CASE REPORT

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Gastric adenomyoma

Adenomiom želuca

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Abstract

Bacground. Gastric adenomyoma is a rare, hamartomatous tumor localized most frequently in the gastric antrum. Review of the available literature shows only sporadic reports or smaller series. **Case reports.** We presented a 72-year-old woman admitted due to epigastric pain with dyspeptic difficulties. Biochemical parameters and tumor markers were within the referential limits. Diagnostic procedures (upper endoscopy, endoscopic ultrasonography and computerized tomography) revealed an intramural tumor prominence with intact mucosa on the posterior wall of gastric antrum, not accessible for biopsy. Surgical treatment was performed with total extirpation of the tumor. Histopathological examination verified adenomyoma with focal low grade epithelial dysplasia. Cytologic immunophenotype was consistent with smooth muscle stromal and epithelial tumor (CK_7 and CK_{20} ++ immunophenotype). Stromal component revealed low proliferative index (Ki-67 protein immunoexpression level 3%), and p53 less than 0.1% in both epithelial and stromal components. Following the operation, the patient remained in good condition. **Conclusion.** Uncertain malignant potential of the gastric adenomyoma in the presented case indicates that timely diagnostics with adequate surgical treatment is crucial for an adequate treatment.

Key words:
adenomyoma; stomach neoplasms; diagnosis; treatment outcome.

Apstrakt

Uvod. Adenomiom želuca predstavlja redak hamartomatozni tumor, lokalizovan najčešće u antrumu. Do danas su opisani samo sporadični slučajevi ili manje serije bolesnika sa ovim entitetom. **Prikaz bolesnika.** Prikazali smo bolesnicu, staru 72 godine, koja ja primljena u bolnicu zbog bolova u epigastrijumu i dispeptičnih tegoba. Laboratorijske analize bile su u granicama referentnih vrednosti uključujući i tumorske markere. Urađenim dijagnostičkim procedurama (ezofagogastroduodenoskopija, endoskopski ultrazvuk i kompjuterizovana tomografija abdomena), utvrđeno je postojanje intramuralne prominencije na zadnjem zidu antruma sa intaktnom mukozom. Operativnim zahvatom urađena je ekscizija *in toto* tumorske mase. Patohistološkom analizom utvrđeno je postojanje adenomioma, sa niskostepenom epitelijalnom displazijom, sačinjenog od kombinovanog epitelijalno-mezenhimalnog tumora. Imunofenotipizacija je pokazala postojanje stromalnog (lejomiomskog) i epitelijalnog tumora sa displazijom (CK_7 i CK_{20} ++), Ki_{67} indeksom 3% i p53 indeksom manjim od 0,1%. Postoperativni tok je protekao uredno i bolesnica je otpuštena u dobrom opštem stanju. **Zaključak.** Nejasni maligni potencijal adenomioma sa displazijom kod ove bolesnice pokazuje da je za uspešno lečenje neophodno brzo postavljanje dijagnoze i adekvatano hirurško zbrinjavanje.

Ključne reči:
adenomiom; želudac, neoplazme; dijagnoza; lečenje, ishod.

Introduction

Gastric adenomyoma is a rare hamartomatous tumor. It is a combined epithelial-stromal proliferation most frequently localized in the gastric antrum (85%), about 4 cm from the pylorus or rarely in peripyloric region (15%) [1, 2]. Delvaux classified submucosal epithelial lesions as follows: hamartoma, gastric glandular heterotopy, submucosal glandular cysts, gastritis cystica profunda and adenoma [3].

Histologically, adenomyoma is composed of hypertrophic smooth muscle fibers of the stroma surrounded by epithelial elements in the form of individual glandular struc-

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tures, Brunner's glands, pancreatic ducts and acini [2]. If tumor is predominantly composed of pancreatic tissue, the most adequate term would be heterotopic or residual pancreatic tissue [1].

We present the case of gastric adenomyoma, with focal epithelial dysplasia with uncertain malignant potential.

Case report

A 72-year-old female patient was admitted to the Department of Digestive Surgery due to epigastric pains, followed by dyspeptic difficulties.

Rutine laboratory tests conducted at the time of referral to the hospital were within the normal ranges including inflammatory parameters. In addition, tumor markers were also within physiological ranges (CEA 2.1 ng/mL, CA 19–9 15.5 ng/mL). Physical examination failed to reveal significant abnormalities; there were no intraabdominal masses and no supraclavicular lymph nodes were palpable.

The patient underwent upper endoscopy demonstrating a submucosal mass in the distal antrum. Multiple biopses were taken, but histology was inconclusive about the nature of the lesion, showing moderate superficial gastritis with foveolar hyperplasia, while *Helicobacter pylori* status was negative.

Endoscopic ultrasonography confirmed the intramural, extramucous prepyloric tumor 25 mm × 15 mm in diameter (Figure 1).

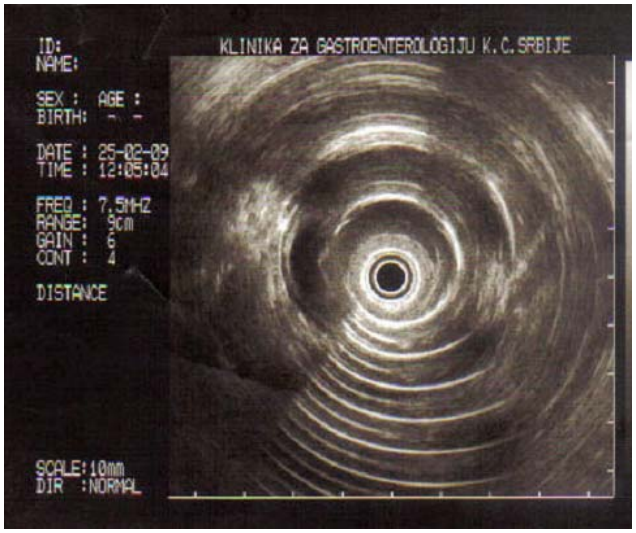

Fig. 1 – Endoscopic echosonography revealed the intramural, extramucous change of 25 × 15 mm in size on the gastric antrum posterior wall

Abdominal CT scan with contrast enhancement revealed the intramural, well-defined, solid-cystic lesion in the distal antrum.

Due to the clinical impression of gastric stromal tumor, a wedge resection was performed (Figure 2). Macroscopic examination revealed the mixed cystic-solid submucosal mass (25 × 15 × 10 mm) with an intact overlying mucosa.

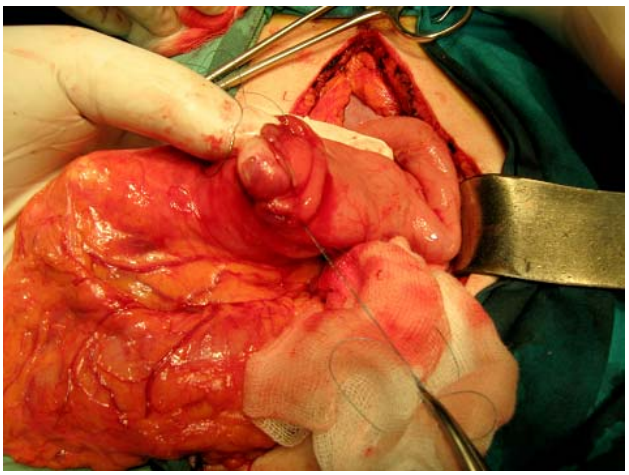

Fig. 2 – The oval, extramucous tumor verified intraoperatively on the posterior wall of the gastric antrum, and extirpated *in toto*

Microscopic examination with hematoxylin eosin staining verified the adenomyoma combined of the epithelial and mesenchymal cells of the distal antrum with epithelial focal low grade dysplasia. According to the cytologic immunophenotype the lesion was consistent with enteral (epithelial) and leiomyogenome (stromal) tumor (Figure 3). Epithelial component revealed CK7 and CK20 positive immunoexpression, with Ki_{67} index of 3 % and p53 less than 0.1% in stromal component. Stromal component manifested low cellularity and low cellular anaplasia with extremely low mitotic index of 0/50 HPF. There was no tumor invasion to adjacent structures and tumor necrosis, while the surface of the lesion, showed some focal pseudocystic degeneration (Figure 4).

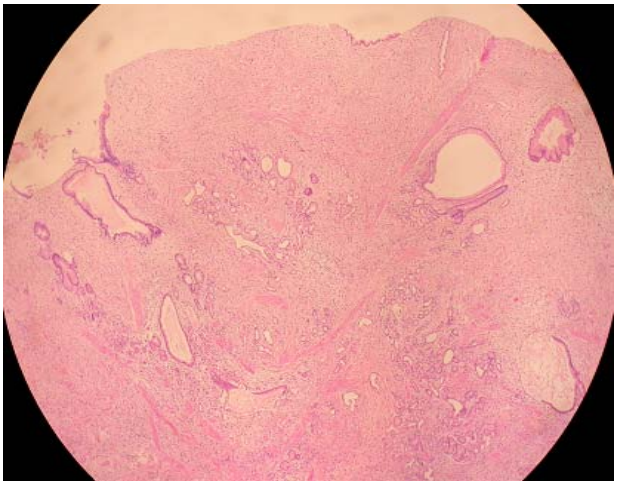

Fig. 3 – Epithelial and stromal overgrowth in a characteristic mixture revealed adenomyoma

Postoperative course was uneventful. Peroral intake started on the 4th postoperative day, and the patient was discharged on the 9th postoperative day.

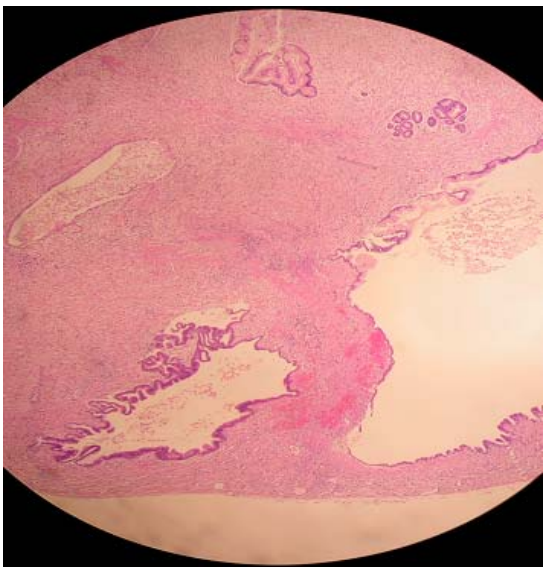

Fig. 4 – Focal low grade dysplasia of adenomyoma with pseudo-invasive features

Discussion

Gastric adenomyoma is an extremely rare benign tumor described also as: myoglandular hamartoma, adenomyomatous hamartoma, gastric adenomyoma, adenomyosis or heterotopy.

The first tumor description dated back in 1903, when Magnus Alsleben presented 5 gastric adenomyomas based on autopsy results. Many years later, in 1955, Cimmino described the gastric adenomyoma as an individual entity hypothesizing that adenomyomas result exclusively from chanes in a gastric wall including aberrant pancreas, Brunner's glands and non-defined myoepithelial growth.

Although gastric adenomyoma are generally accepted to be rare gastric tumors with only sporadic cases described in the literature to date, it has recently been suggested that they may be more common. Ling and Situ [4] reported 9 cases, and Barnert et al. [5] presented a single case of gastric adenomyoma. The largest series so far, having involved 33 patients with gastric adenomyoma, was published by Vandelli et al. [6] in 1993.

While gastric adenomyomas are very rare, pancreatic heterotopy is found in postmortem material in 0.55− 5.7% of the autopsies. It is most commonly localized in the projections of the antrum, pylorus, beginning of the duodenum or jejunum [7].

Histogenesis of the gastric adenomyoma is unknown, but it is believed that it results from developmental abnormalities. It is commonly diagnosed from fourth to sixth decade of age (66%), although there have been some cases described even in 8 months old infants, while the oldest patient was 81 years old [6].

Although gastric adenomyoma should be considered to have uncertain malignant potential, cases with aggressive behaviour have been reported. Chapple et al. [1] were first to describe an extremely rare case of simultaneous presentation of adenomyoma and gastric adenocarcinoma. Kanehira et al. [8] presented the case of associated adenomyoma and superficial adenocarcinoma centered in adenomyoma. Agresta and Della Libera [9] described the synchronous manifestation of adenomyoma and gastric antrum adenocarcinoma together with Hodgkin's lymphoma of the cardia. Nevertheless, the aforementioned sporadic reports cannot support the fact of the adenomyoma-based cancer. Descriptions of the individual cases of gastric adenomyoma associated with gastric malignancy have underlined that the development of neoplasm had been diagnosed long before adenomyoma was detected [1, 8, 9]. Although relation between these two pathological entities remains indefinite, the possibility of malignant alteration of adenomyoma cannot be ruled out with certainty.

The majority of described cases was diagnosed by gastroduodenal contrast medium radiography, in lesser number of cases using an esophagogastroduodenoscopy, and a number of cases was diagnosed during surgery incidentaly or at post-mortem examination.

Adenomyomas often appear as a roundish or oval-shaped mass in the submucosal tissue simulating a lipoma, a neurofibroma or a polypoid formation [10, 11]. Endoscopic biopsy of the mucosa fails to diagnose this benign tumor [12, 13]. Therefore, Hedembroa et al. [13] believe that gastric resection and extirpation of a tumor mass *in toto* is mandatory in order to distinguish the nature of a submucosal lesion.

Treatment of the gastric adenomyoma should be exclusively surgical including a laparoscopic resection. So far, no recurrence after a successful surgical resection has been recorded. Radical surgical treatmant should be reserved only for cases in who benign nature of the tumor is not confirmed [2].

In our case, the upper endoscopy showed the submucosal mass with intact mucosa in distal antrum. However, unless a concern for an alternative diagnosis, the use of biopsy is not recommended in this setting, as most of the lesions, including our case, are localized in the submucosal layer.

Conclusion

Epithelial dysplasia in the presented case indicates that timely and proper diagnostics as well as adequate surgical treatment are crucial for a successful treatment of gastric adenomyoma.

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