

Research Article

Gastric Adenomyoma: An Uncommon Cause of Dyspepsia and a Rare Endoscopic Finding

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Abstract

Background: Gastric adenomyoma (GA) is a rare benign lesion comprising of ducts and glands embedded in smooth muscle stroma. Thirty-seven (37) cases of GA were identified until 1993, however, only 15 cases are said to have been reported from 1993 to 2016. Oesophagogastroscopy has been widely used in evaluating GA. However, the diagnosis of GA remains exclusively histological.

Case: We report a 26-year-old Nigerian woman who presented with recurrent dyspepsia with her endoscopic findings suggestive of GA. GA was confirmed by histology, and she was managed conservatively.

Conclusion: This report will contribute to creating awareness of this uncommon condition and also reminding physicians in considering GA as a possible differential of dyspepsia.

Keywords: gastric, adenomyoma, dyspepsia, endoscopy, Nigerian, woman

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1. Introduction

Gastric adenomyoma (GA) is a rare benign lesion comprising of ducts and glands embedded in smooth muscle stroma [1].

GA was first described in 1903 by Magnus-Alsleben [2]. It has been characterized as a hamartomatous lesion, and even as heterotopic pancreas without an exocrine or endocrine function in some studies [3].

Thirty-seven (37) cases were identified until 1993, however, only 15 cases are said to have been reported from 1993 to 2016 [4, 5]. It is commonly located in the gastrointestinal tract such as in the stomach (25–38%), duodenum (17–36%), and jejunum (15–21%) [6].

In the stomach, the most common location reported is in the antrum (85%) and the pylorus (15%). It is rarely found in the body [7, 8].

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GA can be found among patients between the ages of one month to eighty years, though, commonly seen between the fourth and sixth decade of life. It affects both male and female equally [5, 9].

GA can present as incidental findings during laparoscopic intervention or autopsy. However, non-specific symptoms such as nausea, vomiting, and epigastric pain have been reported. Hematemesis, melena, and anemia have also been reported in some cases [10, 11].

The differential diagnosis of GA includes gastrointestinal stromal tumors, leiomyoma, hamartoma, and a reactive hypertrophic smooth muscle, especially when it presents as a solid submucosal mass or cystic lesion [4, 12].

Oesophagogastroscopy, computer tomography (CT) scan, and endoscopic ultrasonography have been widely used in evaluating GA. Despite the availability of these diagnostic modalities, diagnosing GA can still be difficult [10, 13]. Nevertheless, the diagnosis of GA remains exclusively histological based on the identification of heterotopic tissues, the architectural pattern of the lesion, its relation with the surrounding tissues, and the exclusion of malignancy [4].

We report a case of a young woman with GA who presented with dyspepsia.

2. Case Report

A 26-year-old Nigerian woman presented to the gastroenterology clinic with a six-month history of recurrent epigastric pain. The pain was burning in nature and insidious in onset.

It radiated to the left hypochondrium and was relieved by ingesting antacids. There were no known aggravating factors.

The pain was associated with bloating and nausea, however, there was no history of heartburn or regurgitation. There was no history of jaundice, vomiting, abdominal swelling, weight loss nor change in bowel habit. There was no history suggestive of gastrointestinal bleeding.

The patient's systemic review was not contributory. There was no history of ingestion of non-steroidal anti-inflammatory drugs (NSAIDs) or herbal preparation. Her family and past medical history were not contributory or significant. She neither smoked cigarette nor ingested alcohol.

Except for her tender epigastrium, her physical examination were unremarkable.

The platelet count was $180 \times 10^9/L$ (Reference: $150\text{--}450 \times 10^9/L$); packed cell volume was 38% (Reference: 35–45%); Hemoglobin count was 14g/dl (Reference: 13.5–17.5g/dl), and white blood cell count was $7.4 \times 10^9/L$ (Reference: $4\text{--}12 \times 10^9/L$). Serum creatinine was 70 $\mu\text{mol/l}$ (Reference: 60–100 $\mu\text{mol/l}$), urea was 5.0 mmol/l (Reference: 2.5–6.5 mmol/l), potassium was 3.8 mmol/l (Reference: 3.5–5.0 mmol/l), and sodium was 138 mmol/l (Reference: 135–145 mmol/l). Serum alanine aminotransferase was 10iu/L (Reference: 4–36iu/L), Serum aspartate aminotransferase was 12iu/L (Reference: 4–36iu/L), Serum alkaline phosphatase was 90iu/l (Reference: 45–146iu/L), and Serum bilirubin was 12mmol/l (Reference: 4–17mmol/l). Viral screening for hepatitis B, C, and HIV were all negative. Serum lipase was within normal limit.

The patient's chest X-ray, abdominopelvic ultrasound scan, and electrocardiography (ECG) findings were normal. Urea breath test (UBT) was also negative.

She had an oesophagogastroduodenoscopy (OGD) that revealed a small (1–2 cm), firm, circular, umbilicated subepithelial antral nodule (Figure 1). The mucosa overlying the lesion appeared inflamed. The oesophagus, fundus, and corpus were normal. The duodenal mucosa also appeared normal. Separate biopsies were taken from the antral nodule, fundus, and corpus.

The histology of the biopsied nodule showed columnar epithelium overlying a lamina propria within which were nests of Brunner glands that were separated by smooth muscle bundles and mucous glands. No cytologic atypia was seen (Figure 2). The histological findings were diagnostic of a GA. No *Helicobacter pylori* (*H. pylori*) was seen. The histological findings in the fundus, corpus, and duodenum were normal.

Endoscopic ultrasound (EUS) and immunochemistry were not done as these were not available at the time this patient was managed.

The patient was placed on proton pump inhibitors (PPI) for four weeks with the resolution of her symptoms after two weeks of medical therapy.

The patient was referred to the general surgeons, who advised that patient should be followed-up and monitored due to the absence of dysplasia or malignant cells on a histology, the small size of the antral nodule, and resolution of symptoms with medical therapy.

A repeat OGD and biopsy of antral lesion done six months after showed no significant change in the size of the nodule and no malignant transformation seen on histology. The patient is currently being followed-up in both gastroenterology and general surgery clinic and has remained asymptomatic.

3. Comments

GA is a rare benign tumor first described by Magnus-Alsleben in 1903 [2]. Since then, about 52 cases were reported until 2016 [15]. To the best of our knowledge, our case will be the first to be reported in Nigeria. The index patient's age was within the age range reported for GA; however, we could not ascertain if this lesion was present since childhood.

Our patient presented with only dyspepsia; however, other symptoms such as vomiting, melena, and even gastric outlet obstruction have been reported in other literatures [7, 11]. Common associations of GA such as annular pancreas, Gardner syndrome with duodenal adenomas, and gastric duplication reported in some cases were absent in our patient [12].

The long-term recurrent dyspepsia was the main indication for endoscopy in this patient. The OGD showed small, firm, circular, umbilicated subepithelial antral nodule. This lesion resembled heterotopic pancreas endoscopically, which was a differential diagnosis considered during the evaluation of our patient as GA was regarded as an abortive variant of the heterotopic pancreas in some studies [4].

GA was confirmed histologically with typical histological findings in keeping with this entity. EUS and immunochemistry were not done as they were not available at the time of evaluation. However, histology remains the gold standard for the diagnosis of GA [4].

We believe the dyspeptic symptoms our patient had is most likely due to the presence of GA in the antrum, as the other possible causes of dyspepsia such as NSAID use and *H. pylori* were absent in this case. Moreover, dyspeptic symptoms and even upper gastrointestinal bleeding have been reported in patients with GA [14].

Our patient was managed conservatively with PPI with resolution of her symptoms. The lesion was not resected but rather monitored, and the patient is being followed-up in both gastroenterology and surgical clinic.

Treatment options for GA include endoscopic submucosal dissection, wedge resection of tumor to extensive surgical resections [11, 15]. The advent of frozen section has made intraoperative diagnosis of GA possible with reduction of unnecessary extensive operations [11].

In summary, GA is an uncommon cause of dyspepsia and unusual finding on OGD. This condition is rare in this part of the world, and its presentation can mimic common causes of dyspepsia. Our patient had typical endoscopic and histology findings consistent with GA. We believe that our report of this uncommon condition will further raise

awareness and remind Physicians to consider GA as a possible differential of dyspepsia, thus aiding its prompt diagnosis and treatment.

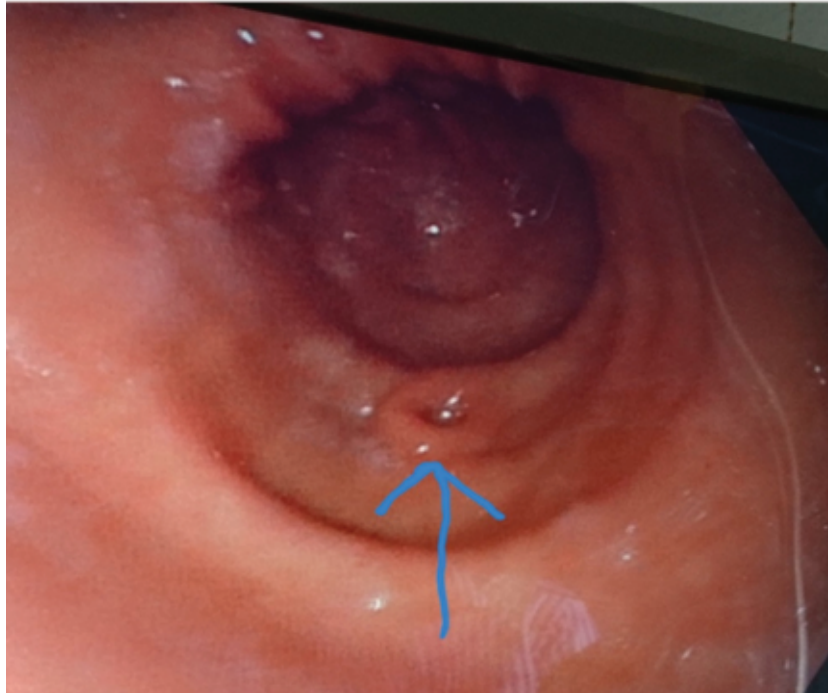


Figure 1: OGD showing a firm, circular, umbilicated subepithelial antral nodule.



Figure 2: Nests of Brunner glands (blue arrow) that are separated by smooth muscle bundles (yellow arrow).

Author Contribution

Opeyemi F Bamidele was the patient's gastroenterologist who reviewed and contributed to the final approval of the version of the manuscript to be published. Matthew O Bojuwoye contributed to the drafting of the manuscript, and Mudashiru Lawal was the patient's pathologist who performed the pathological analysis and interpretation of sample and contributed to manuscript drafting. Ruth A Bello participated in the final drafting of the manuscript and all authors issued the final approval to the submission of this manuscript.

Conflict of Interest

The authors declare that they have no conflict of interest.

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