Case report

Asymptomatic pancreatic heterotopia in an incidental Meckel's diverticulum – A rare case report

Abena Hidangmayum[1], Kishan Prasad H. L.[1], Jayaprakash Shetty K.[1], Shreesha Khandige[2]

[1]Department of Pathology, K S Hegde Medical Academy, Deralakatte, Mangaluru, Karnataka, India
[2]Department of Pathology, GR Medical College, Mangaluru, Karnataka, India

(Received: April 2021 Revised: May 2022 Accepted: June 2022)

Corresponding author: **Abena Hidangmayum.** Email: abenasharma@gmail.com

ABSTRACT

Pancreatic heterotopia is a rare, unusual finding where pancreatic tissues are found out of the normal anatomical location. Various studies have documented an incidence of 0.5 to 13% in autopsy. The reports on pancreatic heterotopia presence is rare, with an incidence estimated to be around 0.2% of all upper gastrointestinal surgeries. The heterotopic pancreas occurs frequnetly in the stomach, duodenum and proximal jejunum. Remote cases of pancreatic heterotopia in Meckel's diverticulum is a rare finding. These heterotopias are often clinically silent and are an incidental findings upon pathological examination or autopsy.

Keywords: Pancreatic heterotopia; Meckel's diverticulum; appendicitis.

INTRODUCTION

ancreatic heterotopia are ectopic pancreatic tissue with no vascular or anatomical connection to the Ppancreas. Its Incidence is recorded to be 0.5 to 13% in the autopsy studies (1). In their descending order of incidence, the common site of pancreatic heterotopia is the stomach, duodenum, jejunum, and ileum. Other rare sites include colon, gall bladder, umbilicus, fallopian tube, mediastinum, spleen, liver and Meckel's diverticulum (2). It is seen more commonly in males than females in a ratio of 3:1 (3). Meckel's diverticulum is a rare entity, but the presence of pancreatic heterotopia in an incidental Meckel's diverticulum is a rarer entity. The incidence of Meckel's diverticulum in the general population is about 2%. Here we discuss a rare case of asymptomatic pancreatic heterotopia in an intraoperative incidental finding of Meckel's diverticulum in a 12-year-old female with acute appendicitis.

Case report

A 12-year-old girl presented with severe and abrupt right lower abdominal pain with fever for two days. Clinical examination revealed tenderness of the right iliac fossa, and a clinical diagnosis of acute appendicitis was made. Routine haematological investigation showed an elevated total count (11,400cells/cumm) with differential counts of neutrophil 78%, lymphocytes 20% and eosinophils 02%. Ultrasonography of the abdomen was consistent with acute appendicitis. The patient was treated with intravenous antibiotics and planned for interval appendicectomy. Intraoperatively, an inflamed appendix was noted along with an incidental finding of Meckel's diverticulum at a distance of 75cm from the ileum, which was resected. Both appendix and Meckel's diverticulum was sent for histopathological examination. The appendicectomy specimen was congested and measured 6.5cm in length. Fecolith was noted in the lumen of appendix. The resected Meckel's diverticulum specimen measured 4cm in length, noted along the anti-mesenteric border of a portion of ileum measuring 3 cm in length. A nodular, firm mass noted in the mucosal lumen of the diverticulum, measuring 1X 0.7cm. The diverticular wall was thinned out, and the mucosa of the ileum was unremarkable. Microscopy showed focal ulceration of appendiceal mucosa with transmural neutrophilic infiltration favouring acute appendicitis diagnosis (Fig.1A). The ileal mucosa was unremarkable (Fig.1B). The diverticulum showed ileal mucosa with the presence of heterotopic pancreatic tissue in lobules consisting of the acini, islet of Langerhans and intralobular duct separated by septae in the muscularis propria (Fig. C, D). A final diagnosis of acute appendicitis with type 1 pancreatic heterotopia in Meckel's diverticulum was made. Intraoperative and postoperative period were uneventful. The patient had a speedy recovery.

DISCUSSION

Meckel's diverticulum is the vestigial remnant of the yolk duct, that functions as a connecting tract between the yolk sac and its primitive mid-gut in the first few weeks of foetal life. It usually disappears by the seventh week of life. Of all congenital anomalies that arises from incomplete obliteration of the yolk- duct, Meckel's diverticulum is the most common. It is usually associated with the presence of ectopic tissue. The gastric mucosa is the most frequent heterotopia found in Meckel's diverticulum, with an incidence ranging from 60 to 82%, followed by pancreatic heterotopia with an incidence of 1 to 16% (4). Meckel's diverticulum is usually seen on the anti-

mesenteric side and comprises the four layers of the intestinal mucosa. Clinically it may be silent. However, it may be symptomatic due to complications associated with the presence of heterotopic tissues. These range from abdominal pain usually secondary to peptic ulceration in cases related to gastric heterotopia, intussusception, per rectal bleeding and intestinal perforation. The most predominat clinical presentation in a young child with symptomatic Meckel's diverticulum is a painless bleed from rectal. In older individuals, the frequent complication commonly observed is the intestinal obstruction. Recent studies reveal that heterotopia is noted in 43% of symptomatic adults and 59% of symptomatic paediatric cases of Meckel's diverticulum (5).

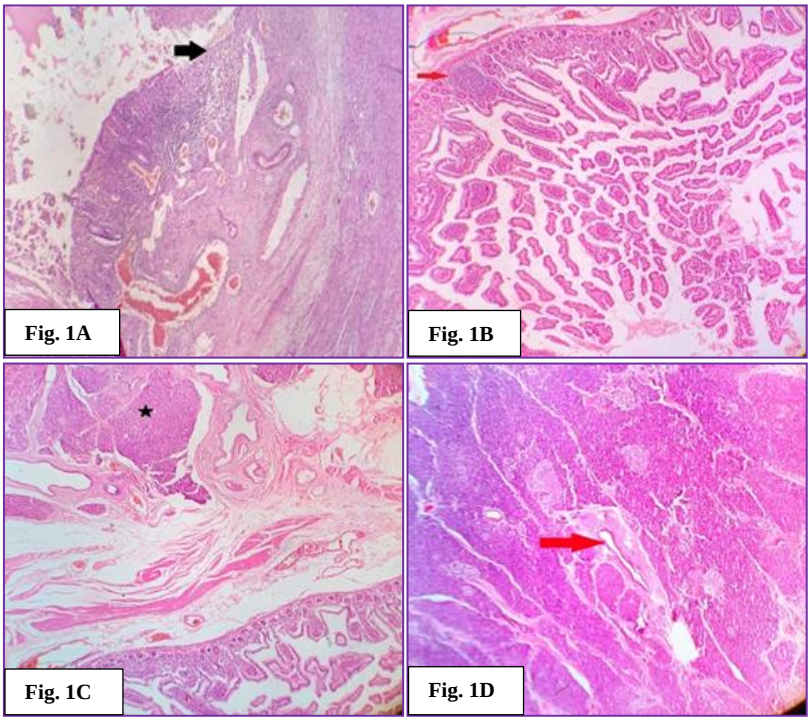

Fig. 1A: Acute appendicitis with focal ulcerated appendiceal mucosa (black arrow) and neutrophilic infiltration; **1B**: Normal ileal mucosa with peyer patches (red arrow); **1C**: Meckel's diverticulum bearing an ileal mucosa with pancreatic lobule (black star); **1D**: Pancreatic lobule with a pancreatic duct (red arrow), acini and islet of Langerhans (H and E,10X)

Heterotopic tissues are a rare clinical entity seen in the gastrointestinal tract. The aetiology of pancreatic heterotopia is still inconclusive. Due to embryological defect during rotation of foregut, it is thought to be the amalgation of dorsal and ventral pancreatic buds. Some tissue separate from the pancreas and develops into pancreatic heterotopia in any site/segment of the alimentary system. Another hypothesis is the pancreatic metaplasia of endodermal tissue (6). As the embryonic pancreatic tissue is close to the foregut during development, the most common pancreatic heterotopia site is in the upper gastrointestinal tract, the stomach being the most frequent site. Other rare sites include gall bladder, jejunum, ileum, colon, mediastinum,liver, spleen,fallopian tube, umbilicus,, and Meckel's diverticulum. The Heinrich criteria classifies heterotypic pancreas into three major types: Type 1 consisting of exocrine glands,islets of Langerhans and the excretory ducts; Type 2 consists of both excretory glands and excretory ducts; Type 3 is formed exclusively by the excretory ducts (7).

Clinically significant pancreatic heterotopias are considered to be larger than 1.5cm in size, beyond which clinical symptoms appear (8). Symptoms may develop as a sequel to pancreatic juice secretion presenting as acute pancreatitis. The clinical significance of heterotopic pancreas hinge on size, site and its associated complications. The presentation may ranges from abdominal pain, anaemia, melaena and ,sometimes,weight loss. Major cases of pancreatic heterotopia form a polypoidal or mass-like lesions in the gastrointestinal tract subsequently leading to clinical symptoms by their mass effects. As a result, it may manifest clinically as intestinal obstruction and intussusception. Heterotopic pancreas in the gastrointestinal tract may also mimic gastrointestinal stromal tumour (GIST), leiomyoma, lymphoma, and carcinoid tumour. Radiologically pancreatic heterotopia is difficult to distinguish from GIST and metastatic tumours, and hence the use of endosonography may be helpful. Definitive diagnosis is yielded with the histopathological examinationof the

resected specimen. Surgical excision is the only definite treatment as medical management isn't effective (9).

In our case, type 1 pancreatic heterotopia was seen in Meckel's diverticulum, one of the rare sites of pancreatic heterotopia. The pancreatic heterotopic nodule size was 1x0.7 cm, which was borderline, and the patient was asymptomatic. Despite modern diagnostic development, a confirmatory preoperative diagnosis of heterotopia in Meckel's diverticulum is still challenging. In difficult cases, laparoscopy is a more endorsed diagnostic modality. However, technetium-99m per-technate scan is the most precise non-invasive investigation performed in such difficult cases (10). Surgical resection and pathological examination are confirmatory for the diagnosis of symptomatic Meckel's diverticulum. In our case, preoperative investigations didn't yield results regarding the existence of Meckel's diverticulum or pancreatic heterotopia. Surgical resection was done, and the histopathological examination gave the final diagnosis.

CONCLUSION

Asymptomatic Meckel's diverticulum is usually a subordinate finding intraoperatively. The presence of heterotopic pancreas in Meckel's diverticulum can be clinically silent. Surgical excision is the only treatment taking into the weightage of its complications and risk of transformation to malignancy. The diagnosis is confirmed only on histopathological examination.

CONFLICT OF INTEREST

Authors declare that there is no conflict of interest.

REFERENCES

1. Sandhyarani, M., Patil, M. R. Meckel's diverticulum – a case study. Int J Anat Var. 2013; 6: 66-67.
2. Song, D. E., Kwon. Y., Kim, K. R., Kim, J. S. Adenocarcinoma arising in gastric heterotopic pancreas: a case report. J Korean Med Sci. 2004;19(1):145-148.
3. Tanaka, K., Tsunoda, T., Eto, T., Yamada, M., Matsuo, S., Izawa, K. Diagnosis and management of heterotopic pancreas. Int Surg. 1993;78:32-35.
4. Riccardo, G., Valeria, B., Giulia, C., Alessia, C., Luisa, F., Elizabetta, T., *et al.,* Heterotopic pancreas in Meckel's diverticulum in a 7-year old child intussusception and recurrent gastrointestinal bleeding: case report and literature review focusing on diagnostic controversies. Afr J Paediatr Surg. 2014;11(4):354-358.
5. Quarrie, R., Lindsey, D., Bahner, D. P. Review of the incidence and management of Meckel's diverticulum. Austin J. Surg. 2014;1(3):1-3.
6. Okamoto, H., Fujishima, F., Ishida, K., Tsuchida, K., Shimizu, T., Goto, H., *et al.,* Intraductal papillary mucinous neoplasm originating from a jejunal heterotopic pancreas: report of a case. Surg Today. 2014;44(2):349-353.
7. Trifan, A., Târcoveanu, E., Danciu, M., Huţanaşu, C., Cojocariu, C., Stanciu, C. Gastric heterotopic pancreas: an unusual case and review of the literature. J Gastrointestin Liver Dis 2012;21(2):209-212.
8. Bromberg, S.H., Neto, C.C., Fernando, A., Borger, A., Franco, M.I.F., Franca, L.C.M., *et al.,* Pancreatic heterotopias: clinicopathological analysis of 18 patients. Rev Col Bras Cir. 2010;37(6):413-419.
9. Kilius, A., Samalavicius, N. E., Danys, D., Zaldokas, G., Seinin, D. Asymptomatic heterotopic pancreas in Meckel's diverticulum: a case report and review of the literature. Journal of Medical Case Reports. 2015; 9(1): 108.
10. Shalaby, R.Y., Soliman, S.M., Fawy, M., Samaha, A. Laparoscopic management of Meckel's diverticulum in children. J Pediatr Surg 2005;40: 562-567.