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Case Report: We are reporting a case of a solitary hamartomatous polyp in the duodenum. A 24-year-old female was referred to our hospital with a diagnosis of duodenojejunal intussusception. We identified a polyp in the distal part of the duodenum. The patient underwent an endoscopic polypectomy. The polyp was confirmed to be a Peutz-Jeghers type of hamartomatous polyp. The patient underwent colonoscopy and small intestines follow-up with no evidence other polyps.

Conclusion: Previous reports have shown that solitary Peutz-Jeghers type hamartomatous polyps have malignant components in some cases. Patients with a duodenal solitary hamartomatous polyp should undergo treatment with complete removal of polyp and they need periodical follow-up examinations.

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CASE REPORT

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INTRODUCTION

Peutz-Jeghers syndrome (PJS) is an autosomal dominant disorder characterized by gastrointestinal hamartomatous polyps in association with mucocutaneous hyperpigmentation [1, 2]. A solitary Peutz-Jeghers type hamartomatous polyp has been considered a variant or separate disease entity without features of PJS [3]. A solitary hamartomatous polyp is histologically the branching of smooth muscles, it is derived from muscularis mucosae, and the overlying mucosa is typically non-dysplastic and maintains its normal architecture. Such hamartomatous polyps occur predominantly in the small intestines, colon and stomach. As compared with Peutz-Jeghers syndrome, Peutz-Jeghers type hamartomatous polyps are diagnosed as having a lower risk of cancer.

CASE REPORT

We are reporting an experience of a solitary hamartomatous polyp in the distal part of duodenum that led to bowel intussusceptions. A 24-year-old woman presented long-term epigastric pain and abdominal discomfort. Upon physical examination, the abdomen

was not tender, no palpable masses were observed, and the vital signs of the patient were normal. Mucocutaneous pigmentation of the perioral region, buccal mucosa, hands and feet was absent. The patient had no family history of Peutz-Jeghers syndrome. Standard laboratory test results were unremarkable. Abdominal computed tomography (Figure 3) revealed an intussusception of the distal part of the duodenum into the jejunum with suspicion of intraluminal expansion. An upper gastrointestinal endoscopy (Figure 1) showed a pedunculated polyp largely filling the lumen of the distal part of duodenum. Under general anesthesia, we introduced a lateral – viewing instrument (TJF-Q180V, Olympus, Tokyo, Japan) into the duodenum, where there was a huge polyp measuring 50 mm in diameter on a wide stem. We performed an endoscopic polypectomy piece wise with a snare (SD-990-25, monofilament, 25mm, Monheim am Rhein, Germany). After removal of the stem, we treated the bottom layer with a non-bleeding artery using argon plasma coagulation (APC). We did not use a balloon-assisted endoscopy due to limited anatomical conditions, which were defined by a large polyp. Our procedure was without complications. A histological examination of the specimens (Figure 2) showed a hamartomatous polyp with a focus of foveolar metaplasia. The patient underwent colonoscopy and small intestines follow-up (MR enterography, capsule enteroscopy) with no evidence other polyps. After endoscopic treatment, concomitant cancer was not found. Patient is monitored and undergoes regularly endoscopic examination of gastrointestinal tract.

DISCUSSION

A hamartomatous polyp without associated mucocutaneous pigmentation or family history of PJS is diagnosed as a solitary Peutz-Jeghers type hamartomatous polyp. It is considered to be either an incomplete variant of PJS or a separate disease entity [3].

Analysis of the loss of heterozygosity at the locus of 19p13.3 and mutation analysis of the *STK11/LKB1* gene were performed, and no evidence of genomic abnormality was found. Such hamartomatous polyps occur predominantly in the small intestines, colon, stomach and rarely arise from the appendix [4–7]. A solitary Peutz-Jeghers type hamartomatous polyp in the duodenum is rare. In previous reports 31 cases of solitary Peutz-Jeghers type hamartomatous polyp in the small bowel were described. The most common site of occurrence of solitary hamartomas in the small bowel is duodenum (mostly second part), less frequently jejunum and ileum. In clinical presentation, the most common symptom was epigastric discomfort, in a small group of patients was described as a first sign bleeding or anemia, one patient with a solitary Peutz-Jeghers polyp was presented with acute pancreatitis, and two patient were presented with acute intussusception of small bowel. Sometimes solitary

hamartomas were incidental findings during endoscopy examination, which was performed for another reason. Endoscopy has a distinct role in the diagnosis and treatment of solitary hamartomas. A solitary Peutz-Jeghers type hamartomatous polyp of duodenum is endoscopically characterized by a lobular or nodular surface. Abdominal CT scan is useful imaging modality. CT scan may help delineate the precise location of polyps. Endoscopic removal is ideal method of treating a polyp, when it is not possible, laparoscopy and bowel resection is effective alternative. Although solitary Peutz-Jeghers type hamartomatous polyps have been considered to have a lower risk of cancer, previous reports have shown cases of solitary Peutz-Jeghers type hamartomatous polyps with malignant components, and one case of solitary hamartomatous polyp had duplicated malignancy in six organs [8, 9].

Figure 1: The giant polyp in distal part of duodenum. (A) The pedunculated polyp with wide stem had an irregularly lobular surface, (B) We performed an endoscopic polypectomy with a snare piecewise.

Figure 2: Pathological findings of endoscopical polypectomy specimens. (A) Macro image: the fragmented polyp have an irregular, multilobulated appearance with a smooth surface, (B) The branching smooth muscle is derived from the muscularis mucosae, and the overlying mucosa is typically non-dysplastic and maintains its normal architecture (H&E stain, x400).

Figure 3: Computed tomography scan of abdomen showing the presence of fluid and soft tissue within the bowel lumen (white arrow), and the fat density suggested a small bowel intussusception.

CONCLUSION

We report one case of duodenal solitary Peutz-Jeghers type hamartomatous polyp, that obstructed small bowel. Polyp was completely removed endoscopically.

Duodenal solitary Peutz-Jeghers type hamartomatous polyps are rare, presumably distinct from Peutz-Jeghers syndrome. A solitary Peutz-Jeghers type hamartomatous polyp of the duodenum should be removed by endoscopic polypectomy or surgical resection and patients should undergo a colonoscopy and a whole-body screening to exclude other malignancies because of the malignancy potential.

Author Contributions

Alexandra Deduchova – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Petr Volsansky – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Filip Neumann – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Radim Zalud – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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