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Original

Pancreatic coristoma within a duodenal diverticulum: a case report

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Abstract

Pancreatic choristoma, also called heterotopic pancreas is a rare condition in gastroenterology. This entity consists of the presence of aberrant pancreatic tissue in some other area of the gastrointestinal tract without vascular or anatomical continuity with the normal pancreas; it has been seldomly reported and could exhibit variable symptomatology.

We herein report the case of a 46-year-old male who presented with abdominal pain and was found, to have a pancreatic choristoma within a duodenal diverticulum through invasive examinations. The diagnosis was made using histopathology and immune-histochemistry testing.

Keywords: *Choristoma, heterotopic pancreas, duodenal diverticulum, gastrointestinal tract, abdominal pain (source: MeSH NLM).*

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Pancreatic choristoma or also called heterotopic pancreas (HP) is a rare pathology in gastroenterology.[1] It consists of the presence of aberrant pancreatic tissue in an area of the gastrointestinal tract where it should not be found, also without vascular or anatomical continuity with the organ itself, this is due to embryogenesis of the tissue in that area without a specific cause.[2-4]

There are few reports of this condition worldwide.[5] It usually has no symptoms during childhood, and in adulthood they are variable; there have been reported cases of patients who debut with upper gastrointestinal bleeding, and others whose only sign is chronic abdominal pain.[3, 6]

The definitive diagnosis is histopathologic.[2, 5, 7] Regarding treatment, surgery remains the best option to avoid the malignant evolution of the aberrant tissue.[8]

We present the case of a patient with chronic abdominal pain secondary to ectopic pancreatic tissue within a duodenal diverticulum.

Clinical case

Male, 46 years old, from Ica, with no pathologic or surgical history. The disease started six months before his admission with epigastric pain associated with a five

kilograms weight loss during those months. He consults Gastroenterology because, during the last week, he has had premature fullness and episodes of emesis of biliary content, of moderate quantity, in addition to the previous symptoms.

Physical examination revealed superficial and deep palpation pain in the epigastrium, with no other findings. An upper gastrointestinal endoscopy was performed, showing a tumor lesion with central depression on the anterior wall of the duodenal bulb, approximately 3 cm in diameter, which was biopsied. A complete abdominal CT scan with contrast was performed showing the same saccular image in the bulb, which was then diagnosed as a duodenal diverticulum (Figure 1).

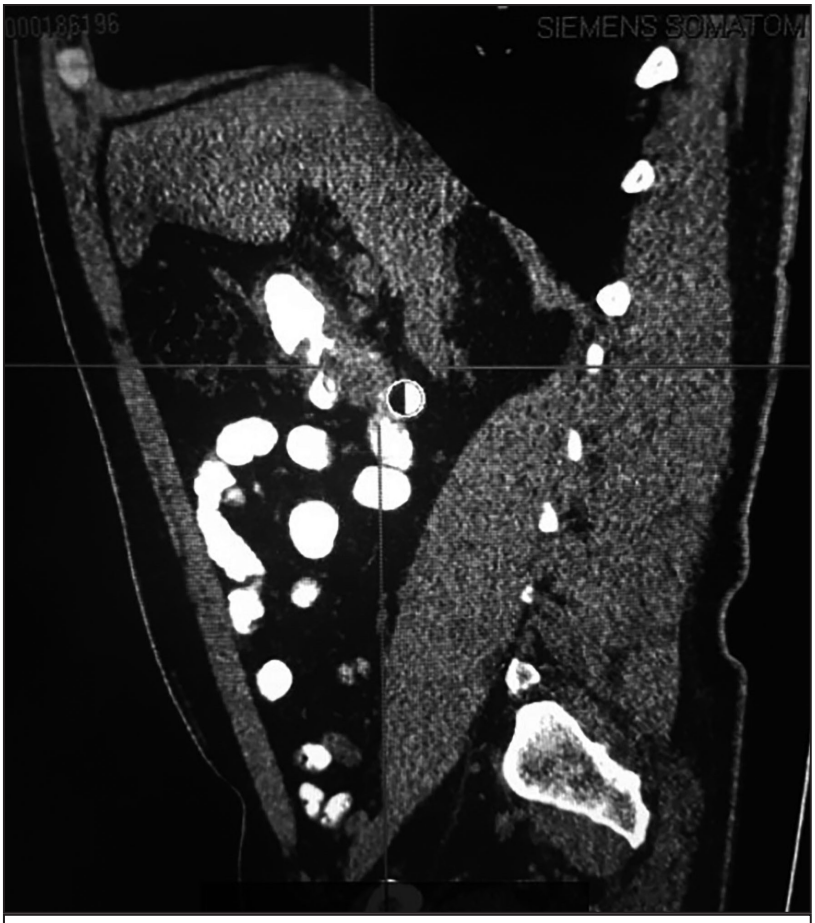
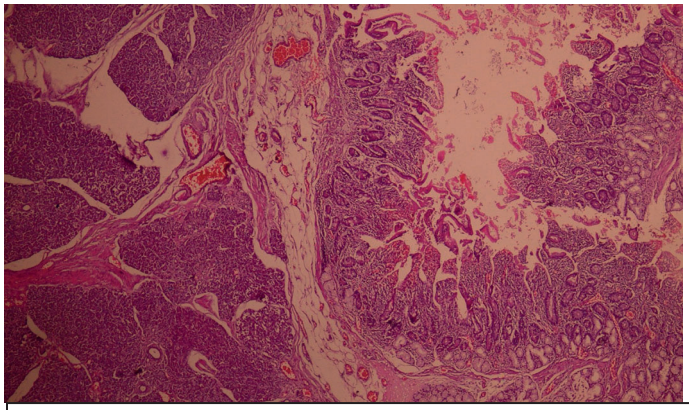


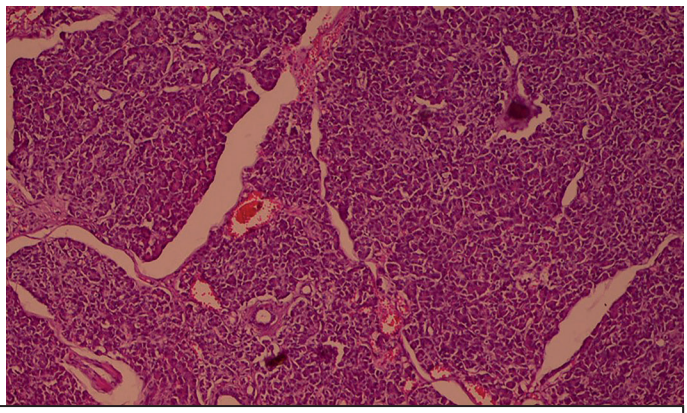
Figure 1. Enterotem with contrast in the anterior face of the duodenal bulb shows a saccular image approximately 2 cm in diameter (ARROW).

An exploratory laparotomy was performed to define the histology of the lesion. A 3.5 cm x 2.5 cm wide-based, hyper vascularized, and the edematous saccular tumor was found, originating in the inferior face of the pylorus, with projection to the gastric area. Diverticulectomy was performed followed by Finney pyloroplasty.

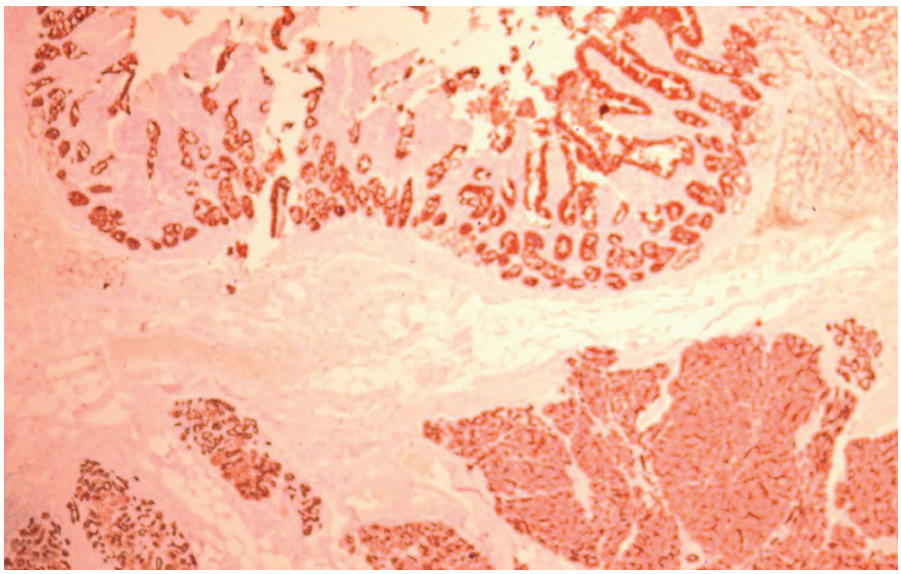
The histopathological study showed in the muscularis propria numerous ducts grouped in lobules and separated from each other by thick and irregular bundles of smooth muscle and dense connective tissue; also, the presence of islets of Langerhans compatible with pyloric pancreatic choristoma (Figures 2 and 3)., The tissue was positive for CK AE1/AE3 (cytokeratins for the identification of keratins and basal) by immunohistochemistry, indicating the presence of two epithelia, both duodenal and pancreatic in the sample (Figure 4); while the chromogranin and synaptophysin studies were negative a fact that ruled out neuroendocrine tumors (Figures 5 and 6). This led to the final diagnosis of a complete type I pancreatic heterotopia. The patient evolved favorably from the procedure and to date is stable.

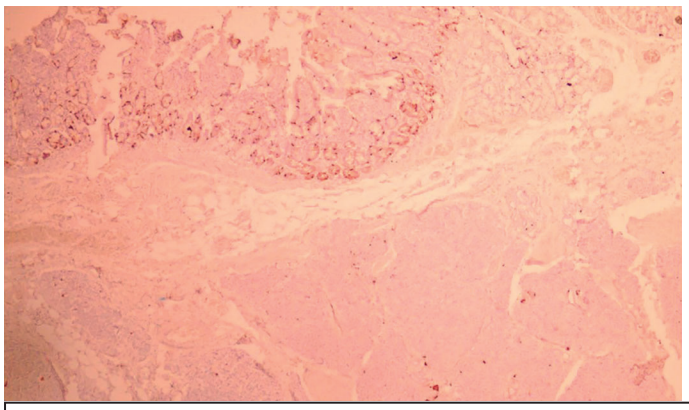
We have the patient's informed consent for the preparation and publication of this article.

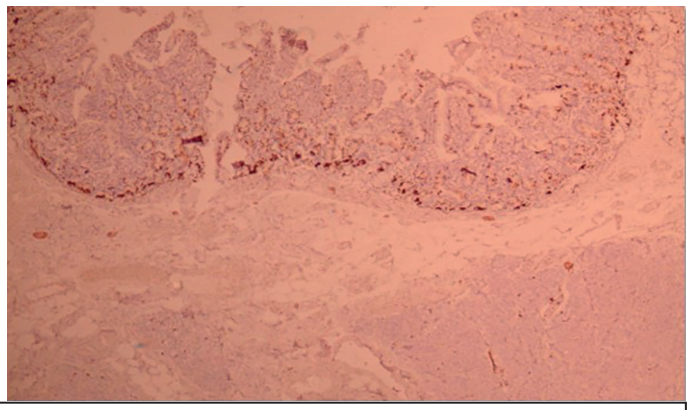




Figures 2 and 3. In the muscularis propria, numerous ducts grouped in lobules and separated by thick and irregular bundles of smooth muscle and dense connective tissue are observed; in addition, there are some islets of Langerhans. The histopathological picture is compatible with pyloric pancreatic choristoma.







Figures 4, 5, and 6. In the immunohistochemical study, CK AE1/AE3 was positive. While in the immunohistochemical study of chromogranin.

Discussion

Choristomas are common in the gastrointestinal tract and are found incidentally during imaging examinations such as CT scans and invasive examinations such as endoscopy.[9] Heterotopic tissue can occur at any location along the gastrointestinal tract, with the most frequent sites being the stomach and small intestine.[1] The aberrant or heterotopic pancreas is an isolated mass of pancreatic tissue, completely separated from the main organ, where its embryogenesis occurs, but its mode of development is unknown.[3, 4] Approximately 5% of the world's population suffers from this condition and the symptomatology does not occur in childhood, but between 40 and 60 years of age; as in the case of our patient.[5]

Pancreatic ectopia is rarely reported in the literature, while the signs and symptoms depend on the organ affected, with the stomach (30%), duodenum (25%), jejunum (16%), ileum (2.8%), and rarely esophagus being affected, with about 20 cases reported in the literature. [10-13] Among the cases of aberrant pancreatic tissue found in the duodenum, the case reports mention a preponderance of localization in the second portion of the duodenum.[14, 15]

The definitive diagnosis is by biopsy, which must be done to rule out malignancy or to define specific inflammatory causes.[16] Sometimes there are alterations in the underlying tissues such as inflammation, fat necrosis, and diverticulum formation.[7]

In 1973, Gaspar Fuentes and collaborators, based on the old criteria of von Heinrich, proposed a new four types classification[2], shown below, based on the structures of the pancreas found in this heterotopic tissue:

type I (atypical pancreas (complete heterotopia)), which corresponds to our case.

type II, only with pancreatic ducts (canalicular heterotopia),

type III, with acinar cells only (exocrine heterotopia) and

type IV, only with islets of Langerhans (endocrine heterotopia).

On the other hand, complications of this disease include pancreatitis, intussusception formation, intestinal obstruction, gastrointestinal bleeding, malignant degeneration, and pseudocysts. [17]

Regarding treatment, surgical resection of the aberrant tissue remains the best option[3]. We are aware of the possibility of malignant degeneration that this ectopic tissue may undergo and that is why it is suggested to preventively monitor the patient periodically after the surgical procedure, to detect possible recurrences, although this is rare and little reported.[8, 9]

Conclusions

Despite its rarity, pancreatic choristoma should be considered within the possibilities of abdominal mass or chronic pain in adult patients.

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