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CASE REPORT / ПРИКАЗ БОЛЕСНИКА

Combined laparoscopic-endoscopic "rendez-vous" procedure in a case of gastric schwannoma in a toddler

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SUMMARY

Introduction Schwannomas are rare benign tumors of the gastrointestinal tract. Despite the differences in features of schwannomas located in the stomach as opposed to peripheral or soft tissue schwannomas, their immunohistochemical characteristics are the same. We present a case of a 14-month-old boy with a gastric tumor who underwent a combined laparoscopic-endoscopic resection surgery, followed by gastric schwannoma diagnosis.

Case outline The patient was admitted to our pediatric hospital with a fever of unknown origin. Endoscopy, performed after recurrent hematemesis, revealed an ulcer in the gastric antrum. At the four-week follow-up, gastroscopic and microscopic findings were normal. Two weeks later, a flank mass in the antrum was detected by an ultrasound examination. A new gastroscopy and CT scan confirmed the presence of a tumor-like mass, 5 cm in diameter. A combined laparoscopic-endoscopic polypectomy was performed with a necessary conversion for complete resection of tumor. The initial histological findings were consistent with a gastrointestinal stromal tumor. Due to this tumor's rarity in childhood, the paraffin-embedded tissue samples were referred for a second opinion. Histological and immunohistochemical characteristics of the tumor made the gastrointestinal stromal tumor diagnosis unlikely and consistent with a completely resected gastric schwannoma. No recurrence of the disease occurred during the seven-year follow-up.

Conclusion Combined laparoscopic-endoscopic surgery is a feasible and effective treatment for large gastric tumors that cannot be excised endoscopically. Schwannoma should be included in the differential diagnostic consideration of gastric tumor lesions even in childhood.

Keywords: stomach; laparoscopy; endoscopy; neurilemmoma

INTRODUCTION

Gastrointestinal (GI) schwannoma was first described in 1988 by Daimaru et al. [1]. The stomach is the most common site, and gastric schwannoma (GS) represents 0.2 % of all gastric neoplasms [2]. They are believed to arise from the disperse autonomic nerve Schwann cells, in contrast to much more common schwannoma of the skin, connective tissue, and other internal organs that arise from peripheral nerves. These differences are also related to schwannomas' gross and histological features in the gastric location [2]. Also, unlike peripheral schwannoma, GS is rarely associated with neurofibromatosis, so it probably has a different genetic basis [3, 4, 5]. However, the immunohistochemical characteristics of schwannomas in all locations are the same. Immunohistochemistry plays a pivotal role in distinguishing GSs from more common mesenchymal tumors in the GI tract: gastrointestinal stromal tumor (GIST), smooth muscle neoplasms and inflammatory fibroid polyp [2–5].

GS occurs predominately in adults and is more common in women. The symptoms are nonspecific and it may be detected incidentally [2, 4, 6].

Endoscopy with simultaneous resection of the lesion is highly advisable for GI lesions in mucosal and submucosal locations. Still, open or laparoscopic surgical resection has been the only available treatment in most cases of deeply sited gastric neoplasms with excellent postoperative prognosis [2, 4, 7, 8].

We present a case of a 14-month-old boy with a gastric tumor who underwent a combined laparoscopic-endoscopic resection procedure, followed by the histopathological diagnosis of GS.

CASE REPORT

The patient was admitted to our pediatric clinic with a fever of unknown origin. Clinical examination did not reveal the cause of the fever. Non-steroidal anti-inflammatory drugs were administered. After recurrent hematemesis, on the seventh day of the hospitalization, endoscopy was performed, which revealed a large ulcer with a diameter of 38 mm in the stomach's

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antropyloric segment (Figure 1A). Afterwards, the patient was under a clinical follow-up and was treated with pantoprazole. After a four-week follow-up, gastroscopic and microscopic findings were normal (Figure 1B). Two weeks later, a flank mass in the antrum was found by an ultrasound examination. New gastroscopy and CT scan confirmed the presence of a well circumscribed, polypoid, submucosal tumor mass measuring 4.8 × 4 × 4 cm, lined with flattened mucosa, in the antropyloric region.

A combined laparoscopic-endoscopic resection, the so-called "rendez-vous" procedure, was performed the next day. Under general anesthesia, the endoscope was inserted, and the location and diameter of the tumor was confirmed (Figure 1C). Then the camera port was inserted in a standard infraumbilical position using an open Hasson technique. A carbon dioxide pneumoperitoneum was established, with a pressure of 8–10 mmHg. Two additional ports (5 mm) were inserted into the left and right upper quadrants. Firstly, a front abdominal wall gastrotomy was performed using endoscopy navigation. Then an excision of the tumor in its entirety was made. The standard laparoscopic operation was practically converted to video-assisted procedure (Figure 1D). It means that the stomach was exteriorized through umbilical port, which was slightly enlarged, and polypectomy was performed extracorporeally. As a result, the defect remained on the back wall due to the radicality of the resection. The operation was completed with the closure of the posterior and anterior stomach wall, both with direct sutures. Finally, a nasogastric tube was placed.

Initially, the tumor, which measured 5 cm in the largest diameter (Figure 1E), was diagnosed as GIST. Due to the extreme rarity of GIST in childhood, tissue samples were sent for a second opinion (Belgrade). The tumor was composed of elongated spindle cells with mild nuclear atypia and moderate mitotic activity (15 mitoses / 50 high-power field). Interstitium was myxomatous with a decreased density of tumor cells. Except in the central area of the ulceration, the tumor was lined with atrophic mucosa. The tumor at the base showed infiltrative growth between the muscle propria fibres (Figure 2A–C). After immunohistochemical staining, tumor cells were positive for vimentin, S-100 (Figure 2D), GFAP, focally CD34, and negative for desmin, muscle-specific and smooth muscle actin, caldesmon, calponin, CD117, DOG1, ALK-1, CK, EMA, BCL-2, CD99, and TLE-1. Histological and immunohistochemical characteristics of the tumor made the diagnosis of GIST extremely unlikely and were consistent with GS. Resection margins were tumor-free. Histological and immunohistochemical analyses were repeated by an expert team (Liverpool, UK), who agreed with the diagnosis of GS. Due to the lack of clinical criteria and negative family history, neurofibromatosis was also unlikely.

The postoperative course was uneventful. No adjuvant therapy was administered. There was no recurrence of the tumor during seven years of follow-up (Figure 1F).

This study was designed as a case report and was conducted according to the guidelines of the Declaration of Helsinki. It was approved by the Institutional Review Board of the Institute for Child and Youth Healthcare of Vojvodina (No 2284 from June 4, 2021).

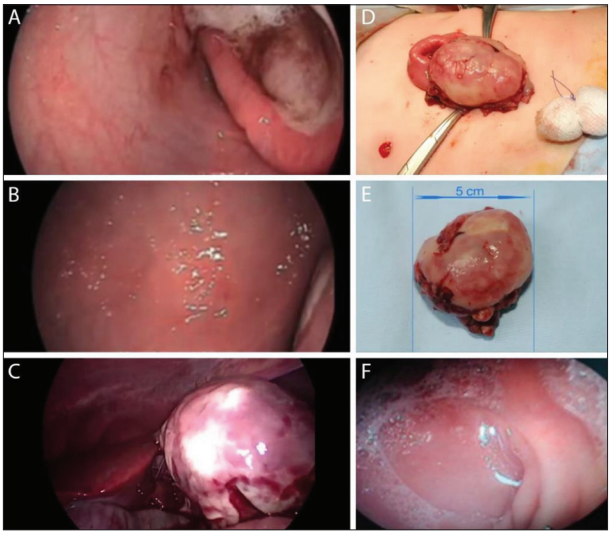

Figure 1. Diagnostic and treatment procedures; A: a large ulcer in the gastric antrum found on the first endoscopy; B: a normal finding on the second endoscopy; C: laparoscopic view of the tumor after opening the anterior wall of the stomach; D: conversion to video-assisted surgery; E: macroscopic appearance of the tumor; F: postoperative endoscopy

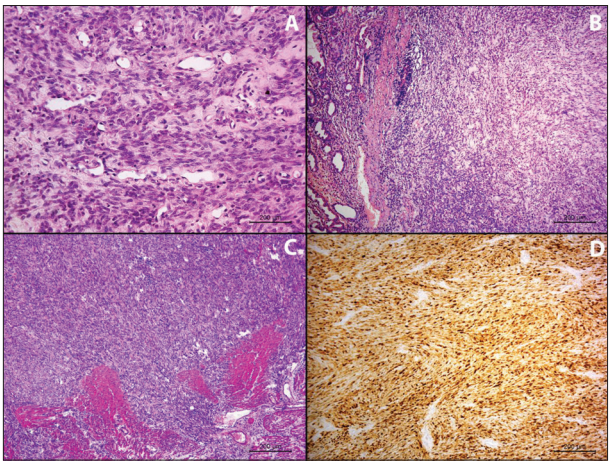

Figure 2. Histology of the gastric schwannoma; A: microtrabecular growth pattern of relatively uniform spindle cells and dilated capillaries in the stroma (H&E, × 200); B: atrophic gastric mucosa above the tumor (H&E, × 100); C: infiltrative growth through the muscularis propria at the base of the tumor node (H&E, × 50); D: immunohistochemically typical diffuse tumor cells positivity for S-100 protein (× 100)

DISSCUSION

Most tumors located in the stomach above the muscularis propria can be excised endoscopically. The available techniques comprise snare polypectomy, endoscopic mucosal resection (EMR), and piecemeal EMR [9, 10]. If the tumor is located in the deeper layers, the possibility of complete resection is questionable. The optimal treatment for such tumors is an open or laparoscopic surgery [11, 12].

Laparoscopic wedge resection is being performed with increasing frequency [13, 14, 15]. In 2004, the National

Comprehensive Cancer Network and The European Society of Medical Oncology published consensus statements doubting the efficacy of laparoscopic resection for GIST of the stomach [16]. In this report, laparoscopic resection is approved for GIST smaller than 2 cm. According to recent publications, many surgeons achieved complete resection (R_0) laparoscopically, with no complications in terms of rupture or tumor spillage [17].

Combined laparoscopic-endoscopic techniques represent an innovative concept in the treatment of colonic and gastric lesions. The idea for this approach came from the paper by Pelizzo et al. [18] published in 2007. The first procedure in our clinic was performed 13 years ago to treat a patient with familial adenomatous polyposis syndrome. Minimally invasive "rendez-vous" surgery allows for the opportunity to make an accurate histological analysis while a high level of patient comfort is maintained [19, 20].

About three-quarters of GI tract schwannomas are located in the stomach, most commonly in the area of the great curvature, followed by antrum and fundus [4, 5, 6, 8]. Lauricella et al. [8] found 686 patients with GS published in the English-language literature over a period of 30 years. Most of the articles dealt with individual cases. Only 10% of all tumors were removed endoscopically, while an equal number of other tumors were removed by total/subtotal gastrectomy and local excision, respectively. Tumor recurrence was very infrequent and did not depend on the surgical method.

All of 221 patients with GS in the literature review by Hu et al. [6] were older than 40 years. The youngest registered patient with GS so far was a 16-year-old girl [21].

The histological features of the tumor in our patient confirm that GS histologically differs somewhat from peripheral or soft tissue schwannoma. Unlike soft tissue schwannoma, GS is usually not encapsulated, lacks nuclear palisading (Verocay bodies), alternating areas of hypercellularity (Anthony A and B), vascular hyalinization and dilatation. GS often has a peritumoral cuff-like lymphocytic infiltration, microtrabecular architecture and cellular atypia [4, 6]. Mitotic activity in our tumor was at the upper limit (criteria for separating benign from malignant GS) [6]. GS is very rarely associated with neurofibromatosis 1 and 2 syndrome.

Immunohistochemical analysis remains the main diagnostic method for distinguishing spindle cell tumors, including mesenchymal tumors of the gastric wall. In our case, repeated analyses in two laboratories specialized in pediatric pathology showed negative immunohistochemical staining results with markers characteristically positive in GIST (CD117, DOG1). The key markers for schwannoma differentiation, S-100 and GFAP, were positive in this case. Also, the immunophenotype of tumor cells did not correspond to some of the very rare childhood tumors (myofibroblastic neoplasms, solid fibrous tumor, synovial sarcomas, and some types of hemangioendotheliomas).

Combined laparoscopic-endoscopic polypectomy is a feasible and effective treatment for large gastric tumors that cannot be excised endoscopically. This is a safe technique that enriches the therapeutic range of the surgeon and healthcare institution in which it is performed. This case suggests that schwannoma should be included in the differential diagnostic consideration of gastric tumor lesions even in childhood.

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Conflict of interest: None declared.

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Комбинована лапароскопско-ендоскопска „рандеву" процедура у случају гастричног шванома код малог детета

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САЖЕТАК

Увод Шваноми су ретки бенигни тумори гастроинтестиналног тракта. Без обзира на разлике у особинама шванома желуца и шванома периферних нерава, њихове имунохистолошке карактеристике су исте. Представљамо четрнаестомесечно дете са гастричним тумором који је ресециран комбинованом лапароскопско-ендоскопском техником, након чега је постављена дијагноза гастричног шванома.

Приказ болесника Болесник је примљен на нашу педијатријску клинику због фебрилности непознатог узрока. Након понављаних хематемеза, током ендоскопије откривен је улкус у антруму желуца. Након четири недеље, резултати гастроскопије и микроскопске анализе били су уредни. Две недеље касније, током ултрасонографског прегледа виђена је полипоидна формација у пределу антрума. Поновљена гастроскопија и КТ потврдили су присуство туморске масе пречника 5 *cm*. Урађена је комбинована лапароскопско-ендоскопска полипектомија са неопходном конверзијом у циљу комплетне ресекције тумора. Иницијални хистолошки резултат указивао је на гастроинтестинални стромални тумор. Због реткости овог тумора у дечјем узрасту, послате су парафинске плочице ради добијања другог мишљења. На основу хистолошке и имунохистохемијске анализе у лабораторијама педијатријске патологије утврђено је да се ради о гастричном шваному, који је у целини одстрањен. Није било рецидива болести током седмогодишњег праћења болесника.

Закључак Комбинована лапароскопско-ендоскопска ресекција могућа је и ефикасна терапија за лечење туморских лезија у зиду желуца које се не могу одстранити ендоскопски. Шваном је неопходно укључити у диференцијалнодијагностичка разматрања туморских лезија желуца, чак и у дечјем узрасту.

Кључне речи: желудац; лапароскопија; ендоскопија; неурилемом