Case Report

Duodenal Duplication Cyst: A Rare Case Report

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ABSTRACT

Gastrointestinal duplication cysts are infrequent congenital abnormalities. 2% to 12% of these abnormalities are localized in correspondence of the duodenum. This disease rarely remains asymptomatic until adulthood. We present a case of A 62-year-old woman with intermittent upper abdominal pain and nausea, describing the diagnostic and therapeutic route.

Key-words: *CT, Duodenal cyst, Duodenum, Endoscopic resection*

Gastrointestinal (GI) duplication cysts are infrequent congenital abnormalities that may occur throughout the gastrointestinal tract [1]. The small intestine, large intestine and esophagus are the sites most commonly affected [2]. The prevalence of GI duplication is 1:4500 to 1:10000 in the general population [2] and the duodenal duplication cyst represents a percentage ranging from 2% to 12% of digestive tract duplications and has an estimated prevalence of less than 1 per 100000 live births [1]. The most common type of duodenal cyst is the cystic and non-communicating type, usually located at the medial border of the second part of the duodenum and extending anteriorly or posteriorly [3]. These are mostly diagnosed in infancy and childhood [4].

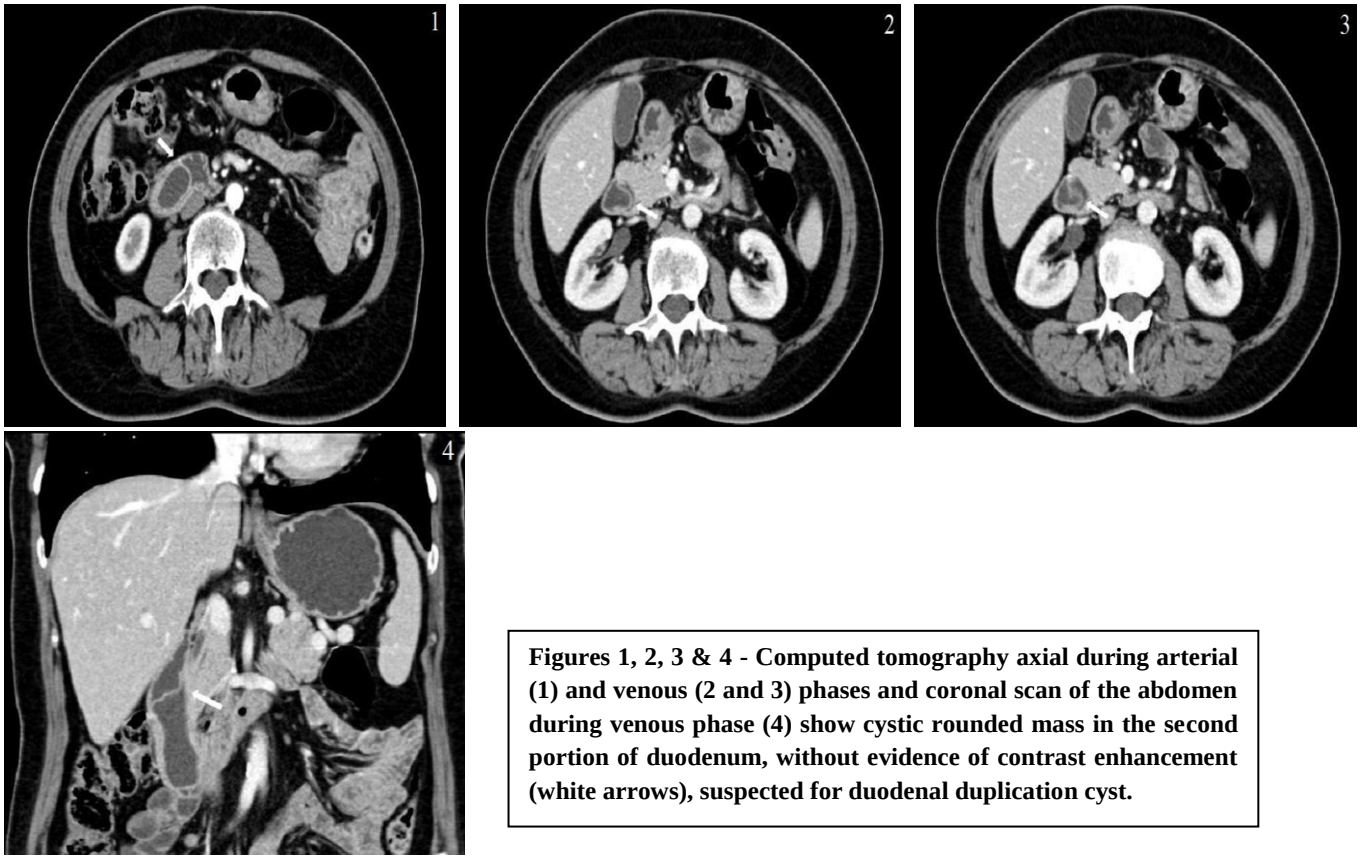
In rare cases, they can remain asymptomatic until adulthood, and 38% of patients are diagnosed after the age 20 years [4]. Clinical manifestations of duodenal duplication cyst include abdominal pain (100%), nausea/ vomiting (52%), pancreatitis (65.7%), cholestasis or hepatitis (26.3%), growth delay or weight loss (10.5%), GI bleeding (7.9%), intussusception/ cyst infection (5.2%) [1].

Malignant change is a rare complication of GI duplications in adult. In 30 cases reported in the English literature, 21 were adenocarcinomas, 4 squamous cell carcinomas, and 5 carcinoid tumours [2]. Duodenal duplication cysts are generally benign lesions; however, three cases of malignant tumours arising from inside have been reported [4]. Ectopic tissue (gastric, squamous, transitional, ciliated mucosa, pancreas, ganglion cells) can be found in its wall [2].

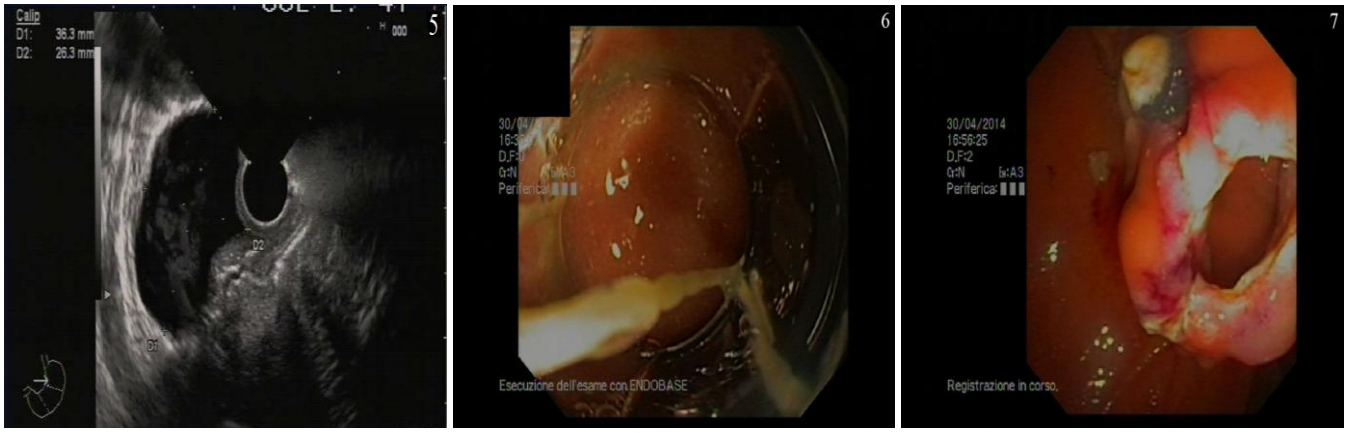
CASE REPORT

A 62-year-old woman presented with intermittent upper abdominal pain and nausea. Her physical examination and laboratory data showed normal findings. CT scan abdomen in arterial (**Fig. 1**) and venous (**Fig. 2, 3 and 4**) phases showed a 36x22 mm rounded cystic mass with fluid content and thin peripheral wall, located within the lumen of second duodenal portion. Neither the arterial (**Fig. 1**) or venous (**Fig. 2, 3 and 4**) phases showed contrast enhancement of the mass.

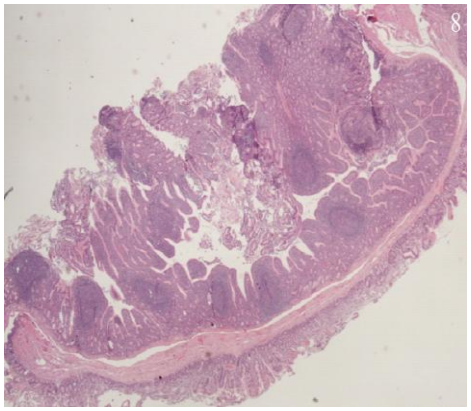
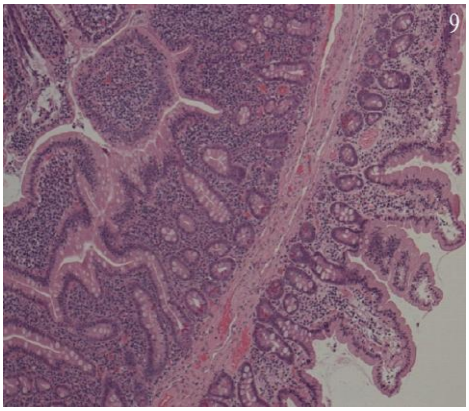
Endoscopic ultrasound (EUS) demonstrated a bulge in the wall with regular mucosa, in the ampullary region, for the entire length of upper knee of the duodenum and in second duodenal portion. This region appears as cystic formation with internal sediment that affects the ampullary region without determining obstruction of the pancreatic duct and common bile duct (**Fig. 5**). As a result of these examinations, a duodenal duplication cyst has been diagnosed. With the patient's written informed consent, an endoscopic partial resection of cystic wall was performed.



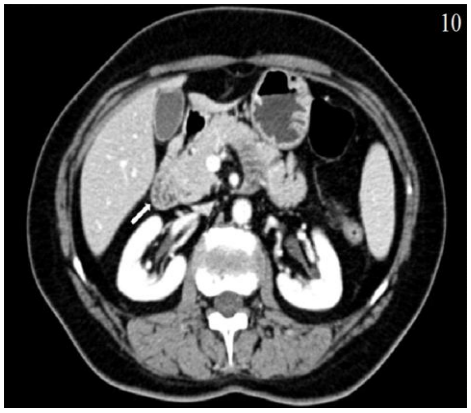
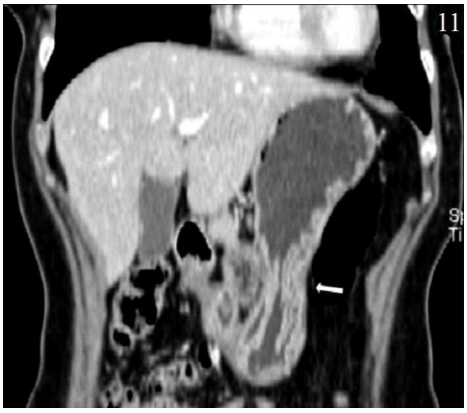
Figures 1, 2, 3 & 4 - Computed tomography axial during arterial (1) and venous (2 and 3) phases and coronal scan of the abdomen during venous phase (4) show cystic rounded mass in the second portion of duodenum, without evidence of contrast enhancement (white arrows), suspected for duodenal duplication cyst.



Figures 5-6-7: Endoscopic ultrasonography showing cystic mass with dimension of 36 x 26 mm with internal sediment in the ampullary region, without obstructing the major pancreatic duct and the common bile duct, compatible with duodenal duplication cyst (5). Endoscopic images show three loops around the prominent portion of the cyst (6), and the conventional polypectomy of the bound portion (7).

Figures 8 & 9 - Histological examination showing intestinal epithelium lines, intestinal duplication with the presence of a smooth muscle coat

Figures 10 & 11 - Computed tomography axial (10) and coronal (11) scan of the abdomen in the venous phase shows the results of endoscopic resection (white arrows).

Under vital signs and electrocardiography monitoring, after CO_2 insufflation, we reached the second portion of duodenum with gastroscope GIF-Q165. After viewing the cyst, 3 loops were positioned on the most prominent portion (Multibrand mucosectomy DT-6-5 Fr) (**Fig. 6**). The ligated portions was snared and resected as in a conventional endoscopic polipectomy, witnessing the onset of clear liquid and plugs (**Fig. 7**). Through the vision with a duodenoscope TJF-160R, coagulation of the edges of cysts was performed with marsupialisation and sphincterotomy with the use of a pre-cut needle. Histological examination of the resected specimen showed intestinal epithelium lines, intestinal duplication with the presence of a smooth muscle coat (**Fig. 8 and 9**). At 3 months follow-up, patient had no symptoms. Laboratory data presented a slight rise of serum amylase (106 UI, NV - 0-94 UI) and lipase (83 UI, NV - 0-60 UI). A contrast enhanced CT abdomen in venous phase (**Fig. 10 and 11**) shows the results of endoscopic mucosectomy.

DISCUSSION

Ultrasound can be useful for the diagnosis of duplication cyst. The duplication content may vary because of bleeding, chronic inflammation, or infection; therefore, it has poor diagnostic value. Inside the duplication cyst may be present enteroliths, most likely related to chronic stasis and alkalinity of intestinal content or the hemorrhage can determine the formation of echogenic debris within the cyst [5,6]. On the other hand, sonographic appearance of the cyst wall can be highly suggestive of the diagnosis of duplication when a multilayered appearance with an echogenic internal mucosal layer and a hypoechoic intermediate muscular layer are present [5]. Ultrasound can usually define the cyst but as a rule, may not show the exact source. The exception is the choledochal cyst where intimate relationship to the biliary tree and pancreas makes the diagnosis possible [6].

Computed tomography (CT) scan abdomen is a useful tool in order to detect duodenal duplication cyst and to precisely define its size and location. Duodenal duplication cyst typically appears as round cystic mass, surrounded by a thin wall and located at the level of the duodenum. The differential diagnosis includes choledochocele, pancreatic pseudocyst and intraluminal diverticulum. The density of duodenal duplication cyst is similar to the density of water. The attenuation increases when it contains hemorrhagic, proteinaceous or purulent material. Cases have been

described in which tumoral nodule was visible in within the duplication. Vegetations and wall nodular mass should be of concern since malignant transformation could be detected in duplications [5].

Magnetic resonance imaging (MRI) may be useful as a non-invasive technique for the diagnosis and accurate preoperative assessment of the cystic mass. Conventional spin-echo T1- and T2-weighted MRI provide accurate information on configuration, location, size and internal content of the mass. Unfortunately on conventional spin-echo MRI, relation between the duplication of duodenum with adjacent organs cannot be clearly demonstrated. To differentiate it from adjacent organs, spin-echo fat-suppressed T1-weighted MR images can be utilized before and after intravenous and oral administration of Gadodiamide Diethylenetriaminepentacetate (Gd-DPTA). Cystic mass has low signal intensity relative to the pancreas (which appears hyperintense) on T1-weighted fat-suppressed images; so, cyst can be easily distinguished from the head of pancreas. Moreover, on fat-suppressed T1-weighted MR imaging after oral administration of Gd-DPTA, we can appreciate a good delineation of the lesion and a better visualization of the extent of stenosis [7].

EUS examination is very important for the differential diagnosis, especially to differentiate it with the choledochocele and to distinguish between solid and cystic lesions [3]. EUS shows duplication cysts as anechoic, homogenous lesions with regular margins arising from the submucosal layer or extrinsic to gut wall; although, a hypoechoic pattern can also be seen with a duplication cyst. Duplication cysts may contain thick mucinous material, septations, fluid levels, debris and sometimes, detached ciliary tufts which could be diagnostic [3].

A meta-analysis of the literature between 1999 and 2009 reported total 47 cases of duodenal duplication cysts [1]. Treatment has classically involved surgical resection because of cases of malignancies have been reported in literature [2]. Endoscopic therapy has been used as an alternative to surgery in selected cases [8]. Marsupialization with partial resection of the cyst and intracystic biopsy is a safe and effective technique, with excellent long-term results. This allows the histological study which can detect any malignant changes [8]. Antaky et al suggested repeat endoscopy and biopsy follow-up at 6 and 12 months after treatment [8].

CONCLUSION

Endoscopic resection of a duodenal duplication cyst represented a safe procedure and can be considered a definitive treatment for these patients.

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