

## CASE REPORT

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# Massive Lower Gastrointestinal Bleeding from Meckel's Diverticulum with Heterotopic Pancreas: Case Report and a Brief Review of the Literature

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### ABSTRACT

**Context** Heterotopic pancreas is defined as pancreatic tissue found out of the pancreas proper with no vascular or anatomical connection. It is a rare clinical entity and can be found at any portion of the alimentary system. **Case report** We describe a case of Meckel's diverticulum with heterotopic pancreas who presented massive lower gastrointestinal bleeding. **Conclusion** Lower gastrointestinal bleeding due to Meckel's diverticulum with heterotopic pancreas is extremely difficult to make a definitive diagnosis preoperatively especially when it onsets rapidly.

### INTRODUCTION

Heterotopic pancreas is defined as pancreatic tissue outside its usual location, and without vascular or anatomical continuity of pancreas proper. Other terms such as ectopic pancreas, accessory pancreas, aberrant pancreas, and pancreatic heterotopia have also been described in the literatures. The incidence of heterotopic pancreas is at 0.5-13.0% in autopsy studies [1]. It is a rare clinical entity and can be found in any site of the alimentary system. Mostly it is reported in the stomach, duodenum and jejunum; other organs include esophagus, terminal ileum, omentum, mesentery, spleen, liver, Meckel's diverticulum, colon, ampulla of Vater, gallbladder, urinary bladder, lung and lymph nodes [2, 3]. We report a case of Meckel's diverticulum with heterotopic pancreas who presented massive lower gastrointestinal bleeding.

### CASE REPORT

A 23-year-old man was admitted to the First Affiliated Hospital of China Medical University with painless severe bloody stools for 6 hours accompanied by dizziness and asthenia. His medical and family records were unremarkable. He had no instance of nausea,

vomiting or fever. On the physical examination, there was no remarkable sign of pain or tenderness, and no mass could be palpated. Digital rectal examination was unremarkable. His hemoglobin level was 84 g/L (reference range: 120-160 g/L). Liver, renal functions, coagulation and platelet level were within normal range. The source of hemorrhage was not identified by upper gastroduodenoscopy. Bleeding required 8 units of blood infusion within 24 hours. <sup>99m</sup>Tc-technetium pertechnetate scintigraphy, capsule endoscopy and angiography were not suggested due to the high risk for the unstable hemodynamics.

An emergency laparotomy was performed immediately. Intraoperative colonoscopy did not identify the source of bleeding. A Meckel's diverticulum was found about 60 cm proximal to the ileocecal valve on the antimesentery side. Consequently diverticulectomy was carried out. Gross examination of the specimen revealed a 1.0 cm diameter mass supplied by varicose vessels arising from the distal end of the diverticulum (Figures 1 and 2). There was an ulcer on the mucosal surface of the diverticulum. On dissection the cut surface was yellowish. Histological examination revealed the presence of pancreatic acini, ducts and islets of Langerhans (Figures 3 and 4). The patient was discharged uneventfully. At his 8-month follow-up, there was no evidence of recurrence.

### DISCUSSION

Meckel's diverticulum is the vestigial remnant of yolk-duct which acts as communicating tract between the embryonic yolk sac and its primitive mid-gut in the first few weeks of fetal life. Meckel's diverticulum is

Received January 20<sup>th</sup>, 2013 – Accepted March 20<sup>th</sup>, 2013

**Keywords** Accessory pancreas [Supplementary Concept]; Choristoma; Hemorrhage; Meckel Diverticulum; Pancreas

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**Figure 1.** The mass (horizontal arrow) arised from the distal end of the diverticulum (vertical arrow).

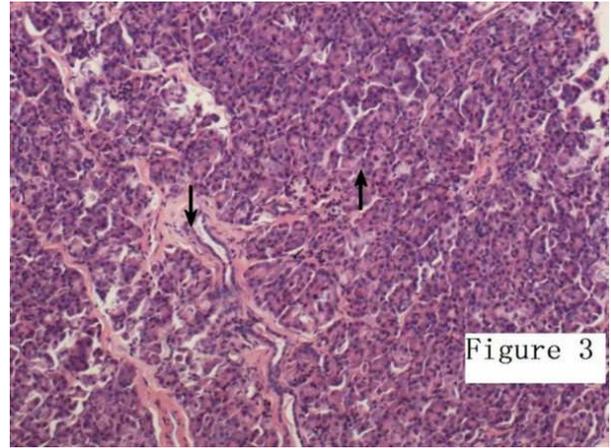
the most common anomaly results from incomplete obliteration of the yolk- duct [4]. Hemorrhage is reported in the majority of Meckel's diverticulum in children but unusual in adult [5]. It is usually painless, which can be slow or even massive, manifesting as anemia or fresh bloody stools. The mechanism of hemorrhage is the ulceration caused by the acid or enzymes secreted from the ectopic digestive mucosa [4].

The etiology of heterotopic pancreatic tissue is still unclear, but it is believed to arise embryonically during rotation of the foregut and fusion of the dorsal and ventral pancreatic buds. Some parts are separated from it and develop into heterotopic pancreas in any portion of the alimentary system. Another best known theory is based on the pancreatic metaplasia of endodermal tissue [6].

Although heterotopic pancreas can be found in many sites throughout the body, it is usually reported in the upper gastrointestinal tract. Bleeding arising from Meckel's diverticulum with heterotopic pancreas is extremely rare described in the literatures. In our review, a retrospective research was performed for articles in the past 30 years with the keywords: choristoma, heterotopic pancreas, ectopic pancreas,



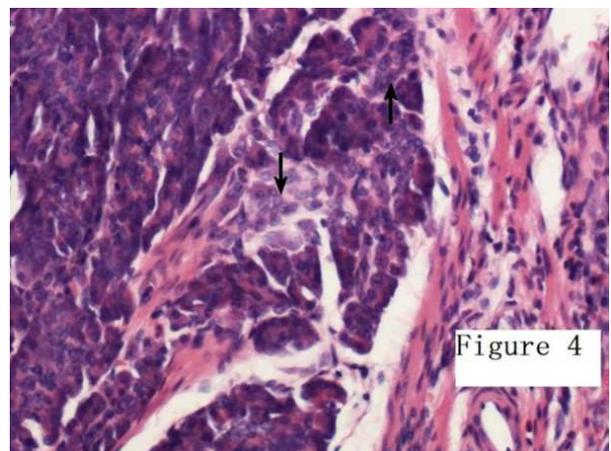
**Figure 2.** The mass was supplied by varicose vessel (arrow).



**Figure 3.** Histological section (H&E x100) revealed the presence of pancreatic acini (up arrow) and ducts (down arrow).

aberrant pancreas, pancreatic heterotopia, accessory pancreas, Meckel's diverticulum, hemorrhage and bleeding. Fifteen cases in English and three in Chinese were reported [7, 8, 9, 10, 11, 12, 13, 14, 15, 16, 17, 18, 19, 20, 21, 22, 23, 24]. Including our case, the study contained 11 males and 7 females (the gender of one case was not mentioned in one article [23]) between 2.5 to 73 years of age (mean equal to 33 years). Bloody stool was the main manifestation in 11 cases (57.9%), abdominal pain in 7 cases (36.8%), and one case was detected incidentally. All cases underwent laparotomy, and the diagnoses of heterotopic pancreas were confirmed based on the pathological examination. Heterotopic pancreas is usually asymptomatic unless the patient has intestinal obstruction, intussusception, inflammation or malignant change [4, 9]. The symptoms may be caused by the secretion of hormones and enzymes from the ectopic tissue. The symptoms may be caused by the inflammation, diverticulitis, ulceration and chemical irritation.

The typical endoscopic appearance is small well-circumscribed submucosal protrusion covered with normal mucosa, characterized as central umbilication which corresponds to the orifice of the duct. The characteristic endoscopic ultrasonographic features of



**Figure 4.** Histological section (H&E x200) showed the pancreatic acini (up arrow) and islet of Langerhans (down arrow).

ectopic pancreas include heterogeneous or mixed echogenicity, anechoic area and fourth-layer thickening which correspond to the presence of acini, duct and hypertrophy of the muscularis propria [25]. Biopsy from the surface often shows normal mucosa. The appearance of heterotopic pancreas on contrast computerized tomography is similar to pancreas proper [26]. The abdominal ultrasonography, X-ray and computerized tomography are not very specific. The routine gastroduodenoscopy and colonoscopy are limited for diagnosis of Meckel's diverticulum with heterotopic pancreas. <sup>99m</sup>Tc-technetate scintigraphy has a high diagnostic accuracy to evaluate gastrointestinal bleeding caused by ectopic gastric mucosa, but it cannot be performed in emergency for the unstable hemodynamics [27]. Angiography can establish a definitive location of gastrointestinal hemorrhage with an accuracy ranging from 33% to 68%, when the amount of hemorrhage is more than 0.5 mL/min [28].

Despite the development of modern diagnostic methods, the definitive preoperative diagnosis still remains challenging. The definitive diagnosis is confirmed by pathological examination after resection. The Heinrich criteria are used to classify heterotopic pancreas: type 1 contains cells of exocrine glands, excretory ducts and islets of Langerhans; type 2 contains only excretory glands and excretory ducts; Type 3 contains only excretory ducts [29].

The differential diagnoses of heterotopic pancreas include gastrointestinal stromal tumor, adenomyoma, leiomyoma, lymphoma, gastrointestinal autonomic nerve tumor and carcinoid tumor.

Surgical resection and pathological examination have been required for the diagnosis for symptomatic Meckel's diverticulum. A definitive diagnosis of Meckel's diverticulum with heterotopic pancreas is extremely difficult to make preoperatively. The indication of surgical intervention for the asymptomatic case is still controversial [5]. Pancreatic adenocarcinoma arising in a Meckel's diverticulum has been described by Koh *et al.* [9]. To prevent later complications, malignant change and re-operation difficulties, surgical intervention might be chosen.

Tumor, Meckel's diverticulum, angiopathy and heterotopic pancreas are the major causes of small intestinal hemorrhage. Lower gastrointestinal bleeding from Meckel's diverticulum as a rare clinical entity in adult patient should be considered as an important differential diagnosis especially in the condition of massive painless hemorrhage.

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**Conflict of interest** The authors have no potential conflict of interest

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