Meckel diverticulum with ectopic pancreatic tissue in a 12-year-old boy with intestinal invagination. Case report and review of the literature

Divertículo de Meckel con tejido pancreático ectópico en un niño de 12 años con invaginación intestinal. Reporte de caso y revisión de la literatura

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Abstract

Introduction: Meckel’s diverticulum is a remnant of the omphalomesenteric duct and its prevalence is 2%. Clinical case: A 12-year-old male with abdominal pain, accompanied by muco-sanguineous evacuations and hematemesis, with right lower quadrant mass. In the laparotomy, invasion was found; intussusception reduction and resection of the Meckel’s diverticulum was performed.

Conclusion: The association of Meckel’s diverticulum with heterotopic pancreatic tissue in children is rare, its main manifestation is intussusception and ultrasound is an option for diagnosis.

KEY WORDS: Meckel diverticulum. Ectopic pancreatic tissue. Intussusception.

Resumen

Introducción: El divertículo de Meckel es un remanente del conducto onfalomesentérico y su prevalencia es del 2%. Caso clínico: Niño de 12 años con dolor abdominal, acompañado de evacuaciones mucosanguíneas y hematemesis, con masa en cuadrante inferior derecho. En la laparotomía quirúrgica se encontró invaginación, y se realizó desinvaginación y resección del divertículo de Meckel. Conclusión: La asociación de divertículo de Meckel con tejido pancreático heterotópico en niños es poco frecuente. Clínicamente, su principal manifestación es la invaginación intestinal en la literatura revisada, y el ultrasonido constituye el mejor estudio para el diagnóstico de invaginación intestinal, no así para el divertículo de Meckel o el tejido pancreático heterotópico, en los que la tomografía sería una opción.

PALABRAS CLAVE: Divertículo de Meckel. Tejido pancreático ectópico. Invaginación intestinal.

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Introduction

Meckel’s diverticulum is a remnant of the omphalo-mesenteric duct, located on the antimesenteric border of the ileum, at 40-60 cm from the ileocecal valve, and it is characterized for containing all the layers normally found in the ileum. The prevalence of Meckel’s diverticulum is approximately 2% in the general population; of total cases, 76% are in adults and 24% in pediatric patients.

In 62% of cases heterotopic gastric mucosa can be found, in 6%, pancreatic tissue, in 5%, gastric and pancreatic mucosa, in 2%, jejunal mucosa and in 2% Brunner’s tissue. Clinical presentation can include intestinal bleeding, pain, intestinal occlusion or intussusception. It can be diagnosed with technetium-99 scintigraphy when there is ectopic gastric mucosa, but computerized tomography is the method that has been shown to be the most specific, showing a tubular structure filled with fluid or intestinal intussusception images.

The purpose of this paper is to present a case of Meckel’s diverticulum with heterotopic pancreatic tissue in a 12-year-old boy with intestinal intussusception, as well as to perform a review of the literature, since this is a rare pathology and we did not find cases reported in the national literature.

Clinical case

Twelve-year-old boy with a history of laparoscopic appendectomy at 10 years of age. He attended the emergency department with an 8-hour history of abdominal pain, located at the mesogastrium with 10/10 intensity, accompanied by muco-sanguineous stools in seven occasions, which subsequently were melenaic. There were episodes of vomiting on eight occasions, with gastro-alimentary content and then with blood. On physical examination, blood pressure of 129/48 mmHg, mean blood pressure of 73 mmHg, heart rate at 100 beats per minute, respiratory rate of 28 breaths per minute, temperature of 36 °C, 86% saturation and weight of 51 kg were observed. Dehydrated, conscious, oriented, pale oral mucosa, lung fields with air adequately flowing in and out. Precordium with heart sounds increased in intensity, without murmurs. Soft, deppressive abdomen. With a diagnosis of digestive tract bleeding of etiology to be determined, management with omeprazole, acetaminophen and ondansetron was administered for 48 hours, with no improvement. Laboratory tests reported hemoglobin 15.00 g/dL, platelets 193,000, white blood cells 10,600 x 10^3 μL, neutrophils 89.70%, lymphocytes 9.10%, monocytes 1%, basophils 0.20%, creatinine 0.82, glucose 128 mg/dL, sodium 139 mEq/L, potassium 4.1 mEq/L, calcium 9.40 mg/dL and C-reactive protein (CRP) 0.793 mg/dL. A pan-endoscopy did not reveal active bleeding in the stomach or esophagus. Since on abdominal palpation a mass was felt in the lower right quadrant, an ultrasound was carried out, which reported a fluid-free image with a “bull’s eye” appearance, suggestive of invagination. The intussusception head was displayed at the hepatic angle, with a pediculate, rounded image dependent on a handle of the small intestine being identified at the center (Fig. 1).

With the diagnosis of intestinal intussusception with more than 48 hours of evolution, and considering the patient age, performing a surgical examination was decided, where double invagination was found: ileo-ileal and ileocolic intussusception (Fig. 2), with the first ileoileal segment harboring a Meckel diverticulum inverted on its axis and this segment doubly invaginated to the ascending colon. Desinvagination by taxis was carried out, together with intestinal resection of the Meckel diverticulum located at 40 cm of the ileocecal valve (Fig. 3), with 5 cm of intestine at each side and end-to-end anastomosis. Postoperative evolution was favorable, and the patient was discharged without complications, and at the moment of the report, he was in good health conditions.

On histopathological examination, macroscopically, the intussuscepted Meckel diverticulum was observed with its three layers and, at the center, solid-looking, light yellow tissue, reminiscent of pancreatic tissue (Fig. 4). On histological section, pancreatic acini and slightly dilated ducts could be appreciated; there were no Langerhans islets (Fig. 5).

Discussion

Heterotopic pancreas is defined as pancreatic tissue without a true anatomical or vascular connection with the pancreas, and was first reported by Jean Schultz in 1729. Other terms used are ectopic pancreas, accessory pancreas and aberrant pancreas. In 1859, Zenker reported the first case of ectopic pancreas in a Meckel diverticulum.

Heterotopic pancreas origin is still unclear, but it is believed to arise embryonically during rotation of the foregut and fusion of the dorsal and ventral pancreatic tissue, where some tissue separates from the
pancreas and develops in any portion of the intestine. Another best known theory is based on the pancreatic metaplasia of endodermal tissue. Due to the proximity of embryonic pancreatic primordial buds and the foregut during development, 70-90% of ectopic pancreas occurs in the upper portion of the digestive tract.

Most patients with Meckel’s diverticulum are asymptomatic and the diagnosis is usually made by scintigraphy with technetium-99, tomography, endoscopy or during surgical examination motivated by other diseases. In children, most cases are detected due to the presence of intestinal intussusception. Heterotopic pancreatic tissue visualization is difficult, and we only found one report where heterotopic pancreatic tissue could be identified in a Meckel’s diverticulum as a homogeneous, hypoechoic and well-defined mass on ultrasound.

According to Heinrich’s criteria, heterotopic pancreas is classified as type 1 if it contains cells of exocrine glands, excretory ducts and islets of Langerhans, type 2 if it contains excretory glands and excretory ducts, and as type 3 if there are only excretory ducts. The most common is type 2, followed by type 1, while type 3 is uncommon. Our case corresponded to type 2.

In our literature review, we were able to document 10 cases reported in children from 23 days to 15 years of age, 60% in children older than 2 years and 70%
in males (Table 1). Around 50% had a presentation with intussusception, 30% with Meckel’s diverticulum and, in two cases, exploratory laparotomy was performed without an accurate diagnosis being established, and was based only on the presence of abdominal pain, vomiting or abdominal distension. In five cases, the type was reported according to Heinrich’s histological classification, with types 1 and 2 being the most common5,6,15-20.

We consider that heterotopic pancreatic tissue favors the presence of intestinal intussusception, and the literature also refers that if a patient develops acute pancreatitis in the “mother” organ, inflammation will also affect heterotopic foci, causing abdominal pain with elevation of pancreatic enzyme blood levels, which did not occur in our patient14.

Conclusions

Association of Meckel’s diverticulum with heterotopic pancreatic tissue in children is rare. Clinically, its main manifestation is intestinal intussusception in the reviewed literature. Ultrasound is the best study for diagnosis of intestinal intussusception, but not so for Meckel’s diverticulum and heterotopic pancreatic tissue, where tomography would be an option.

References


Table 1. Documented cases of children from 23 days to 15 years of age with Meckel’s diverticulum and heterotopic pancreatic tissue

<table>
<thead>
<tr>
<th>Author, year</th>
<th>Age</th>
<th>Gender</th>
<th>Presentation</th>
<th>Histopathological classification</th>
</tr>
</thead>
<tbody>
<tr>
<td>Guanà et al.6, 2014</td>
<td>7 years</td>
<td>Female</td>
<td>Intussusception</td>
<td>Type 1</td>
</tr>
<tr>
<td>Baysoy et al.15, 2010</td>
<td>5 years</td>
<td>Male</td>
<td>Intussusception</td>
<td>Unknown</td>
</tr>
<tr>
<td>Xiao et al.5, 2009</td>
<td>12 years</td>
<td>Male</td>
<td>Intussusception</td>
<td>Type 2</td>
</tr>
<tr>
<td>Huml et al.16, 2009</td>
<td>1 month</td>
<td>Male</td>
<td>Pain, nausea and vomiting</td>
<td>Type 1</td>
</tr>
<tr>
<td>Ogata et al.17, 2007</td>
<td>1 year</td>
<td>Female</td>
<td>Meckel’s diverticulum, intestinal malrotation</td>
<td>Type 1</td>
</tr>
<tr>
<td></td>
<td>4 years</td>
<td>Female</td>
<td>Intussusception</td>
<td>Unknown</td>
</tr>
<tr>
<td></td>
<td>10 years</td>
<td>Male</td>
<td>Meckel’s diverticulum</td>
<td>Type 2</td>
</tr>
<tr>
<td></td>
<td>23 days</td>
<td>Male</td>
<td>Meckel’s diverticulum, bile duct atresia</td>
<td>Unknown</td>
</tr>
<tr>
<td>Ogata et al.17, 2007</td>
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<td>Unknown</td>
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Figure 4. On the antimesenteric border, intussuscepted Meckel’s diverticulum with its three layers is observed, and on the center, solid-looking, light yellow tissue, reminiscent of pancreatic tissue.

Figure 5. Histological section with pancreatic acini and slightly dilated ducts. There are no islets of Langerhans.


