Case Report
Polypoid adenomyoma of the gastric fundus: an unusual endoscopic and pathologic presentation

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Abstract: Gastric adenomyoma (GA) is a rare benign lesion, most frequently found in the pyloric/antral region of the stomach. It consists of a mixture of smooth muscle and glandular structures, occasionally cystically dilated, lined by tall columnar epithelium, Brunner’s glands, and sometimes pancreatic parenchyma. In this report, we describe an unusual case of adenomyoma of the fundus of the stomach, which presented as a polyp on endoscopy, and was found to be a polypoid, cystic lesion pathologically; both of these features are rare for a gastric adenomyoma.

Keywords: Gastric adenomyoma, polypoid, fundus

Introduction

Gastric adenomyoma (GA) is a rare benign lesion, most frequently found in the pyloric/antral region of the stomach. The first case was reported in 1903 by Magnus-Álsleben [1]. It is considered hamartomatous in nature and consists of a mixture of smooth muscle and glandular structures, occasionally cystically dilated, lined by tall columnar epithelium, Brunner’s glands, and sometimes pancreatic parenchyma [2]. GA occurs mainly in adults, but newborns and children are not spared, with ages ranging from 1 week old to 81 years old. Rarely malignant transformation has also been reported [3, 4]. In this report, we describe an unusual case of adenomyoma of the fundus of the stomach, which presented as a polyp on endoscopy, and was found to be a polypoid, cystic lesion pathologically; both of these features are rare for a gastric adenomyoma.

Case report

A 59-year old woman presented with retrosternal pain and a physical examination did not reveal significant abnormalities. All laboratory tests were within normal range and she underwent gastroesophageal endoscopy for heartburn. At endoscopy, the esophagus and duodenum were unremarkable. The stomach showed a small amount of bile but on retroflexed view, a large submucosal, circular mass covered by normal gastric mucosa was seen in the gastric fundus near the gastroesophageal junction. The rest of the stomach was unremarkable. Biopsies of the fundic lesion and of the antrum were taken in an attempt to characterize the lesion and to exclude the presence of H. pylori organisms. The biopsy showed only unremarkable oxyntic mucosa and H. pylori organisms were absent.

The patient underwent a CT scan of the abdomen and pelvis, which failed to identify the gastric lesion. No other significant abnormalities or lymphadenopathy were noted. The only unrelated finding was the presence of an aneurysm of the distal aorta measuring 2.7 cm.

At the follow-up visit, it was decided to remove the fundic lesion by endoscopic excision. During the procedure, mucinous/clear fluid exuded from the cystic lesion.

Macroscopic examination showed a mixed cystic and solid submucosal lesion, which consisted of a piece of polypoid tan-brown tissue measuring 2.0 × 1.4 × 1.5 cm. The lesion was
entirely submitted for histological evaluation. Microscopically, the surface was lined by unremarkable gastric oxyntic mucosa which was embedded within the gastric wall forming an intramural submucosal cyst (Figure 1A). The cyst wall was also lined by gastric foveolar, antral-type epithelium with oxyntic glands, resting on a loose connective tissue, resembling lamina propria, rich in plasma cells, lymphocytes, lymphoid aggregates surrounded by smooth muscle, which was in continuity with the muscularis mucosae of the overlying surface oxyntic mucosa (Figure 1B). The epithelium lining the cyst showed focal reactive changes and was negative for dysplasia. The lumen of the cyst was occupied by a protuberant, polypoid structure connected with the cyst wall by a short stalk (Figure 1A). The polypoid intraluminal structure consisted of broad interdigitating bundles of bland smooth spindle cells in continuity with the smooth muscle of the muscularis mucosae (Figure 1C). No mitotic figures

Figure 1. A: The cystic consists of gastric foveolar-type lined mucosa with an intraluminal polypoid structure attached to the cyst wall by a short stalk. B: The wall of the cyst consists of three layers: the most external facing the gastric lumen is comprised of gastric oxyntic mucosa; the intermediate is shared muscularis mucosa and the proper lining of the cyst which includes gastric foveolar epithelium, antral and oxyntic type glands. C: The polypoid intraluminal structure is comprised of interdigitating bundles of bland smooth muscle. Neither cytological atypia nor mitotic figures are seen. D: The surface of the intraluminal polypoid structure appears partially ulcerated. E: Immunohistochemistry for caldesmon highlights the muscular component of the lesion.
or cytological atypia were seen. The surface of
the polypoid structure was focally lined by
antral-type mucosa with areas of granulation
tissue and fibrinopurulent exudate, consisting
with ulceration (Figure 1D). Neither ectopic
pancreatic tissue, nor Brunner’s glands were
identified within the bundles of smooth muscle
or in the wall of the cyst. Immunohistochemistry
showed the spindle cells to be immunoreactive
for desmin, SMA and caldesmon (Figure 1E).
Immunohistochemistry showed the spindle
cells to be negative for CD117, DOG-1 and
S-100. CD117 highlighted numerous interstitial
cells of Cajal cells intermixed with the smooth
muscles fibres. The histopathological findings
were in keeping with a benign, adenomyoma of
the gastric fundus.

Discussion

GAs are rare benign hamartomatous lesions,
which consist of a variable mixture of bundles
of smooth muscle and epithelial elements in
the submucosa. The epithelial element may
include gastric foveolar glands with/without
goblet cells, Brunner’s glands and pancreatic
elements. Adenomyomas can be found in the
stomach, duodenum, jejenum, ampulla and
gallbladder where they present as solid mural
nodules. Presentation as a cystic polypoid
lesion is rare. The origin of the adenomyoma is
controversial. Gastrointestinal epithelium origi-
nates from endoderm, whereas muscle, con-
nective tissue and peritoneal elements derive
from the mesoderm. Abnormal interactions
between endodermal and mesodermal ele-
ments may result in the formation of adenomy-
omas. Primordial epithelial buds may develop
and variably differentiate, resulting in the spec-
trum of adenomyoma, including aberrant pan-
creas and Brunner’s glands [5, 6]. The smooth
muscle bundles may arise from embryonic
muscle or may represent a proliferation of nor-
mal muscle induced by the misplaced epitheli-
um [7]. These lesions are classified according
to which cell type predominate; aberrant pan-
creas is the most common variant [5].

GAs of the stomach are most frequently found
in the distal stomach, antrum and pylorus. They
may occur at any age and there is a slight pre-
dilection for women [7]. The presentation of
these lesions is quite variable and depends on
the site and size of the lesions. They may be
asymptomatic or cause obstruction, bleeding
with melena [8], and intussusception [9] or
cause non-specific symptoms such as epigas-
tric pain and vomiting. Asymptomatic lesions
can be found incidentally during endoscopy or
imaging for other reasons.

Given the submucosal localization of these
lesions, endoscopic biopsies have a very low
yield [10] and surgical resection is usually nec-
sessary to confirm the diagnosis. Histologically,
the diagnosis generally is straightforward and
based on the presence of bundles of smooth
muscle intimately associated with epithelial
elements. Invasive gastric adenocarcinoma
associated with GAs has been reported [3, 11].
The differential diagnosis depends on the age
of the patient and includes Brunner’s gland
hamartomas, which generally occurs in the first
part of the duodenum, myofibromas of the pylor-
us composed by myoid cells with thin-walled
blood vessels. When epithelial elements are
lacking or scarce, gastrointestinal stromal
tumors (GISTs) and leiomyomas should be con-
sidered [9]. Another condition which may enter
in the differential diagnosis is gastritis cystica
profunda. This condition is characterized by
misplacement of hyperplastic foveolar glands,
cystically dilatated into the gastric submucosal
layer. The lack of smooth muscle bundles
around the cysts helps to distinguish gastritis
cystica profunda from GAs.

To the best of our knowledge this is the first
case in the literature of a GA of the gastric fun-
dus (in close proximity of the gastroesophageal
junction). Our lesion was cystic and contained
an unusual combination of epithelial elements
and smooth muscle: antral mucosa with oxyntic
glands embedded in a loose connective tissue
resembling the lamina propria and associated
smooth muscle from the muscularis mucosae.
These morphological features are unusual for
GAs which typically consists of glands embed-
ded within muscle in the wall. They are rarely
cystic, and this “cyst within a cyst” appearance
is even more unique.

Yoon et al reported a case of GA of the gastric
body [7]; and their lesion shows similar fea-
tures to the GA described herein. Both were
submucosal with mixed cystic and solid compo-
nents, filled with mucinous/clear liquid. How-
ever, histologically, Yoon et al describe broad
interdigitating bundles of smooth muscle pres-
tent between the ducts, which were occasional-
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ly dilated and lined by tall columnar cells [7]. In our case, the cyst was lined by gastric mucosa with antral and oxyntic type glands while the bundles of smooth muscle were mainly protruding into the lumen of the cyst and was in continuity with the smooth muscle of the muscularis mucosae of the overlying oxyntic gastric mucosa.

In conclusion, although GAs are commonly located in the distal stomach, we describe a rare, incidental case of GA of the gastric fundus, which appeared as a polyp on endoscopy, and was a polypoid cystic lesion pathologically. Histologically, it was typified by gastric type mucosa with oxyntic glands, devoid of both Brunner’s glands and pancreatic elements, both of which are rare for a GA.

Disclosure of conflict of interest

None.

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