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# Inflammatory Myofibroblastic Tumor Mimicking Gastric Duplication Cyst: A Dilemma in Clinical and Radiological Diagnosis

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***Authors' contributions***

*This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.*

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## ABSTRACT

A gastric duplication cyst is a rare congenital malformation of the foregut but inflammatory myofibroblastic tumor (IMT) is a more rare diagnosis. These are mostly asymptomatic and diagnosed incidentally on imaging done for some other cause. It is very difficult to differentiate between these two conditions based on clinical and radiological studies done preoperatively. Endoscopic Ultrasound with Fine Needle Aspiration (EUS-FNA) allows us to correctly diagnose these conditions. However, EUS-FNA carries a high risk of iatrogenic rupture of these lesions with

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significant infective potential. We present a case of IMT presenting as a Gastric duplication cyst with a brief review of the literature.

*Keywords: Gastric duplication cyst; IMT; tumor.*

## 1. INTRODUCTION

Inflammatory Myofibroblast Tumors (IMT) is a very rare type of tumor comprising myofibroblastic spindle cells and inflammatory cells. It was first observed in lungs by Bunn in 1939. Pathogenesis of this IMT has been postulated to range from infective, and reactive to a more recent belief of neoplastic. A recent change in the same is evidenced by recurrence, metastasis, and cytogenetic studies done in this field [1,2]. IMTs do not cause any specific symptoms and are almost always diagnosed incidentally or misdiagnosed [3]. Definitive management remains a mystery but surgical excision remains the treatment of choice. Gastrointestinal duplication cysts are rare congenital malformations that can occur anywhere in the gut. Foregut duplication cysts can be further divided into types based on the embryonic origin [4]. Treatment often involves surgical removal to prevent complications. Gastric duplication cyst makes up for 4 - 9% of all intestinal duplication cysts [5] Gastric Duplication cysts can be asymptomatic or can present as epigastric pain, vomiting, gastric outlet obstruction, weight loss, or mass per abdomen [6]. Both of the conditions are diagnosed based on radiological diagnosis with Endoscopic Ultrasound and Computed Tomography (CT) scan. Definitive diagnosis is obtained with tissue diagnostic tests like fine needle aspiration biopsy and histopathological examination (HPE) of surgical specimens. In the present case HPE was in favour of tumor consisting of spindle cells like GIST and myofibroblastic tumor This created a dilemma for definitive diagnosis in the preoperative period with contrasting findings in postoperative diagnosis. This report signifies the need to consider the alternative diagnosis mentioned above in case a tissue diagnosis is not available for preoperative diagnosis and its management.

## 2. CASE REPORT

A 37-year-old gentleman presented with complaints of epigastric and supraumbilical pain for 3 months insidious onset, dull aching, mild to moderate in intensity, nonprogressive with no specific aggravating or relieving factors. The pain was non-radiating and non-referring in nature. He also complained of nausea for 2-3 months. There was no history of fever, retrosternal pain or vomiting. The patient had no associated systemic symptoms. Past history & family history was unremarkable. The patient denied a history of any addictions. His bowel and bladder habits were unaltered.

Clinical examination showed hemodynamically stable with no systemic signs seen. Abdominal examination was unremarkable with no tenderness, guarding, or palpable abdominal lump. Contrast Enhanced Computed Tomography(CECT) scan revealed a well-defined isodense lesion in the perigastric region closely abutting the posterior gastric wall and body of the pancreas (Fig. 1). The lesion was 6.1x3.9x3.6 cms and showed thin peripheral postcontrast enhancement. There was no solid component noted. The possibility of malignant potential was low in the scan. His upper Gastrointestinal endoscopy showed no mucosal abnormality. Endoscopic Ultrasonography (EUS) was suggestive of small cystic lesions 3.5cm x 4.5cm arising from layer 4 of the stomach at the level of the cardiac end from the posterior wall (Fig. 2). It showed heterogeneous contents and septations. Biopsy was attempted but the yield was negative. Impression most likely of a gastric duplication cyst was given.

Considering the inability to acquire tissue diagnosis the decision to do a laparoscopic excision of the cyst was taken. Nathanson retractor was inserted through the epigastric port and the liver was retracted away from the operative site. The cyst was found to be ruptured and present in the lesser sac continuous with the posterior wall of the stomach (Fig.3). The cyst was excised in toto and separated from the gastric wall leaving a rent in the muscularis layer of the gastric wall. The cyst was placed in an endo bag and sent for histopathology (Fig. 4). The opening in muscularis was closed with polydioxanone 3-0 simple interrupted sutures.

Intraoperatively upper Gastrointestinal endoscopy was done to rule out mucosal breach and to check the adequacy of the repair. The stomach was insufflated with air and leakage of

gastric repair was tested from the laparoscope by keeping the suture line dipped in normal saline to look for any bubbling (Fig. 5). Repair was found to be adequate. An abdominal drain was placed in the lesser sac. Check dressing was done on day 2. The drain was removed on day 5. The liquid diet was started on day 3 and shifted to a full diet on day 5. The patient tolerated the procedure well and was discharged home with relieved complaints and stable vitals.

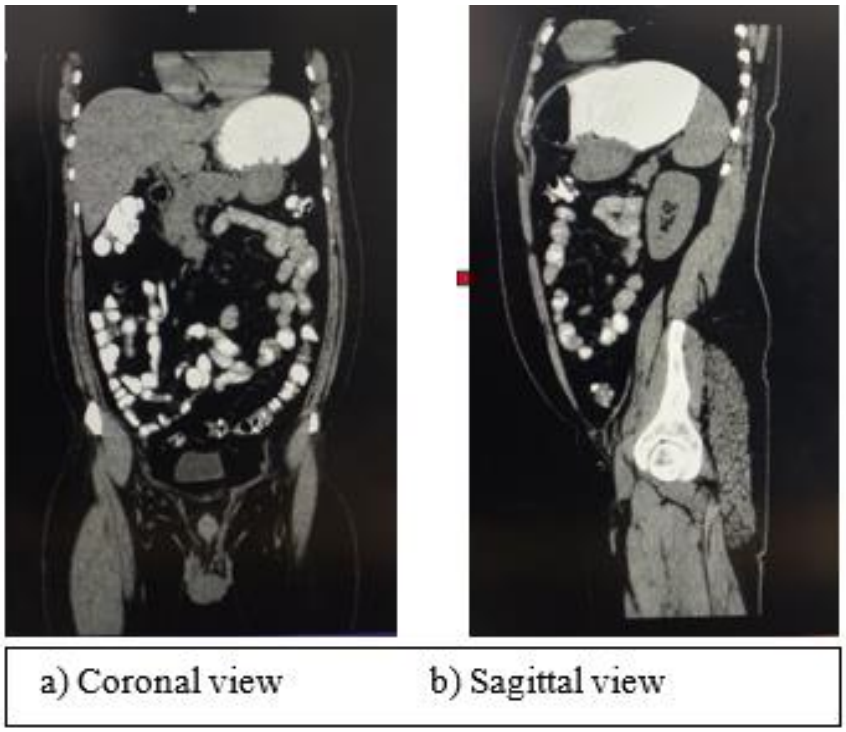


**Fig. 1. Lesion in the perigastric region closely abutting the posterior gastric wall and body of the pancreas**

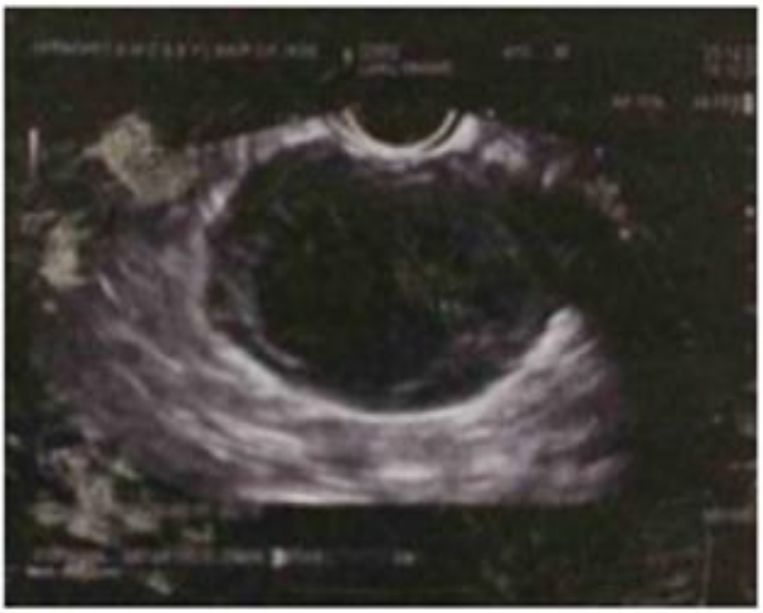

**Fig. 2. Stomach at the level of cardiac end from posterior wall**

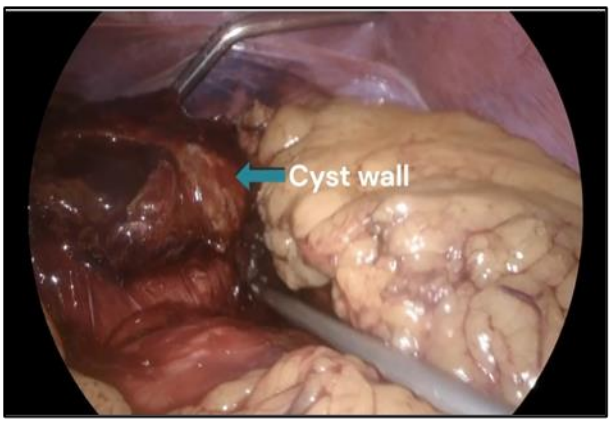


**Fig. 3. Lesser sac continuous with the posterior wall of the stomach**

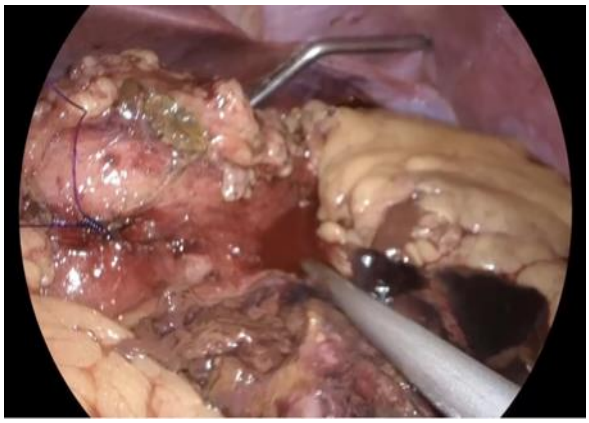

**Fig. 4. Cyst placed in an endobag**

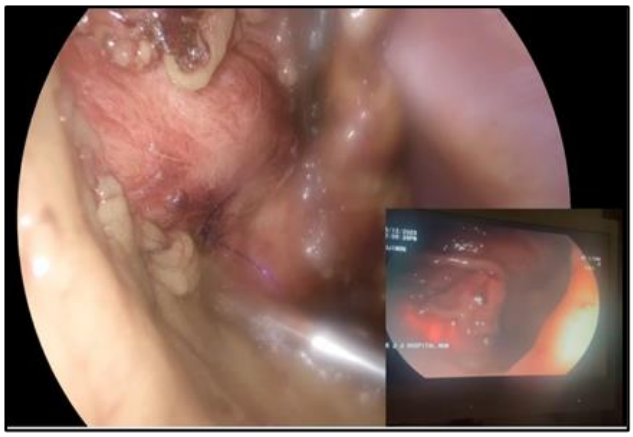

**Fig. 5. Line dipped in normal saline to look for any bubbling**

Histopathology report was suggestive of a spindle cell tumor most likely a GIST or myofibroblastic tumor. IHC testing was immunoreactive for smooth muscle actin, immunopositive for desmin and pancytokeratin while being immunonegative for ALK/CD117/DOG1/CD34/S100 giving us the definitive diagnosis of an inflammatory myofibroblastic tumor. A follow-up of 6 months has shown him to be disease & symptom-free.

## 3. DISCUSSION

Gastric duplication cyst, Gastrointestinal stromal tumors, and Inflammatory myofibroblastic tumors. Are rare tumors of the Gastrointestinal tract. The present case highlights this varied differential diagnosis based on preoperative evaluation and post-operative histopathological reports. Gastric duplication cysts are generally present in children and young adults and rarely in the fourth decade of life like the IMT [4]. Duplication cysts are thought to arise due to a developmental anomaly during the embryonic stage of growth. Endoscopic ultrasound is the investigation of choice. Typically seen as an anechoic to isoechoic cystic lesion arising from layers 3-5 of the stomach usually arising from the muscularis propria [6-8],.Lungs and gastrointestinal tract are the most common tissue of origin in either of the swellings and IMT rarely occurs in the oral cavity and maxillofacial areas [9]. Some authors suggest that EUS-FNA is necessary to rule out other conditions and malignant transformation within the cyst whilst ensuring a definitive diagnosis. IMT might require multiple biopsies to make a definitive diagnosis Role of fine needle aspiration is controversial as it known risk of rupture of the cyst, perforation(0.02%) infection (0.05%) hemorrhage(0.13%), pain(0.34%), pancreatitis(0.44%), and can result in dissemination in case of malignancy [10]. In the

present case, the cyst ruptured due to an attempted EUS-guided FNA.

Surgical excision remains the treatment of choice in such cases. This may result in a rent in the gastric wall which should be closed with sutures. It is imperative to rule out leaks after gastric wall repair. This can be done with simultaneous intraoperative upper gastrointestinal endoscopy and laparoscopy. Air insufflated through the endoscope and dipping the suture line in normal saline laparoscopically is a useful test. Leaks can be detected with evidence of any bubbling present which is seen laparoscopically which is many times done after a Sleeve Gastrectomy also in our center.

Both IMT & duplication cysts present similarly, however, the final diagnosis is confirmed on histopathology and immunohistochemistry after surgical excision. The present case highlights the importance of the same. IMT have a spindle cell morphology but they lack the cellular atypia of sarcomas. They are typically reactive to vimentin and Smooth Muscle actin on IHC [11]. The tumor might be ALK-positive or negative but always negative for CD21, CD117, CD23, and, S100.ALK immunohistochemistry is helpful in the diagnosis and prognosis of IMT. ALK positivity is only present in 56% of the cases and has been proven to be associated with local recurrence which is favorable rather than distal metastasis in ALK-negative tumors[12]. Immunonegativity in our case suggests we do a close follow-up with this patient in the future for signs of metastasis or recurrence. Duplication cysts mimic the histopathological appearance of the organ of origin and lack the IHC features mentioned above.

## 4. CONCLUSION

This report brings forth a case of cystic lesion arising from the stomach where the patient presented to us with complaints of epigastric pain with no positive examination findings. Preoperatively cystic lesions of the stomach can be confused as duplication cysts like in the present case. In such cases, malignant potential is low as compared to GIST or myofibroblastic tumors and other cystic lesions. Surgical management is indicated in symptomatic gastric cysts. However, a GIST or myofibroblastic tumor has to be surgically excised even if it is asymptomatic. EUS-guided Fine needle aspiration biopsy is controversial in such cases as evidence suggests a significant risk of rupture and infective potential of the procedure. A combination of intraoperative endoscopy along with laparoscopic leak test helps prevent complications.

## DISCLAIMER (ARTIFICIAL INTELLIGENCE)

Author(s) hereby declare that NO generative AI technologies such as Large Language Models (ChatGPT, COPILOT, etc) and text-to-image generators have been used during writing or editing of manuscripts.

## CONSENT

As per international standards or university standards, patient(s) written consent has been collected and preserved by the author(s).

## ETHICAL APPROVAL

As per international standards or university standards written ethical approval has been collected and preserved by the author(s).

## COMPETING INTERESTS

Authors have declared that no competing interests exist.

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