Gastrointestinal (GI) organs affect 1 out of 4500 to 1 out of 10000 births [3]. By definition, they are composed of a layer of smooth muscle and are lined with a GI epithelium [4]. These entities can be located in any part of the alimentary tract [5]. They can be intramural

or intraluminal [6]. GI duplications are most commonly found in the small bowel (47%), whereas duplication in the duodenum is more atypical [7-11]. The symptoms caused by this type of pathology are usually nonspecific. Duodenal duplications can be associated with recurrent episodes of pancreatitis, jaundice [4], and obstruction of the bowel with abdominal pain, vomiting, loss of weight, and nausea [1,3]. A review of current literature revealed only 16 adult patients with symptoms of gastric outlet obstruction. These findings confirm that duodenal duplication cysts only occasionally manifest with vomiting and gastric outlet obstruction in adults. Chen et al. [1] reported that most of the patients (72%) who experienced episodes of vomiting because of duodenal duplication cysts were younger than 10 years old. Chen et al. [1] also noted that 44% of the cysts reported in past articles measured between 2.0 cm and 4.0 cm. Of the patients included in this literature review, which was limited to patients with symptoms of gastric outlet obstruction who were older than 18 years of age, a great majority of the cysts were larger than 4.0 cm and the average cyst size was 9 cm. Patients who are older than 18 years might have larger cysts, and these larger cysts might be associated with symptoms of gastric outlet obstruction. Duodenal duplication cysts have also been found to communicate with the pancreatico-biliary duct in some cases (30% of cases according to Gjeorgjievski et al. [12]). It was previously stated that duodenal duplication cysts are usually composed of clear fluid, but some have also been observed to contain bile, pancreatic fluid, or calcified stones [13] or enteroliths [14]. According to Antaki et al. [3] the diagnosis of duodenal duplication cysts can be achieved through the exclusive use of MRCP and EUS. The solid or cystic nature of the lesion can adequately be assessed using EUS, and the intramural, extramural or tubular features of the mass can also be determined. The composition of the lesion wall can be identified using EUS. MRCP is particularly helpful for evaluating whether the cyst is in communication with the pancreatico-biliary tract [1]. A recent report described the use of Meckel technetium scanning to visualize the ectopic gastric mucosa in the cyst. Since the gastric mucosa in duodenal duplication cysts can be associated with malignancy, Meckel technetium scanning might be useful for reinforcing the surgical indications [12]. The differential diagnosis of a duodenal duplication cyst includes mesenteric cysts; however, this type of cyst is composed of a thin layer of flattened epithelium, whereas duodenal duplication cysts have a much thicker wall. Duodenal duplication cysts can also be confused with other cystic masses such as choledochoceles, although these structures are lined with biliary mucosa and do not have a layer of smooth muscle [1,13]. Duodenal duplication cysts have also been mistaken for pancreatic pseudocysts [15], leiomyomas [16], and cystic dystrophies of the duodenum [17]. The most common complication associated with duodenal duplication cysts is pancreatitis [4]. The rupture of a duodenal duplication cyst has also been reported [18,19]. The treatment of duodenal duplication cysts remains controversial. Some authors have reported that an endoscopic approach is ideal [3,9,10], while others consider that duodenal duplication cysts can only be treated using total resection [11,20,21]. The endoscopic treatment of a duodenal duplication cyst consists of the incision of the cyst, allowing the contents of the cyst to drain into the duodenum. Some duodenal duplication cysts have been subjected to malignant transformations of their intestinal wall [22]. The malignant transformation of duodenal duplication cysts can be associated with the presence of ectopic gastric mucosa in the cyst, and 15% of duodenal duplication cysts have been found to contain ectopic gastric mucosa [3]. This result explains why the total resection of duodenal cysts has been recommended in medical literature. In our review, only one patient had a malignant transformation; however, the transformation was not associated with ectopic gastric mucosa. The malignant transformation of duodenal duplication cysts can also occur as a result of chronic inflammation [3]. Different types of procedures can be performed for the treatment of duodenal duplication cysts, such as a cystoduodenostomy or a pancreatico-duodenectomy. A laparoscopic approach can be used as an alternative to a laparotomy. However, the exclusive laparoscopic resection of a duodenal duplication without the resection of other organs has never been reported. The majority of patients included in our review underwent a cystoduodenostomy via a laparotomy. Only one patient underwent the removal of a duodenal duplication cyst laparoscopically accompanied by a resection of the duodenum [23-29]. Our objective was to remove the cyst laparoscopically. Duodenal duplication cysts are usually situated along the inner curvature of the duodenum, making it difficult to remove the duodenal duplication cyst alone [5]. In our case, the duodenal duplication cyst was located along the exterior of the duodenal curvature, and its resection without touching the duodenum was considered to be feasible preoperatively. All the patients included in our review as well as the presently reported patient had uneventful recoveries. However, the observations made in the present review were based on a small number of patients and must be interpreted with caution.

Conclusion

Duodenal duplication cysts are a rare pathology that can be difficult to diagnose. This literature review suggests that EUS, MRCP and MRI are useful for diagnosis. Because of the risk of malignant degeneration, we recommend the total resection of duodenal duplication cysts. We also recommend the laparoscopic resection of the cyst, if possible.

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