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Adenocarcinoma Arising from A Gastric Duplication Cyst in a Pregnant Patient: A Case Report

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Abstract

Objective: To report a rare case of Poorly Differentiated Adenocarcinoma Arising from a Non-Communicating Gastric Duplication Cyst (GDC) in a 25-year old pregnant patient diagnosed by routine Hematoxylin-Eosin and panel of Immunohistochemistry (IHC) stains. ***Clinical Feature:*** An incidental finding of a cystic mass measuring 21.0 cm in widest dimension located superior to the uterus and within the omentum was noted on her prenatal ultrasound at 32 weeks of AOG. INTERVENTION AND OUTCOME. Exploratory laparotomy and excision done with intraoperative findings of mass closely adherent to the transverse colon and mesocolon, posterior to pancreas and first part of the duodenum with normal bilateral adnexa. Histopathologic findings of the mass show multiple cystic structures with gastric mucosa, well-circumscribed smooth muscle layer and ectopic pancreatic tissues. The malignant transformation shows undifferentiated neoplastic cells in tight clusters and immunohistochemical staining with Cytokeratin, CAM 5.2 and Carcinoembryonic Antigen shows positive reactivity. Ki-67 proliferation index is high with 30 – 40% positivity. No reactivity in the neoplastic cells with Smooth Muscle Actin, p63, Chromogranin, DOG-1, LCA, PLAP and S-100. Two weeks post-excision, patient's condition deteriorated and eventually went to Cardiopulmonary Arrest secondary to Multiorgan Failure; Malignancy Stage IV. Clinico-radiologic, histopathologic and immunohistochemical findings is consistent with a Poorly Differentiated Adenocarcinoma Arising from a Non-Communicating Gastric Duplication Cyst.

Conclusion: GDC is a rare malformation with malignant transformation arising from it is extremely rare. Non-specific clinical presentation may not lead to a prompt diagnosis, especially in the patient where symptoms was masked by pregnancy. A high index of suspicion coupled with radiologic, histopathology correlation, with the aid of immunohistochemistry is important for early diagnosis and treatment.

Keywords: Gastric Duplication Cyst; Malignant Transformation; Poorly Differentiated Adenocarcinoma; Immunohistochemistry (IHC) stains

Introduction

Gastric duplication cyst (GDCs) is a rare phenomenon and account for only 2–9% of all gastrointestinal duplications. The majority is cystic, non-communicating and surrounded by a smooth muscular coat. GDCs are more common in young children and are rarely diagnosed in the adult population, who may present with symptoms of abdominal pain, gastric outlet obstruction or a palpable abdominal mass [1].

Malignant transformation of GDCs is rare, and only 14 cases have been reported in the English literature. Thirteen (13) cases was collated and reported by Zhu, et al in 2015 and 1 additional case by Chan, BPH et al in 2018. Adenocarcinoma is the most common histopathology, although squamous cell carcinoma, neuroendocrine, and gastrointestinal stromal tumors have also been reported [2].

Case Summary

This is a case of a 25 year old pregnant patient with intra-abdominal mass. An incidental finding of a cystic mass measuring 21.0 cm in widest dimension located superior to the uterus and within the omentum was noted on her prenatal ultrasound at 32 weeks of AOG. Three (3) weeks post Low Transverse Cesarean Section (LTCS), due to increasing abdominal pain and gradually enlarging abdominal girth, patient was referred to surgery department. Pertinent laboratory tests showed increased lipase and amylase results. CT scan was done, with considerations of a mesenteric vs. ovarian cyst (fig. 1). Exploratory laparotomy and excision done with intraoperative findings of the mass closely adherent to the transverse colon and mesocolon, posterior to the pancreas and first part of the duodenum with normal bilateral adnexae (fig. 2). Initial histopathologic results revealed an Undifferentiated Neoplasm probably arising from a Gastric Duplication Cyst with suggestion of further work-up with immunohistochemical stains for a definite diagnosis. Two (2) weeks post-excision, multiple bleeding liver nodules was noted. Patient condition deteriorated and eventually went to Cardiopulmonary Arrest secondary to Multiorgan Failure; Malignancy Stage IV.

Age/sex	***Symptoms***	***Impression***	***Gastric Duplication***	***Malignant Transformation***	***Follow-Up***
25/ F	Incidental finding of a cystic mass on her prenatal ultrasound at 32 weeks of AOG; abdominal pain and gradually enlarging abdominal girth	Mesenteric vs. Ovarian Cyst	Size= 21.0 cm in widest dimension	Poorly Differentiated Adenocarcinoma	Died 2 weeks post-excision
			Location= adherent to transverse colon and mesocolon, posterior to the pancreas and first part of the duodenum; normal bilateral adnexae		

			Microscopic findings=- multiple cystic structures with gastric mucosa, well-circumscribed smooth muscle layer and ectopic pancreatic acini.		

Table 1: Summary of Patients' Characteristic Findings.

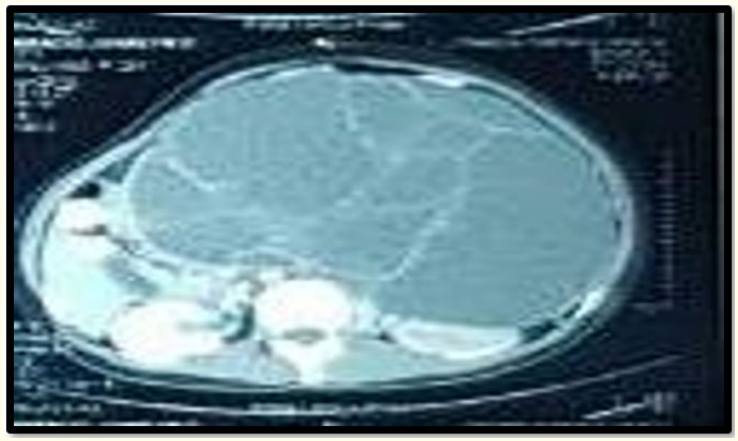


Figure 1: CT scan with contrast discloses a large and bulky thin septated fluid cystic mass characteristic of mesenteric vs ovarian in nature.

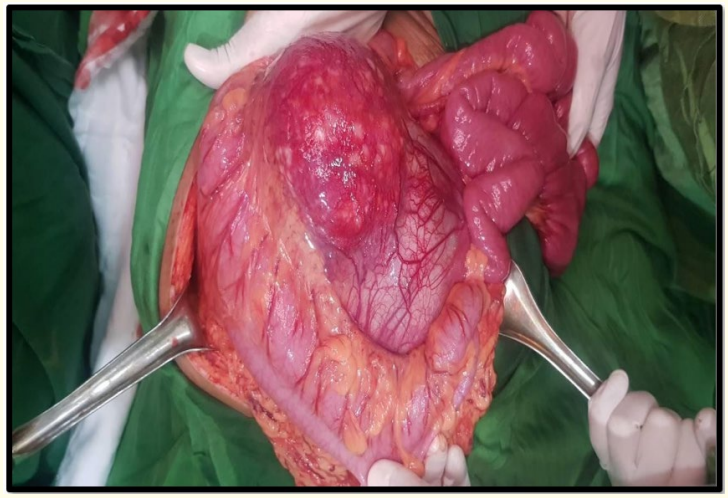


Figure 2: Intraoperative findings of the mass closely adherent to the transverse colon and mesocolon, posterior to the pancreas and first part of the duodenum.

Gross, Histopathologic and Immunohistochemical Findings

Received is a tan-brown to dark-red, ovoid, rubbery, cystic tissue measuring 21.0 x 18.0 x 8.0 cm (Fig. 3). Cut sections of the mass shows several multi-loculated cystic structures measuring 3.5 cm to 5.0 cm in greatest diameter containing mucinous material.

Microsections show multiple cystic structures (Fig .4) with the innermost layer of the gastric wall lined by gastric foveolar epithelium with mucinous simple columnar epithelium and gastric glands in the mucosa followed by well-circumscribed smooth muscle layer. Malignant transformation is noted in the mucosa and in the outermost layer (Fig. 5). Ectopic pancreatic acini are seen in the periphery (Fig. 6). The malignant transformation show sheets of neoplastic cells in tight clusters. The individual neoplastic cells are pleomorphic, with increased nucleo-cytoplasmic ratio, irregular hyperchromatic nuclei, ample to scant cytoplasm with prominent nucleoli. Necrotic areas are noted with a mitotic count of 1-2/hpf (Fig. 7).

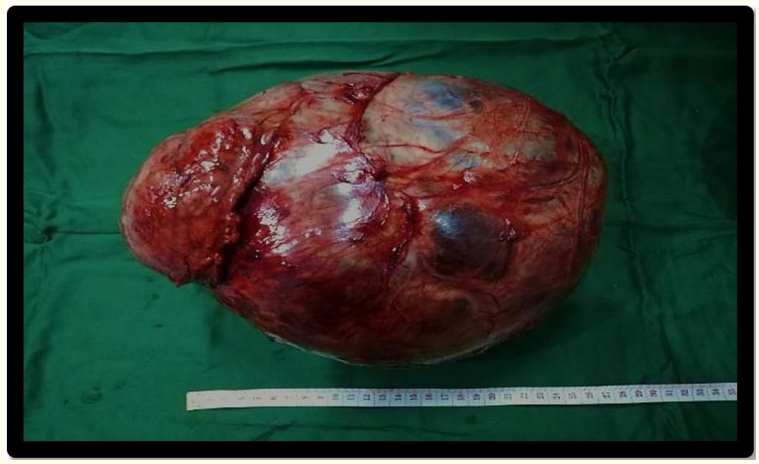


Figure 3: Gross findings: tan-brown to dark-red, ovoid, rubbery, cystic tissue measuring 21.0 x 18.0 x 8.0 cm.

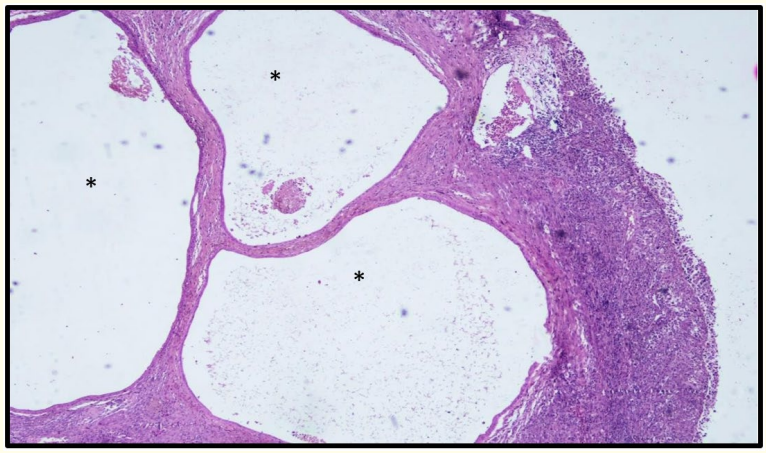


Figure 4: Hematoxylin and Eosin (H & E, scanner) showing multicystic structures (*).

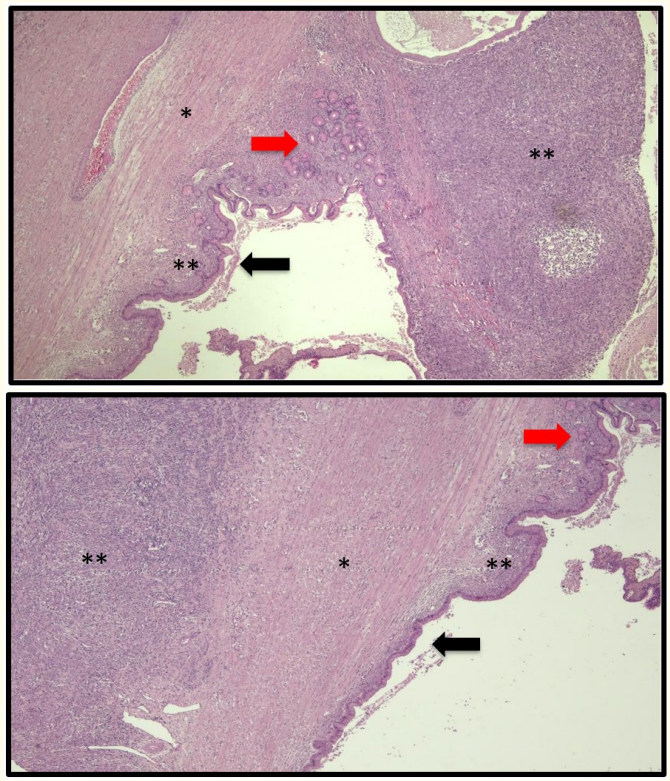
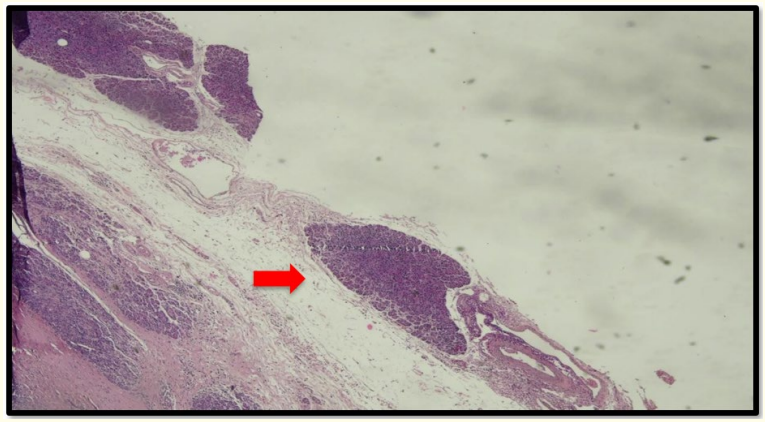


Figure 5: Hematoxylin and Eosin (H & E) (A. scanner view; B. 10x) revealed the diagnosis of GDC: cystic structures exhibited the innermost layer of gastric wall lined by gastric foveolar epithelium with mucinous simple columnar epithelium (black arrow) and gastric glands (red arrow) in the mucosa followed by well-circumscribed smooth muscle layer (*). Malignant transformation is noted in the mucosa and outermost layer (**).



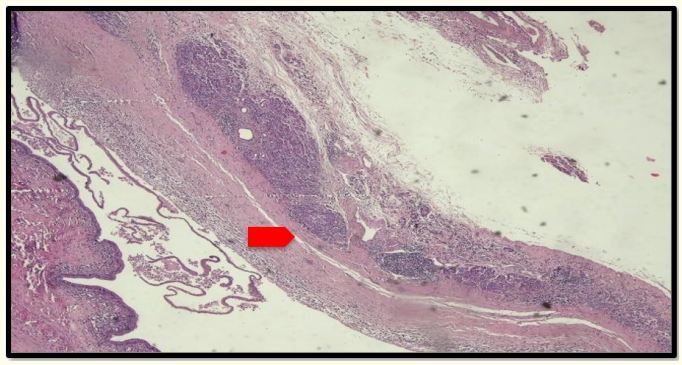


Figure 6: Hematoxylin-Eosin (H&E) (A. scanner view; B. 10x) shows ectopic pancreatic tissues (red arrow) seen in the periphery.

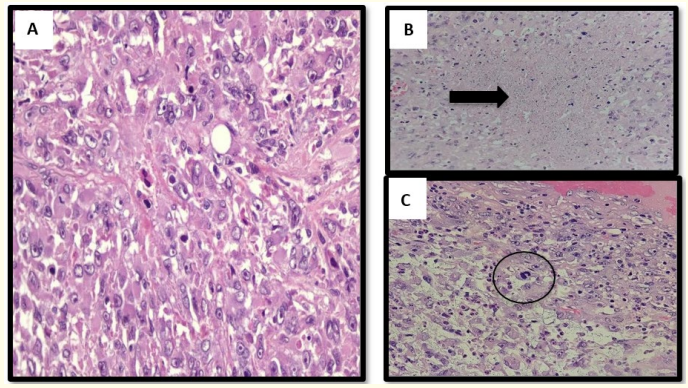


Figure 7: Hematoxylin and Eosin (H & E) (40X) A. the malignant transformation shows sheets of neoplastic cells in tight clusters. The individual neoplastic cells are pleomorphic, with increased nucleo-cytoplasmic ratio, irregular hyperchromatic nuclei, ample to scant cytoplasm with prominent nucleoli; B. necrotic areas; C. mitotic count of 1-2/ hpf.

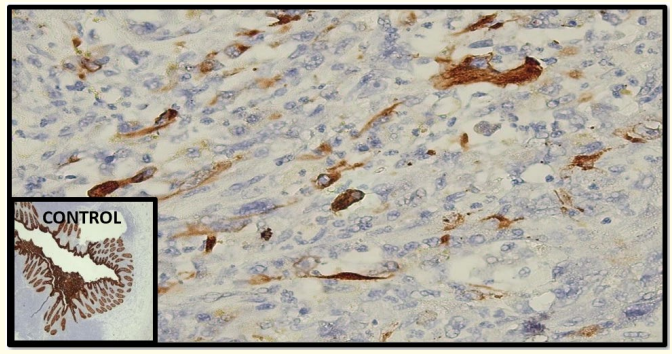


Figure 8: Cytokeratin. Patient (40X), showing positive cytoplasmic expression in neoplastic cells; Control (10x), positive.

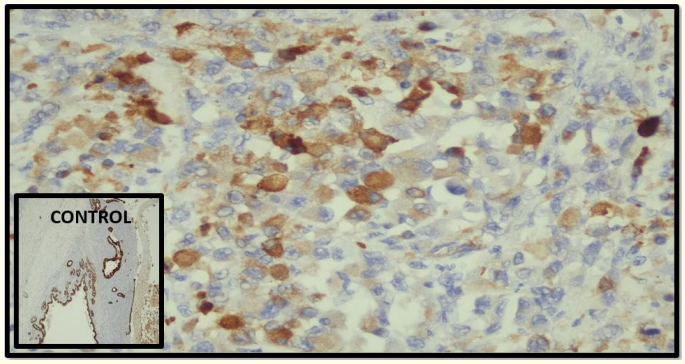


Figure 9: CAM 5.2. Patient (40X), showing positive cytoplasmic expression in neoplastic cells; Control (40x), positive.

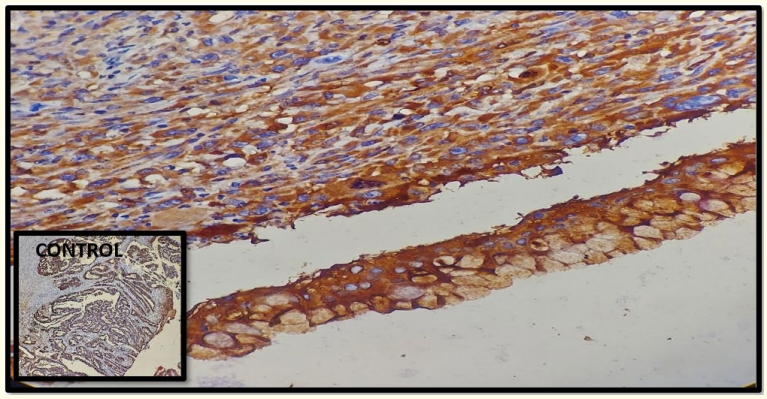


Figure 10: CEA. Patient (40X) showing positive cytoplasmic and membranous expression in neoplastic cells. Control (10x) positive.

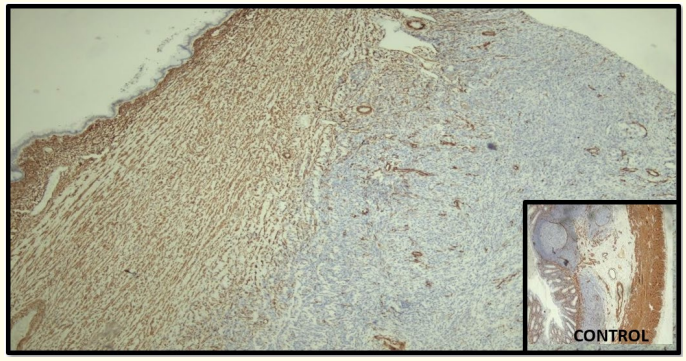


Figure 11: Smooth muscle actin (SMA). A Patient (scanner), showing positive diffuse cytoplasmic and membranous expression in normal smooth muscle layer (*) and negative expression in the neoplastic cells (**); Control (10x), positive.

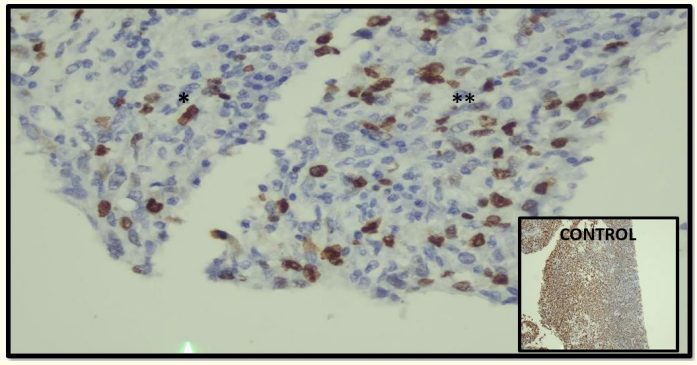
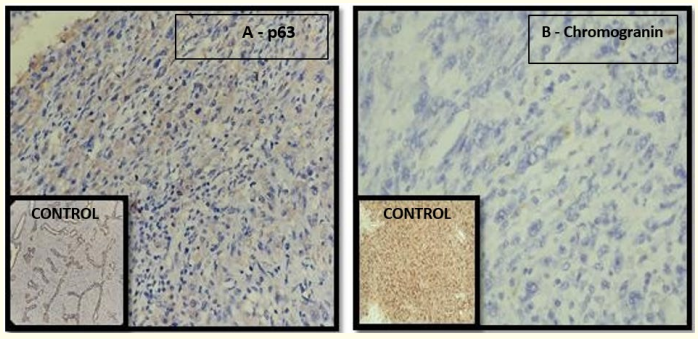
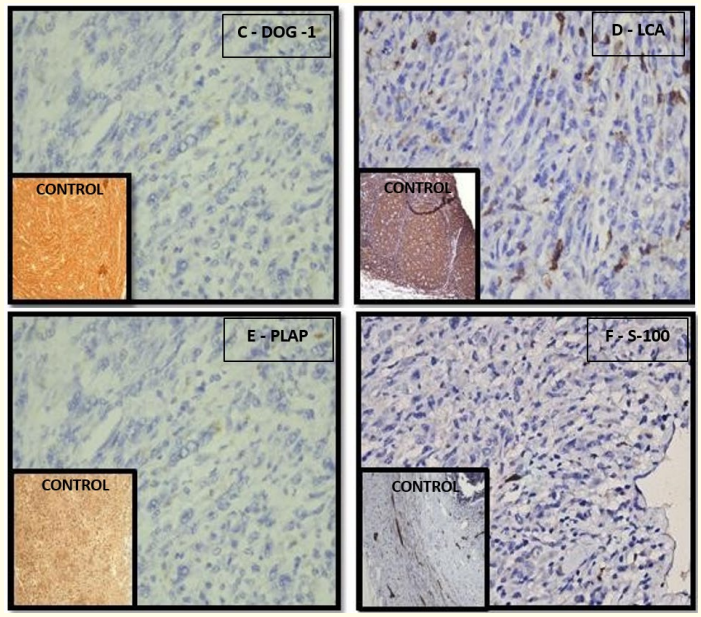


Figure 12: Ki-67%. Patient (40X) showing high proliferation index with 30-40 % nuclear staining in the neoplastic cells. Control (10x), positive.





*(LCA: Leukocyte Common Antigen; PLAP: Placental Alkaline Phosphatase; DOG-1: Discovered on GIST 1).

Figure 13: A-F, Patient (40x) showing negative expression in the neoplastic cells. Control positive.

IHC	*Patient*	*Adeno Carcinoma*	*SCCA*	*NET*	*GIST*	*Lymphoma*	*GCT*	*Neural Tumor*	*Smooth muscle Tumor*
1.CK	(+)	(+)	(+)	(-)	(-)	(-)	(-)	(-)	(-)
2.CAM 5.2	(+)	(+)	(+)	(-)	(-)	(-)	(-)	(-)	(-)
3. CEA	(+)	(+)	(-)	(-)	(-)	(-)	(-)	(-)	(-)
4. p63	(-)	(-)	(+)	(-)	(-)	(-)	(-)	(-)	(-)
5. Chromogr-anin	(-)	(-)	(-)	(+)	(-)	(-)	(-)	(-)	(-)
6. DOG-1	(-)	(-)	(-)	(-)	(+)	(-)	(-)	(-)	(-)
7. LCA	(-)	(-)	(-)	(-)	(-)	(+)	(-)	(-)	(-)
8. PLAP	(-)	(-)	(-)	(-)	(-)	(-)	(+)	(-)	(-)
9. S-100	(-)	(-)	(-)	(-)	(-)	(-)	(-)	(+)	(-)
10. SMA	(-)	(-)	(-)	(-)	(-)	(-)	(-)	(-)	(+)
*SCCA- Squamous Cell Carcinoma; NE- Neuroendocrine Tumor; GIST- Gastrointestinal Stromal Tumor; GCT- Germ Cell Tumor.									

Table 2: Immunohistochemical (IHC) Profile of the Malignant Transformation (Histologic Subtype) and other Differential Diagnoses.

Discussion

Duplication of the gastrointestinal tract is a rare congenital anomaly. Most cases will occur in females compared to males (8:1), with the majority of cases being diagnosed within the first 3 months of life and rarely after 12 years of age. Symptoms may include abdominal pain, gastric outlet obstruction or a palpable abdominal mass [1].

The pathogenesis for of alimentary tract duplications are still uncertain and several theories have been proposed, but most researchers believe that abnormal recanalization after the solid epithelial stage of embryonic bowel development is the most likely mechanism [4].

There are 2 morphologic types of duplications, cystic and tubular. Cystic duplications are the most common and have no communication with the bowel lumen. Tubular duplications communicate with the lumen in 75 % of cases. The ileum is the predominant site of duplication [2].

The stomach accounts for 2-9 % of all duplications diagnosed, which is the least common site. Gastric Duplication Cysts (GDC) can be found anywhere in the stomach, the most common is the greater curvature. They may communicate with the gastric lumen, however the majority is non- communicating with a cystic configuration [1]. In this case, it is cystic and non-communicating.

The common pathologic characteristics that are used as criteria for the diagnosis are: lesion is coated by smooth muscle, continued with the stomach, and inner lined with mucosae, which can be epithelium of any portion of the gastrointestinal tract. In addition, the lesion should be attached to the gastric wall. Ectopic pancreatic mucosa has been reported [5]. Both gastric and pancreatic ectopic tissues can be seen, which are the most common and tend to be the most clinically significant, as patients develop complications such as pancreatitis [1]. In this case, the multi-cystic structures exhibited the innermost layer of the gastric wall with gastric foveolar epithelium lined by mucinous simple columnar epithelium and gastric glands in the mucosa followed by well-circumscribed smooth muscle layer. Ectopic pancreatic tissues were noted and was the cause of the increasing levels of lipase and amylase in the patient. Since the case is non-communicating, it is not attached or continuous with the stomach. Fulfillment of a non-communicating GDC is achieved.

GDC is a benign lesion, but has the potential for malignant transformation from either the duplication itself or the adjacent gastric wall. The mechanism of such malignant transformation process is still uncertain. In the event of presence of any suspicion of malignant transformation process in the gastric duplication, it is recommend surgical resection of the lesion once detected [5].

Malignant transformation of GDCs is rare, and only 14 cases have been reported in the English literature. Thirteen (13) cases was collated and reported by Zhu, et al in 2015 and 1 additional case by Chan, BPH et al in 2018. Adenocarcinoma is the most common histopathology, although squamous cell carcinoma, neuroendocrine, and gastrointestinal stromal tumors have also been reported [2]. In our case, a Poorly Differentiated Adenocarcinoma is the histologic subtype of the malignant transformation. To the best of our knowledge, no local data is ever reported or published yet.

Conclusion

Gastric duplication cyst is a rare malformation and a malignant transformation arising from it is extremely rare. Non-specific clinical presentation may not lead to a prompt diagnosis, especially in the patient where symptoms was masked by pregnancy. A high index of suspicion coupled with radiologic, histopathologic correlation and with the aid of immunohistochemistry is important for early diagnosis and prompt treatment.

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