Multiple Gastric Antrum Mucosal Schwann Cell Hamartoma on Incidental Screening Gastroscopy: A Case Report

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Case Report

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Abstract

Background Mucosal Schwann cell hamartoma (MSCH) is a newly recognized neural lesion of the gastrointestinal (GI) tract. MSCHs are mostly solitary and occur in the colon and rectum. We present a case with multiple MSCHs arising in gastric antrum.

Case presentation A 49-year-old woman with atrophic gastritis was accidentally found to have multiple mucosal protrusion lesions in the gastric antrum during endoscopic follow-up. Biopsy specimens revealed a spindle cell lesion in the lamina propria with positive reaction to the S-100 and SOX-10 protein. The final diagnosis was MSCH.

Conclusions: MSCH is a benign mucosal spindle cell lesion of the GI tract and does not associate with inherited syndromes. It is most commonly seen in the colorectum, but can also be found in stomach and gallbladder. In this case, MSCH occurred in gastric antrum with multiple lesions. MSCH is benign lesion and successive treatment, work-up and follow-up are unnecessary.

Background

Mucosal Schwann cell hamartoma (MSCH) is a newly recognized neural lesion of the gastrointestinal tract (GI), first proposed by Gibson and Hornick in 2009 [1]. MSCH is characterized by an ill-defined proliferation of spindle cells within the lamina propria purely composed of S100-positive Schwann cells. Different from the neural lesions that have significant associations with inherited syndromes, such as neurofibromatosis type-1 (NF1) and multiple endocrine neoplasia type 2B (MEN 2B), MSCHs have no known syndromic association but are often incidental findings. Most reported MSCHs in the literature occur in the colon and rectum, predominantly in the rectosigmoid region. Only few cases were reported in the gastroesophageal junction (GEJ) [2] and gallbladder [3]. Only one case was reported in gastric antrum [2], which appeared as isolated polypoid nodule. In this report, we are reporting a case of MSCH with multiple patchy mucosal uplifts arising in gastric antrum.

Case Presentation

A 49 year-old woman underwent gastroscopy due to upper abdominal pain, upper abdominal distension and acid reflux. Gastroscopy showed atrophic gastritis. The symptoms abated after treatment. Gastroscopy in follow up after one year showed that multiple patchy mucosal uplifts with slight depressions in the center in the gastric antrum (Fig. 1a, b). Atrophic gastritis was still present. No significant abnormalities were found on laboratory and other tests. Patient hospitalized and treated with gastroscopic electrosurgery of the gastric sinus Schwann cell tumor. Two biopsy specimens revealed spindle cell lesions in the lamina propria (Fig. 2a). The spindle cells had eosinophilic cytoplasm and oblong nuclei with certain neural pattern and without cellular atypia (Fig. 2b). Mitosis, necrosis and ganglion cells were not seen among the spindle cells. The lesions were considered suspicious for a gastrointestinal stromal tumor (GIST) firstly. On immunohistochemical stains, the lesional spindle cells

were diffusely and intensely positive for S-100 and SOX-10 protein (Fig. 2c, d). There was no positivity for synaptophysin, neurofilament protein, smooth muscle actin, desmin, DOG1, CD34, CD117 and Ki-67. Extensive sampling of the gastric mucosa showed moderate chronic gastritis with mild activity, mild atrophy and extensive intestinal metaplasia of the epithelium. No additional spindle cell lesions were found. There was no reported family history of personal history of familial hereditary polyposis syndromes, neurofibromatosis, von Recklinghausen syndrome or Cowden syndrome. The final histological and immunohistochemical results confirmed the diagnosis of Multiple Gastric Antrum Mucosal Schwann Cell Hamartoma.

The patient was treated with acid suppression, gastric protection and symptomatic support after the gastroscopic electrosurgery. After four days of hospitalization, the patient was discharged without any short-term postoperative complications. The gastroenterologist recommended that the patient be discharged with omeprazole enteric dissolved tablets, Rebapent tablets and rehabilitative new liquid, and after two months of follow-up observation, the patient is generally doing well.

Discussion And Conclusions

MSCHs occur mostly in the colon and rectum that showed solitary single spindle cell proliferation without axons or ganglion, were not associated with hereditary diseases, revealed a positive reaction to the S-100 protein in immunohistochemical staining. MSCHs of the GI tract are rare benign lesions, which were different from other neural tumors [1]. Recently, a few cases in colon and rectum [5-8], GEJ [2] , stomach [4] and gallbladder [3] have been reported successively. The literatures showed there was a slight female predominance and most often occur in elder people. MSCH usually produces no symptoms and is detected by chance in most of the cases during gastrointestinal endoscopy performed as routine screening, but the patient may present with diarrhea, melena, abdominal pain or abdominal discomfort, and anemia [9]. This case presented abdominal pain, upper abdominal distension and acid reflux, which may be symptoms of atrophic gastritis. Gastrointestinal endoscopy shows that solitary small polyp accompanied or not accompanied by other lesions. In this report, we describe a case of multiple MSCH arising in gastric antrum on the background of atrophic gastritis. The lesions were mucosal uplifts with slight depression rather than polypoid, consisting of poorly circumscribed proliferation of spindle cells in the lamina propria, with benign cytological appearance and by pure Schwann cell immunophenotype. The histologic and the immunohistochemical features of our case were certainly consistent with MSCH.

The histological differential diagnosis of spindle cell proliferation in the stomach includes GISTs, neural tumors, fibrous lesions, and neoplasms of smooth muscle. It is easy to distinguish MSCH from GISTs, fibrous lesions, and neoplasms of smooth muscle with immunohistochemical stains. GISTs are characterized by peculiar immunoreactivity to C-KIT/CD117 and DOG1 antibodies. Fibrous lesions, such as inflammatory fibroid polyps and benign fibroblastic polyps, are benign fibrous lesions negative for S-100. In neural cell tumors, schwannoma, neurofibroma, mucosal neuroma, perineurioma and granular cell tumor are the main differential diagnosis. Schwannoma is the most challenging differential diagnosis, because Schwann cell hamartoma and schwannoma are both proliferation of pure Schwann cells.

However, they are different diseases. Schwannoma is a tumor showing expansive proliferation, with S-100- positive cells and/or a lymphoid cuff in the gastrointestinal tract. Unlike schwannomas, Schwann cells proliferate between gastric foveola and lamina propria in MSCH. The lack of axons and ganglion cells argues against lesions such as ganglioneuromas, neurofibromas, or mucosal neuromas. Moreover, these tumors display significant association with inherited syndromes. Neurofibromas have a strong association with NF1 and mucosal neuroma is highly associated with MEN 2B. The case in our report showed no signs of NF1, MEN 2B, Cowden syndrome, or other inherited syndromes. Perineuriomas have perineurial cell differentiation and are positive for glucose transporter-1 and claudin-1 instead of S-100 protein. Granular cell tumor had numerous eosinophilic granules in cytoplasm and oval cell nucleus in contract to spindle cells in MSCH.

MSCH is an uncommon mucosal spindle cell lesion of the GI tract. It is most commonly seen in the colorectum, but can also be found in stomach and gallbladder. MSCH does not associate with inherited syndromes. MSCH is benign lesion and additional treatment, work-up and follow-up are unnecessary.

Abbreviations

MSCH: Mucosal Schwann cell hamartoma; GI: Gastrointestinal; NF1: Neurofibromatosis type-1; MEN 2B: Multiple endocrine neoplasia type 2B; GEJ: Gastroesophageal junction; GIST: Gastrointestinal stromal tumor

Declarations

Ethics approval and consent to participate

Written informed consent was obtained from the patient. The ethical approval and documentation for a case report was waived by Shandong Provincial Hospital affiliated to Shandong First Medical University.

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Informed consent statement

Informed consent was obtained from the patient.

Written informed consent

Patient has provided informed consent for publication of the case.

Conflict-of-interest statement

The authors declare that there is no conflict of interest related to this report.

Consent for publication

The patient has given informed consent.

Availability of data and materials

All data generated or analysed during this study are included in this published article.

Authors's contributions

Chang Lu is a major contributor in collecting and sorting out the basic data. Jizhen Feng revised the manuscript. Shulei Zhao provided clinical data. Zhigang Yao and Xiankui Cheng helped revising the manuscript. Jiamei Li wrote the manuscript, designed and organized the study. This manuscript has been read and approved by all authors. This manuscript has been read and approved by all authors.

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Figures

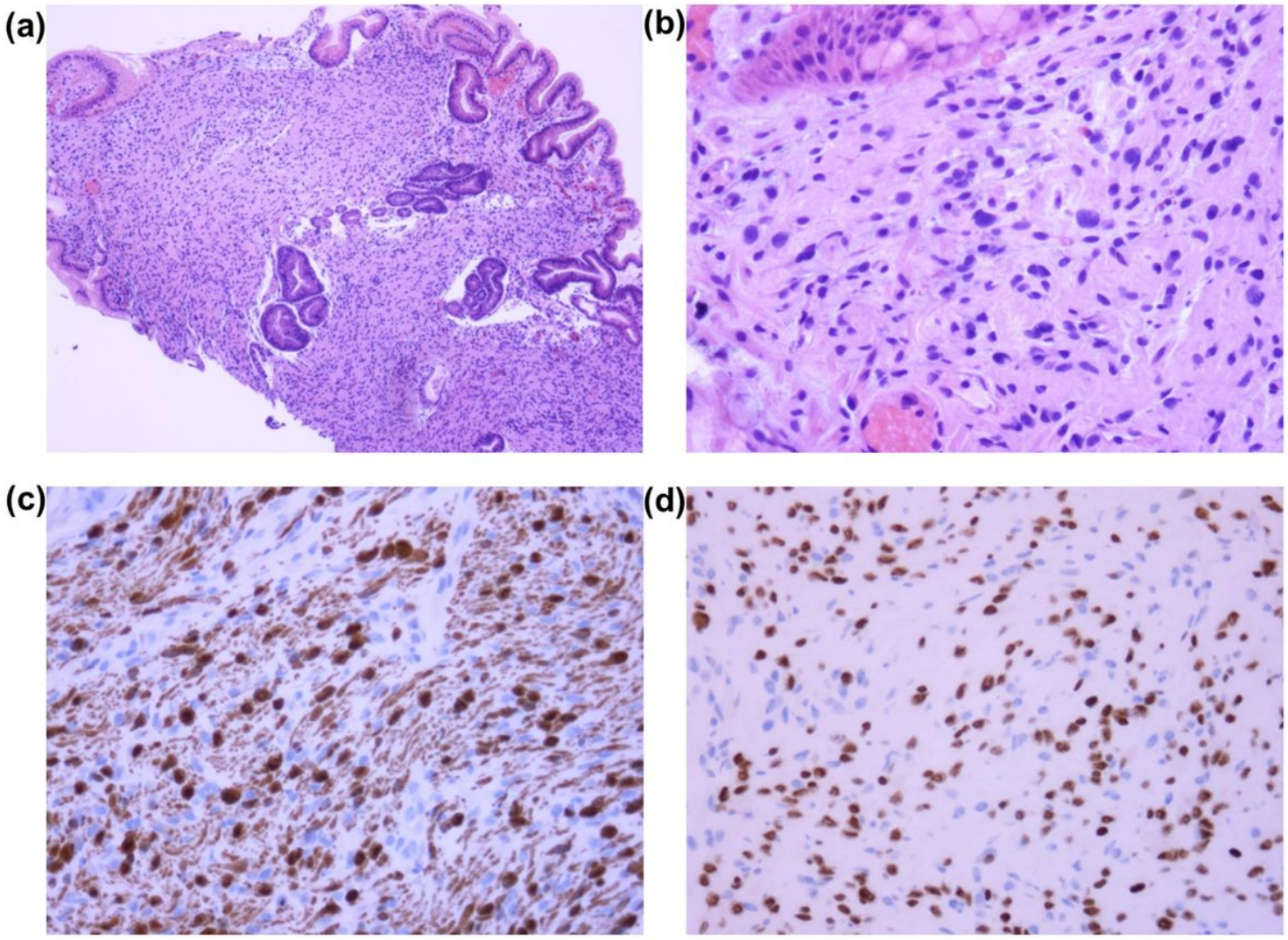

Figure 1

Endoscopic findings from stomach using gastroscopy. a, b Photos taken from gastric antrum showed multiple patchy mucosal uplifts with slight depressions in the center.

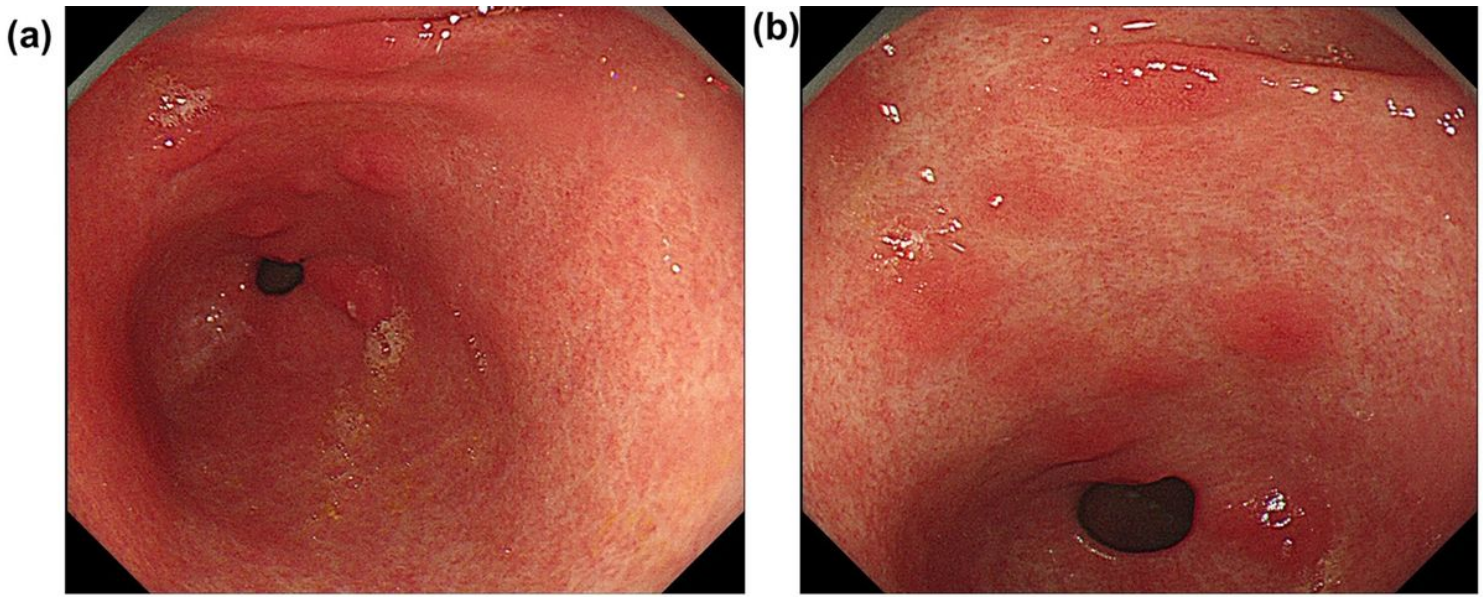

Figure 2

Histological findings and immunophenotype. a A diffuse proliferation of spindle cells in the lamina propria [hematoxylin and eosin (HE) 40×]. b A diffuse cellular proliferation of uniform bland spindle cells with elongated, tapering nuclei, abundant, dense eosinophilic cytoplasm, and indistinct cell borders (HE, 200×). c The spindle cells of the lesion show intense, diffuse positivity for S-100 protein (original magnification ×100). d The spindle cells of the lesion show intense, diffuse positivity for SOX-10 protein (original magnification ×100)

Supplementary Files

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