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Ectopic Pancreas with Manifested Intestinal Obstruction in a Newborn - A Case Report and Literature Review

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Abstract

Ectopic pancreas is not an uncommon congenital anomaly manifesting the heterotopic pancreas tissue separated from the normal pancreas. The main ectopic site is the proximal gastrointestinal tract. The majority of ectopic pancreas is asymptomatic, whereas some subjects cause diseases such as bleeding and luminal obstruction. The distal bowel obstruction caused by ectopic pancreas is rarely reported. Herein, we reported a newborn presented with distal bowel obstruction complicating bowel gangrene 31 h after delivery. Surgical resection cured this disease.

Keywords: Ectopic pancreas; Intestinal obstruction; Neonate

Introduction

Ectopic Pancreas (EP) is pancreatic tissue which is not anatomically connected to the primary pancreas. It is commonly found in the upper gastro intestinal tract and the Meckel's diverticulum. EP has also been reported in other locations such as the gallbladder, the biliary tree, the mesentery and even the fallopian tubes [1]. Almost all patients are asymptomatic, although some rare complications such as bleeding, intussusception, and ectopic pancreatitis have been observed [2]. The prevalence of ectopic pancreas ranges from 0.55% to 13.7% in autopsy series [1]. Symptomatic ectopic pancreas has rarely been reported in neonates according to our literature review. Here we present a rare case with intraluminal bowel obstruction, caused by ectopic pancreas in the ileum after birth.

Case Presentation

A full-term female newborn that had undergone uneventful prenatal examinations was transferred to our department at 31-h old after experiencing bilious vomiting. A physical examination upon arrival revealed mild distention of her abdomen which was otherwise soft without tenderness. Hypoactive bowel sounds were recorded. She had passed meconium within 24 h after birth with the laboratory data showing unremarkable findings. There was continuous bilious drainage from the nasogastric tube. A KUB X-ray revealed dilated small bowel loops compatible with mechanical ileus (Figure 1). An emergent abdominal CT scan showed no evidence of volvulus. A follow-up KUB taken 24 h later showed persistent ileus. A pediatric surgeon was consulted and an exploratory laparotomy was performed to investigate further the unknown etiology of mechanical obstruction.

The operation revealed an intraluminal mass measuring 1.2 cm occupying most of the lumen in the terminal ileum located about 10 cm from the ileocecal valve. The adjacent sections of the small bowel exhibited poor perfusion. The mass lesion and ischemic bowel (25 cm in length) were resected, and primary small bowel anastomosis was performed. The pathology report a lobulated mass composed of variable-sized ducts, which is compatible with ectopic pancreas (Figure 2). After surgery, the hospital course was uneventful, and the patient exhibited normal bowel function during a follow-up examination one year later.

Discussion

Mechanical intestinal obstruction is a common surgical emergency occurring during the neonatal period in approximately 1 in 1,500 live births [3]. There are many causes of intestinal obstruction such as atresia, stenosis, internal hernia, and intestinal malrotation with or without volvulus [3]. Early diagnosis and appropriate management are important for both prevention of intestinal gangrene and resection.

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Table 1: Symptomatic ectopic pancreas in neonates in there viewed literature.

Author	Year	Sex	Age	Symptoms	Location	Treatment
Visentin et al.	1991	M	18 days	Vomiting	Pylorus	Biopsy and watchful waiting
Würfel. and Riebel	1992	M	23 days	Vomiting	Omentum near pylorus	Tumor excision
Hayes-Jordan et al.	1998	M	2 days	Vomiting	Antrum	Tumor excision
Kawashima et al.	1998	M	7 days	Projectile vomiting	Pylorus	Tumor excision (with duodenal duplication)
Özcan et al.	2002	M	1 month	Vomiting, failure to thrive	Pylorus	operation
Fragoso et al.	2003	M	21 days	Post-prandial vomiting	Pre-pyloric	Tumor excision
Sharma et al.	2004	M	1 month	Vomiting	antrum	Tumor excision
LeeWT et al.	2005	M	8 days	Protrusive mass with discharge	Umbilicus	Tumor excision
Shiratori et al.	2008	F	4 days	Bowel perforation	Jejunum and ileum	Operation
Saka et al.	2009	M	6 days	Bilious vomiting	Jejunum	Tumor excision
KimS-H et al.	2014	M	20 days	Vomiting	Pylorus	Excision
KimH-S et al.	2021	F	1 day	Bilious vomiting	Jejunum	Tumor excision
Presented case	2021	F	1 day	Bilious vomiting	Ileum	Tumor and ischemic bowel excision

M: Male; F: Female

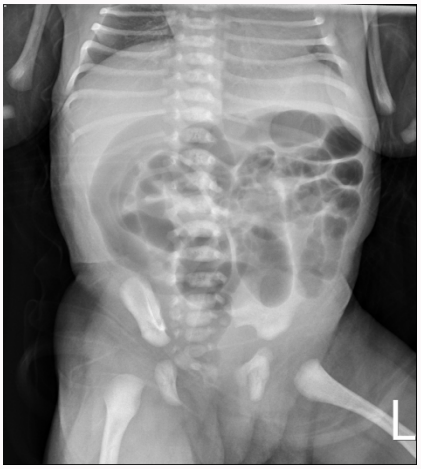

Figure 1: Abdominal radiography showed multiple dilated small bowel loops during the visit.

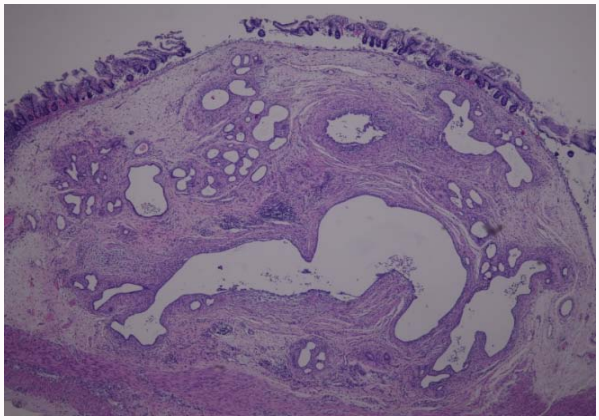

Figure 2: The pathology of surgical specimen showed a lobulated mass composed of variable-sized ducts, which is histologically compatible with ectopic pancreas.

Ectopic Pancreas (EP) is defined as the abnormal location of pancreatic tissue with ductal and vascular systems that do not communicate with the main body of the pancreas. Its prevalence is approximately 0.54% in children, according to a recent retrospective study carried out in a single center [1]. Two prevailing pathogenetic mechanisms have been proposed to explain the development of ectopic pancreatic tissue. One is that fragments of pancreatic tissues separate from the developing pancreas during the rotation of the foregut, and the other is that the pancreatic metaplasia forms from multipotent stem cells [1]. The Gaspar-Fuentes modified classification identifies four types of ectopic pancreas [4]: Type I is typical pancreatic tissue, with ducts, acinar cells and islet cells, and is similar to the normal pancreas; Type II (the canalicular variety) contains pancreatic ducts only; Type III (the exocrine pancreas) contains acinar tissue only; and Type IV (the endocrine pancreas) contains islet cells only. CT scans have shown that acinus-dominant lesions exhibited an enhancement similar or greater to that of the orthotopic pancreas, whereas duct-dominant lesions were shown to be hypovascular with fewer enhancements [5]. Pathology findings classified our case as type II EP and the lesion was undetectable on the abdomen CT scan.

EP is found at any age, with most cases being asymptomatic and incidental findings. EP is most commonly located in the stomach and the duodenum, followed by a Meckel's diverticulum [1]. EP has been identified in children presenting with following symptoms: Bleeding, umbilical discharge, vomiting, recurrent abdominal pain, and intussusception [6]. The children with symptomatic EP were older than those whose EP was an incidental finding [1]. There have also rare cases of malignant transformations of ectopic pancreatic tissue [7]. The most common approach to managing symptomatic EP is surgical excision.

Only a few cases of symptomatic EP have been reported during the neonatal period (Table 1). Most of these cases affected male babies (83.3%), and most of them were located near the pylorus (66.7%), causing non-bilious vomiting. Other symptoms caused by EP were one case with an umbilical mass with discharge and three cases (25% of all cases) with an obstruction of the small intestine. One particular case was a preterm baby with 34 weeks of gestation who presented with a bowel perforation at 4-day-old, and was found to have a lot of ectopic pancreatic tissue EP on the anti-mesenteric side of the jejunum and ileum [8]. Although proximal bowel obstruction is the most common symptom found in neonates according to our literature review, our case demonstrates an extremely rare instance of neonatal EP with distal bowel obstruction at the level of the ileum

complicated by bowel ischemia. Based on our findings, pediatricians should take the possible presence of EP into consideration when faced with neonates presenting with mechanical bowel obstruction.

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