# **Case Report**

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# Gastric duplication cyst: rare cause of upper gastrointestinal bleeding in a young adult, with a review of literature

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#### **ABSTRACT**

Duplication cyst along the alimentary canal is a congenital anomaly which is rare and usually incidentally found at endoscopy or radiological imaging. It can develop anywhere along the alimentary canal with only 4% occurrence in the stomach. Only few cases of gastric duplication cysts were reported to be symptomatic along with its complications. Diagnostic modalities include oesophageo-gastroduodenoscopy (OGDS), endoscopic ultrasound (EUS), computerized tomography (CT) scan and magnetic resonance imaging (MRI). However, the best option to confirm the diagnosis is complete resection of the lesion. We are reporting about a teenage girl whereby she presented with sudden episodes of hematemesis, which led to a syncopal attack. She also complained of intermittent central abdominal pain for a month prior to her presentation. Physical examination was unremarkable. However, her blood investigation showed a significant drop in hemoglobin. The patient had an endoscopy during which a submucosal lesion at the greater curvature was identified and the provisional diagnosis we had was a gastrointestinal stromal tumor (GIST). CT scan of the abdomen was also done showing an endoluminal cystic gastric lesion measuring approximately  $2\times2\times2$  cm. The lesion was confined to the stomach with no local infiltration of the adjacent structures. Decision was made to proceed with a laparoscopic wedge resection after outweighing the risk and benefits of surgery. Gastric duplication cyst is a rare anomaly. Despite its rarity, it should be included as a differential when we encounter a submucosal lesion due to its tendency for malignant transformation and other complications.

Keywords: Duplication cysts, GIST, Laparoscopy, EUS

## INTRODUCTION

Submucosal lesions can occur anywhere along the alimentary tract and are most commonly found at the esophagus and stomach. These lesions may be benign or malignant.<sup>1</sup>

In a study by Polkowski et al he revealed that submucosal lesion arising from the stomach were the second most common based on its anatomical location.<sup>2</sup>

Gastric submucosal lesions include smooth muscle neoplasm (54%), heterotopic pancreas (16%), cystic lesions (9%), lipoma (5%), carcinoid (3%), other lesions

(10%) such as fibroma, fibrovascular polyp, granuloma, inflammatory fibroid polyp, intramural metastasis, varix).<sup>2</sup>

However, it remains unclear when we encounter such lesions if a EUS guided-fine needle aspiration cytology (EUS-FNAC) or surgical resection would be best option.<sup>3</sup>

Gastric duplication cyst was among the rare submucosal lesions and is usually asymptomatic. Only a small number of cases present with complications for example hemorrhage, perforation, malignancy or complete gastric outlet obstruction. These cysts are usually incidental findings upon radiological imaging or endoscopy.<sup>4-5</sup>

This case report is about our encounter with a submucosal lesion in which our provisional diagnosis was a GIST but histology came back as a gastric duplication cyst.

#### **CASE REPORT**

A 14-year-old girl was incidentally diagnosed with a gastric duplication cyst confirmed on histopathology following a laparoscopic wedge resection. This young girl initially presented with 3 episodes of hematemesis within 24 hours, which led to a syncopal attack. She did experience intermittent central abdominal pain for a month prior to her presentation. She was otherwise a healthy teenager with no other comorbidities. On examination, she did not have any abdominal tenderness and aside from a low hemoglobin count (9 to 7 g/dL), her renal profile, coagulation profile was unremarkable.

Given the extent of hematemesis, the patient had an OGDS during which a submucosal lesion (Figure 1 A, B C and D) at the greater curvature was identified. There was no active bleeding evident during the procedure. The provisional diagnosis was a GIST.

An outpatient early CT scan of the abdomen and pelvis was then arranged showing an endoluminal cystic gastric lesion measuring approximately 2×2×2 cm (Figure 3 A and B). The lesion was confined to the stomach with no local infiltration of the adjacent structures.

After the scope and CT imaging, the patient was given another clinic appointment to further discuss about surgical options. After discussion with the caretaker and the patient, decision was made to proceed with a laparoscopic wedge resection.

She was admitted one day prior to surgery whereby we did a OGDS to mark the lesion. The lesion was marked with methylene blue stain (Figure 2 A and B) to facilitate the resection. Intraoperatively, the marked lesion was not visible laparoscopically. As a result, an on table OGDS had to be done to locate the lesion that required resection. Wedge resection was done with a margin of 1 cm laparoscopically. A polypoid lesion measuring  $3\times2\times2$  cm at the greater curvature was resected.

Histopathologic examination of the lesion demonstrated a 3cm cyst lined by gastric alveolar epithelium with intervening areas of simple columnar epithelium with apical mucous and cilia. In other area, the cyst wall was made up of fibrous stroma covered by aggregates of foamy macrophages. No heterotopic pancreatic tissue is observed (Figure 4). There were no features suggestive of intestinal metaplasia or malignancy.

She had an uneventful recovery and was discharged home after 3 days of hospital stay. Upon follow up after surgery, she was well with no evidence of complication.

She also claims that she has been asymptomatic since surgery was done.









Figure 1 (a-d): Endoscopy of a lesion at the greater curvature with smooth mucosa surface.

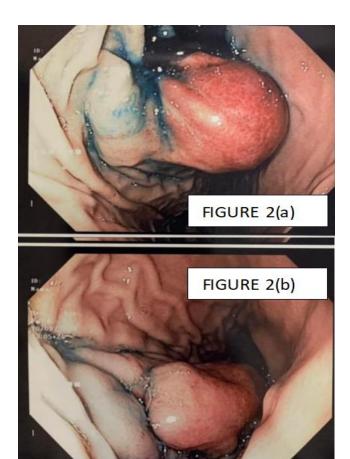


Figure 2 (a and b): Endoscopy of lesion marked with methylene blue stain.

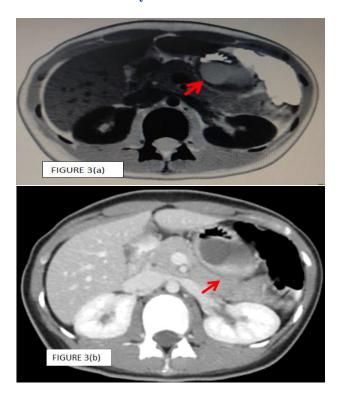


Figure 3 (a and b): CT scan of the abdomen of an endoluminal cystic gastric lesion measuring approximately 2×2×2 cm (red arrow).

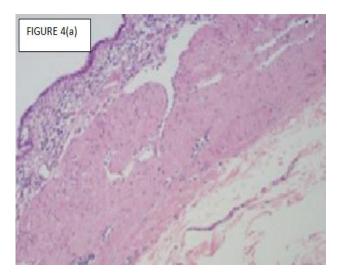


Figure 4: Microscopy of cystic lesion lined by gastric alveolar epithelium with intervening areas of simple columnar epithelium with apical mucous and cilia. Cyst wall was made up of fibrous stroma covered by aggregates of foamy macrophages.

## DISCUSSION

Duplication cysts are very rare and it is usually benign. Only a glimpse of cases had malignant transformation and its mechanism is still poorly understood.<sup>6</sup>

In a study by Zhu et al they reported a case series on 3 patients with malignant transformation of duplication cyst. All 3 patients were above the age of 40 and biopsy of the cyst was done prior to definitive surgery. Among the 3, 2 patients' diagnosis was confirmed to be adenocarcinoma only after surgical resection as biopsy was inconclusive.<sup>7</sup>

This congenital anomaly can be found anywhere along the alimentary tract and most commonly found in the proximal small intestine.<sup>8</sup>

Rowling et al on her observations on gastric cysts in the British journal of surgery stated that this cyst is most frequently found at ileum (35%), the oesophagus (19%), the jejunum (10%), the stomach (4%) and the colon (7%).

Gastric duplication cysts are the most uncommon as compared to the other anatomical locations of duplication cyst. Most cases were found to be female predominant (8:1) and being diagnosed within the first 3 months of life and rarely after 12 years of age. <sup>10</sup>

These cysts are usually asymptomatic and are typically discovered incidentally upon radiological imaging or during endoscopy. Some patients tend to present with abdominal pain, vomiting, dysphagia, dyspepsia and even anemia. There are few cases that presents with complications such as hemorrhage, perforation, malignancy or complete gastric outlet obstruction

depending on location or type of ectopic tissue present.<sup>11</sup> Some cysts may consists of gastric and pancreatic ectopic tissue. Hence presenting with features of peptic ulcer disease and even pancreatitis depending on the location of the cyst.<sup>12,13</sup>

Endoscopically, this cyst grossly appears as a smooth and regular bulge and most commonly located at the greater curvature of the stomach. Occurrence at the lesser curvature is only around 5.5%. <sup>14</sup> These macroscopic features often cause a misdiagnosis as its appearance may be similar to a GISTs.

Other modalities to diagnose this cyst includes EUS, CT scan and MRI.

EUS is currently the best imaging method for diagnosis and as it is able to differentiate cystic from solid lesions.<sup>3</sup> This however is limited to certain cases as the imaging is dependent on the cystic contents as the contents ranges from free-flowing fluid to think proteinaceous material.<sup>15,16</sup>

Takahara et al published a paper on imaging modalities for gastric duplication cyst and noticed that misdiagnosis from CT scan was ranging from 43 to 70% of cases. MRI was also shown to be inferior to EUS.<sup>17</sup>

Therefore, in most of the cases that was reported, a surgical resection with histopathological examination on the specimens were required to obtain a definite diagnosis.

Gastric duplication cyst may sometimes be misdiagnosed as a GISTs due to the similarities in their morphological feature upon endoscopy. Hence, EUS-fine-needle aspiration (EUS-FNAC) was recommended but its significance is still in debate. 3,18,19

In this patient, a complete resection of the lesion was justified by the patients' symptoms and unknown risk of malignant transformation. Modes of surgery is dependent upon the surgeon's preference as well as patient factors.

## **CONCLUSION**

In conclusion, duplication cysts of the stomach should be considered as one of the differentials when we encounter submucosal gastric masses despite the rarity. Complications such as bleeding, perforation, obstruction and malignant transformation has been reported. As a result, surgical excision should be performed to avoid such complications and especially in symptomatic patients.

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