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# Benign pyloric adenomyoma presented as gastric outlet obstruction: a case report and review of the literature

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## Abstract

**Background** Gastric adenomyoma is a rare benign tumor composed of glandular structures and smooth muscle fibers. While some classify gastric adenomyoma as a hamartoma, others view it as an abortive form of heterotopic pancreas. Despite its benign nature, there is a risk of malignant transformation. Predominantly found in the antrum, gastric adenomyoma affects all ages but is most common in adults aged 40–60 years. Symptoms are nonspecific, and its similarity to other lesions complicates diagnosis. This paper aims to provide a review of medical literature on gastric adenomyoma and its diagnosis and treatment methods, along with presenting an additional case report on the same topic.

**Case presentation** We present the case of a 55-year-old Syrian man who experienced vomiting, weight loss, and chronic partial constipation. An obstructing mass in the pylorus was detected, and then an open surgery was performed to excise the lesion. A biopsy of the resected mass was obtained for histopathological examination. The final diagnosis of the lesion was pyloric-region adenomyoma with severe pyloric stenosis. After the successful surgery, the patient recovered without any recurrence or complications.

**Conclusions** Several diagnostic approaches are available, including radiological studies, endoscopic examination, and fine needle aspiration guided by endoscopic ultrasonography. Treatment options involve endoscopic submucosal dissection and complete laparotomy resection. Further studies and thorough reviews are recommended to better understand the best clinical practices. Practitioners should consider gastric adenomyoma when encountering a mural gastric lesion.

**Keywords** Gastric adenomyoma, Gastric outlet obstruction, Partial gastrectomy, Case report

## Introduction

Gastric adenomyoma (GA) is classified by the World Health Organization as a rare benign tumor of the stomach, characterized by a unique histological composition of glands and cysts lined by columnar or squamous epithelium, embedded with a prominent smooth muscle stroma [1–3]. On the one hand, some authors categorized gastric adenomyoma as hamartoma. On the other hand, some suggested defining it as an abortive variant of heterotopic pancreas [4]. Despite its benign classification, a possibility of malignant transformation still exists

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[4]. Although GAs are predominantly found in the gallbladder, sporadic cases in the stomach have been documented, with the majority (85%) occurring in the antrum, and the rest (15%) have been reported in the pylorus [1, 4].

Gastric adenomyoma ranges in size from 0.6 to 4.5 cm and can affect individuals across a wide range of age, from infancy to advanced age, with the highest occurrence observed in adults between the fourth and sixth decades of life [4, 5]. Clinical presentation varies widely; nonspecific symptoms such as nausea, vomiting, epigastric pain, hematemesis, melena, and hematemesis are repeatedly noticed [4]. GA poses a diagnostic challenge due to its resemblance to other gastrointestinal lesions, for instance, gastrointestinal stromal tumors (GISTs), leiomyoma, or leiomyosarcoma [3]. This confusion can lead to a misdiagnosis and inappropriate management strategies.

Herein, we report a rare case of gastric adenomyoma that caused stomach outlet obstruction that had been suspected to be a malignant tumor before histopathological examination was performed. It is worth noting that this is one of the first case reports of gastric outlet obstruction due to pyloric adenomyoma in an adult patient. In this paper, we have also briefly conducted a review of the medical literature for similar case reports.

## Case presentation

A 55-year-old Syrian man from rural areas near Aleppo City presented to the gastrointestinal department with a chief complaint of chronic vomiting of food remnants persisting for 4 years and a weight loss of 34 kg during the last month before presentation. The patient also experienced chronic partial constipation. During gastrointestinal endoscopy at our hospital, an enlarged food-filled stomach and severe pyloric edema were observed that obstructed the gastric outlet and stopped further passage of the endoscope toward the duodenum. The primary diagnosis was gastric outlet obstruction due to pyloric stenosis. To assess the extent and cause of the obstruction, a computed tomography (CT) scan of the abdomen was performed, revealing a severely dilated stomach with a 4 cm mass in the pylorus and 1 cm thickening of the pyloric wall (Fig. 1).

Based on these findings, open surgery was performed to resect the mass that was initially suspected to be a malignant tumor causing the obstruction (Fig. 2). During surgery, multiple enlarged lymph nodes were found surrounding a 4×3 cm mass in the pylorus. The mass and surrounding lymph nodes were excised via distal partial gastrectomy with free surgical margins. The resected mass was sent for histopathologic examination. The normal reactive lymph nodes excised from the lesser and greater omentum indicated the absence of metastatic disease.

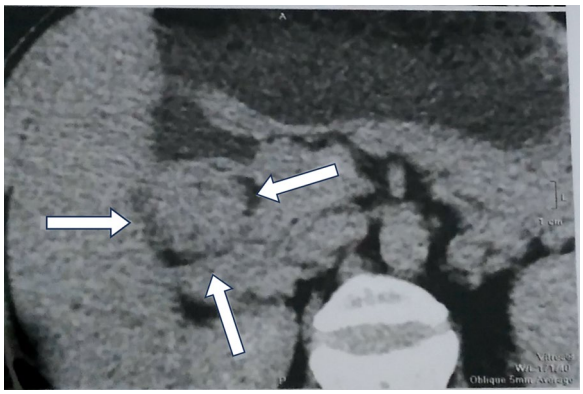

**Fig. 1** Computed tomography scan revealing the mass in the pyloric region of the stomach. White arrows point at the lesion

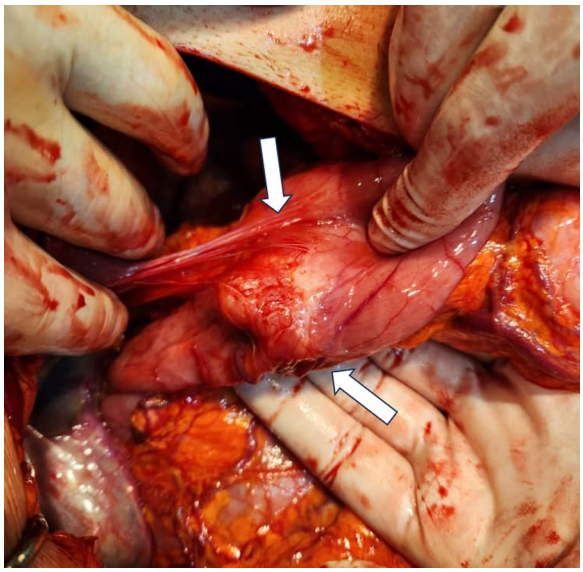

**Fig. 2** White arrows point at the mass of lesion located in the pyloric region of the stomach during surgery before its resection later

Histopathological analysis revealed normal gastric epithelium, with well-differentiated glands showing some areas of dilation lined by columnar to cuboidal cells in the muscularis propria and submuscular layer. No atypia or mitotic activity was observed, and the proliferative glands were surrounded by smooth muscle bundles (Fig. 3). The final diagnosis of the obstructing lesion was pyloric-region adenomyoma with severe pyloric stenosis. The patient's condition improved gradually after the successful surgery, and he was discharged home after the fourth day of hospitalization. After 1 year of follow-up, our patient experienced

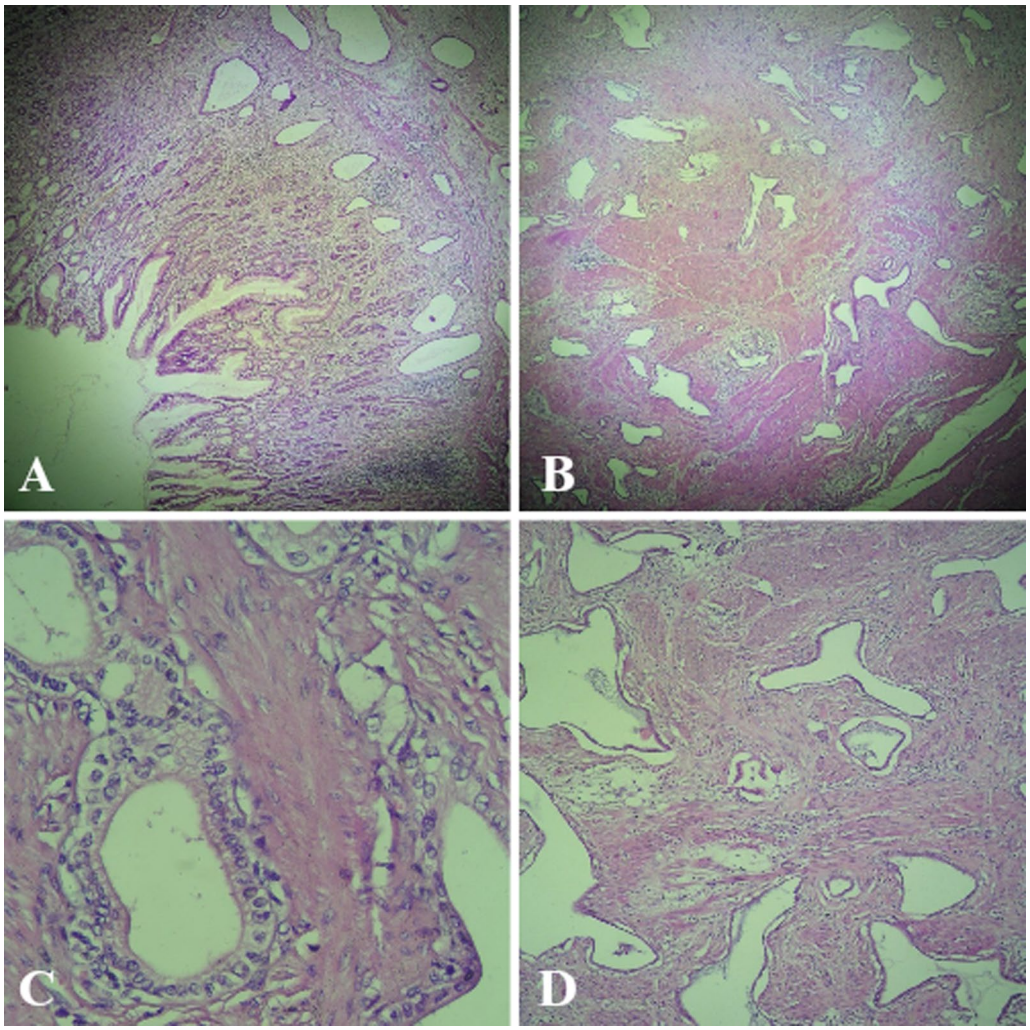


**Fig. 3** Pathohistological examination of the gastric adenomyoma lesion. **A** Normal gastric epithelium. **B**, **D** Islands of epithelial ducts and glands surrounded by smooth muscle bundles. **C** No presence of atypia or mitosis in the proliferative ducts

neither recurrence nor complications related to the surgery.

## Discussion

Gastric adenomyoma was initially described in 1903 by Magnus-Alsleben. Subsequently, Anand *et al.* [6] mentioned that only 52 case reports were published in medical literature until the year 2017. Later on, and according to the review of Barré *et al.* [3], nine cases of gastric adenomyoma have been reported and published during 2017 and 2020. In this paper, we have reviewed the Medline-indexed literature published after 2020 via PubMed using the search strategy "(adenomyoma) AND (gastric OR stomach)" with applying (Case Reports) filter to the results. Only eight articles were reached, and of them, merely four papers reported patients with GA [7–10]. Therefore, we claim, to our knowledge, that a total of approximately 65 articles have reported GA in the medical literature until the date of publishing this current case report, making our article the 66th.

Of these cases, very few presented with symptoms resembling gastric duplication cysts or pyloric stenosis, or manifested as gastric outlet obstruction [3]. Takeyama *et al.* [5] reported a case of GA causing obstruction in a 1-month-old girl. Similarly, Rhim *et al.* [11] reported a case of GA causing obstruction in a 1-week-old boy, emphasizing the importance of considering GA in the differential diagnosis of gastric outlet obstruction in infants, particularly when radiological features deviate from those typical of hypertrophic pyloric stenosis. Notably, cases of GA presenting as gastric outlet obstruction are more commonly reported in pediatrics, and building

on the findings of the review by Barré *et al.* [3], the presentation of obstruction is more common when lesions are located closer to the gastric outlet (that is, cases with GA in the pylorus usually manifest as stomach outlet blockage).

Symptoms are either absent or nonspecific in the majority of adult cases, which may introduce a reason why GA is considered a low-occurrence lesion [2, 8]. Usual manifestations include epigastric pain and/or discomfort, hematemesis, dyspepsia, vomiting, anemia, melena, and occasionally peritonitis due to perforation [4, 8]. In pediatrics, intussusceptions are the most frequent complications [5]. Our patient had chronic food emesis for 4 years and 34 kg weight loss, which made us concerned for malignancy; after performing endoscopy and discovering the pyloric stenosis and stomach outlet blockage, we immediately decided to carry out laparotomy.

Preoperative diagnosis of GA is challenging as doctors are unfamiliar with it [12]. Radiological diagnosis for gastric adenomyoma encompasses barium meal X-ray, CT scan, ultrasonography, gastrointestinal endoscopic ultrasound, and occasionally magnetic resonance imaging (MRI) [8]. However, several authors, including Vandelli *et al.* [13] and Zhu *et al.* [2], argued that endoscopic examination alone may be insufficient for diagnosing GA due to the limitations of superficial biopsies as they do not provide accurate representative tissue samples. Moreover, they highlighted that endoscopic features are not definitive for distinguishing adenomyoma from tumors that may present as antral umbilicated submucosal masses [2, 4, 8]. They advocated for resection followed by histological studies and immunohistochemical stains for definitive diagnosis, and recommended the intraoperative diagnosis via frozen section to avoid excessive surgical procedures [2].

In contrast, Barré *et al.* [3] presented a unique perspective and proposed a novel approach by utilizing the endoscopic ultrasonographic-directed fine needle aspiration (EUS-FNA) to diagnose mural-based lesions, aiming to reduce the need for surgical intervention, particularly in asymptomatic cases. We recommend their diagnostic algorithm, which initially distinguishes between mural or luminal gastric lesions, and then, if the lesion is luminal as in the case of gastric cancer, alternative diagnostic methods should be considered. Accordingly, Kagawa *et al.* [12] considered EUS the most useful approach to provide diagnostic details for gastric wall lesions. However, diagnosing GA relies entirely on histological studies, which involves identifying heterotopic tissues, the lesion's architectural pattern, its relationship with the adjacent tissues, and the exclusion of malignancy. The key information provided by EUS is the precise location of the tumor within the gastric wall and the identification of suitable physicians for endoscopic removal of the lesion [4]. GA should be distinguished from both gastritis cystica profunda and high-grade adenocarcinoma [2], and despite being an uncommon tumor, gastric adenomyoma needs to be considered when a gastrointestinal stromal tumor or another mural gastric lesion is suspected [1].

In this present case, we opted for distal partial gastrectomy following endoscopy due to the previously mentioned reasons. A suspicion of malignancy existed since the nature of the symptoms was misleading. To better assess the size and location of the lesion, we chose to perform a CT scan. Furthermore, our definitive diagnosis was only confirmed through complete resection and subsequent pathohistological studies. Heretofore, no GA typical treatment has been stated [8]. In a retrospective study by Wang *et al.* [14] on 15 patients with GA, endoscopic submucosal dissection (ESD) resulted in successful clinical outcomes. They stated that ESD has been repeatedly used in treating small gastrointestinal tract neoplastic lesions as this procedure recorded minimal preoperative pain and trauma, concise period of hospital stays, and low disease recurrence rate. Usually, gastric lesions that cause thickness in the antral and/or pylorus wall are managed with laparoscopic wedge resection [9]. All in all, medical literature included multiple successful surgical methods to treat GA such as endoscopic mass resection, laparoscopic mass resection, complete laparotomy resection (that is, partial gastrectomy), subtotal gastrectomy, or total gastrectomy based on the patient's condition and the severity of the lesion [8].

## Conclusions

Gastric adenomyoma is a rare lesion that can be misdiagnosed as it presents with nonspecific symptoms. Multiple approaches of diagnosis are reported such as radiological studies, endoscopic examination, and endoscopic ultrasonographic directed fine needle aspiration. Treatment options include endoscopic submucosal dissection and complete laparotomy resection. We highly recommend further studies, research, and thorough reviews on this lesion to achieve better knowledge and understanding of the best clinical choices. We also advice practitioners to consider gastric adenomyoma when a mural gastric lesion is discovered.

**Abbreviations**

| | |
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| GA | Gastric adenomyoma |
| GIST | Gastrointestinal stromal tumors |
| CT | Computed tomography |
| MRI | Magnetic resonance imaging |
| EUS-FNA | Endoscopic ultrasonographic-directed fine needle aspiration |
| ESD | Endoscopic submucosal dissection |

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**Author contributions**
AA designed the work concept, drafted the manuscript, and guided the teamwork. SSD participated in academic writing. SMO assisted in academic writing and literature search. ND helped in academic writing and literature search. LA participated in academic writing and extracted the references. RA performed the histopathological examination and collected the patient's data. NM conducted the surgery and documented the patient's follow-up. All authors read and approved the final manuscript.

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## Declarations

**Ethics approval and consent to participate**
Not applicable.

**Consent for publication**
Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

**Competing interests**
The authors declare that they have no competing interests.

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## Publisher's Note