ELSEVIER

Contents lists available at ScienceDirect

### **International Journal of Surgery Case Reports**

journal homepage: www.elsevier.com/locate/ijscr



#### Case report

## Gastric perforation and generalized peritonitis due to heterotopic pancreas, an unusual case report $^*$

María N. Gómez <sup>a,b,\*</sup>, Aurora García Gómez <sup>a</sup>, Irma Sánchez Montes <sup>a</sup>, Jesús Villagran Uribe <sup>a</sup>, María Fernanda Torres Ruiz <sup>c</sup>, María E. Baridó Murguía <sup>d</sup>

- a Department of General Surgery, General Hospital "Dr. Miguel Silva", Morelia, Michoacán, Mexico
- <sup>b</sup> Street Pirul 309, Fray Antonio de San Miguel Iglesias, CP 58270 Morelia, Michoacán, Mexico
- <sup>c</sup> Advanced endoscopy and Endoscopic Surgery, National Institute of Respiratory Diseases, Mexico City, Mexico
- <sup>d</sup> Department of Surgery, Medical South, Private, Mexico City, Mexico

#### ARTICLE INFO

# Keywords: Mucinous cystadenoma Bilateral salpingo-oophorectomy Total abdominal hysterectomy Borderline Histopathological

#### ABSTRACT

*Introduction:* Heterotopic pancreas is the presence of normal pancreatic tissue that is in an anatomical site different from the pancreas.

Case report: Adolescent who was admitted to the emergency room due to intense, generalized abdominal pain, which did not go away with anything, was exacerbated with movements, 24 h later vomiting that occurred as soon as he ate food was added, and severe and progressive abdominal distention, reason for admission to the service of emergencies. He had no significant family or personal medical history.

On physical examination, he was found to be sick with a painful appearance, pale, sweaty, feverish, with tachycardia, he complained of intense pain, distended abdomen, with loss of hepatic dullness, with involuntary muscular resistance, with positive rebound sign, absent peristalsis. Laboratory studies were taken that reported anemia, leukocytosis and neutrophilia, blood chemistry, electrolytes, and coagulation times within normal limits; abdominal x-rays showed subdiaphragmatic free air.

Discussion: The prevalence of pancreatic heterotopia in the literature varies from 0.6 % in autopsy series to 14 %, however, the increasingly frequent use of upper gastrointestinal endoscopy and endoscopic ultrasound allows early diagnosis, since whether directed or incidentally, which avoids serious complications such as gastric perforation, because it is unusual for it to occur as in this clinical case. Specifically in the stomach, involvement of the submucosa layer has been described in 73 %, muscular layer in 17 % and subserosa in 10 % of cases respectively. In cases where the disease is in an asymptomatic stage, it can be treated conservatively; it seems to be more common in men, as in our case, than in women.

Conclusion: In patients with chronic epigastric pain, it is essential to rule out conditions such as heterotopic pancreas, to avoid serious complications such as perforation.

#### 1. Introduction

The presence of normal pancreatic tissue that is in an anatomical site different from the pancreas, and that does not maintain vascular, neuronal or anatomical continuity with said gland, is known as a heterotopic pancreas or ectopic pancreas [1].

It has all the following histological characteristics: formation of pancreatic acini, development of ducts and islets of Langerhans. It was described for the first time in 1727, in a diverticulum of the ileum, by

Jean-Schultz; It has been reported most frequently in the stomach in 24 to 38 % of cases [2], in the duodenum in 9 to 36 % [3], and the jejunum in 0.5 to 27 % [4], as well as in other sites. Meckel's diverticulum, abdominal esophagus, bile ducts, liver, spleen, mesentery, adrenal gland, etc. Let us remember that the pancreas has its origin in two primitive buds, the dorsal bud and the ventral bud, which arise respectively from the duodenum and the base of the liver, and occurs in the fifth week of gestation; During the seventh week of gestation, the two buds fuse: the ventral part gives rise to the head of the pancreas and

 $<sup>^{\</sup>star}$  All authors declare that we did not obtain any type of financing for the preparation of this work.

<sup>\*</sup> Corresponding author at: Department of General Surgery, General Hospital "Dr. Miguel Silva", Morelia, Michoacán, Mexico.

E-mail addresses: normagomezherrera@yahoo.com.mx, marianorma1306@gmail.com (M.N. Gómez), aurora.garcia5655@alumnos.udg.mx (A.G. Gómez).

the uncinate process, while the dorsal part gives rise to the body and tail of the pancreas.

The cause of heterotopic pancreas is unknown; It is possible that early in fetal life, during rotation of the foregut and fusion of the dorsal and ventral parts of the pancreas, small parts are separated from it, and continue to develop in the wrong location [5] or caused by metaplasia of totipotent endodermal cells [6].

We present the case of an adolescent with generalized peritonitis due to gastric perforation secondary to a heterotopic pancreas in the lesser curvature of the stomach, a very unusual complication in this disease, who remained for 2 years with epigastric pain, with remissions and exacerbations, he received several treatments due to the diagnosis of

gastritis and even gastroenteritis in his area, he went to the emergency room with us until he developed an acute abdomen. This case report has been reported in line with the SCARE Criteria [7].

#### 2. Case report

16-year-old patient, with a history of chronic abdominal pain for 2 years, "burning" type, in the epigastrium, intense that subsided with medical treatment based on analgesics, pump inhibitors and sometimes antibiotics, due to apparent gastroenteritis, 3 days before his admission, the pain appeared again in the epigastrium and later became generalized throughout the abdomen, it did not go away with anything, it was

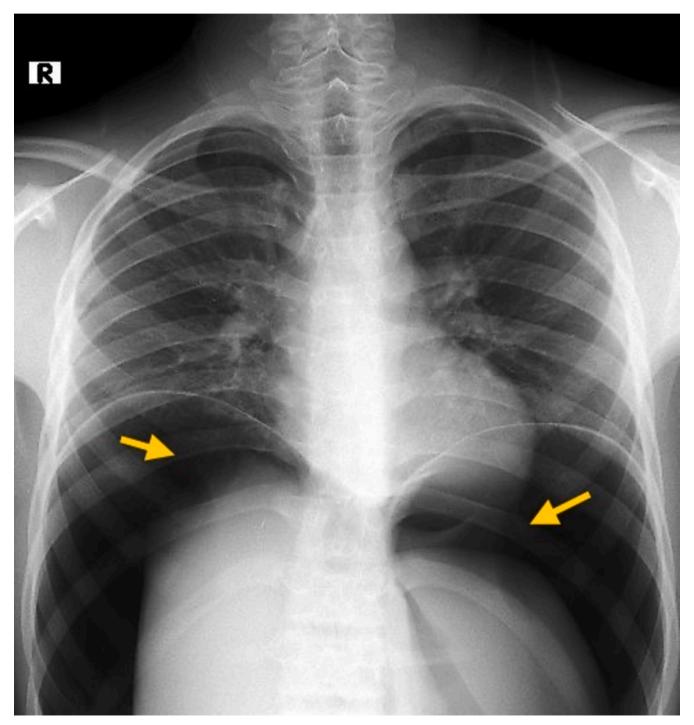


Fig. 1. Chest teleradiography showing large amounts of subdiaphragmatic free air.

exacerbated with movements, 24 h later vomiting was added that occurred as soon as he ate food, vomiting what he ate, followed by severe and progressive abdominal distention, reason for admission to the emergency department. He had no significant family or personal medical history, the parents and grandparents were apparently healthy, he had all the corresponding vaccinations for his age.

On physical examination, he was found to be ill with a painful appearance, pale, sweaty, febrile, with tachycardia of 118 per minute, polypnea of 24 per minute, temperature of 37.8 degrees Celsius, he complained of intense pain, ballooning abdomen, with loss of hepatic dullness, with involuntary muscular resistance, with positive rebound, absent peristalsis, laboratory studies were taken that reported anemia with hemoglobin 11 g/dL, leukocytosis of 14 thousand, and neutrophilia, blood chemistry, electrolytes and coagulation times within limits normal

A diagnosis of acute abdomen was made due to probable perforation of a hollow viscus. Simple abdominal x-rays were taken, standing and recumbent, which showed free air and abundant free fluid throughout the abdomen (Fig. 1); No additional study was performed, it was rushed to the operating room, to exploratory laparotomy, the following findings were found: segment of hardened gastric wall with tumor appearance in a dimension of  $6\times 6$  cm, with an ulcerated area and perforation in the middle end. of the lesser curvature of the stomach, approximately 3 cm, with gastric contents throughout the abdomen, the abdominal cavity was washed with abundant physiological solution until it was clean, a wide wedge gastric resection was performed, which included the entire

area of the stomach. Tumor with a margin of 4 cm of apparently healthy tissue, and the stomach was closed in two planes, an omentum patch was placed, 2 drains were placed, one subhepatic and another in the right parietocolic groove, the abdominal cavity was closed in planes with continuous stitches in peritoneum and separate stitches in aponeurosis and skin; sample was sent to pathology; no intraoperative complications, but with a severe systemic inflammatory response, he was admitted to the intensive care unit due to difficulty in being extubated, which occurred 24 h postoperatively, however, nosocomial pneumonia was added, which was resolved in 10 days and The patient was discharged due to improvement, on day 14 of his hospital stay; The histopathological report (Fig. 2) of the surgical specimen was: Heterotopic pancreas measuring  $4 \times 3$  cm, was found 5 cm from the proximal surgical edge and 3 cm from the distal edge, located at the lower end of the lesser curvature of the stomach, which included the entire thickness of the gastric wall, foci of acute and chronic nonspecific pancreatitis; in addition to nonspecific chronic gastritis with complete intestinal metaplasia without atypia; lymphoreticular hyperplasia in 9 of 9 lymph nodes of the lesser gastric curvature.

He was seen in the outpatient clinic with a 6-month follow-up and a year after his treatment, a telephone consultation was carried out where he stated that he felt well, so he was discharged.

#### 3. Discussion

At the hospital, only patients 18 years of age and older are treated;

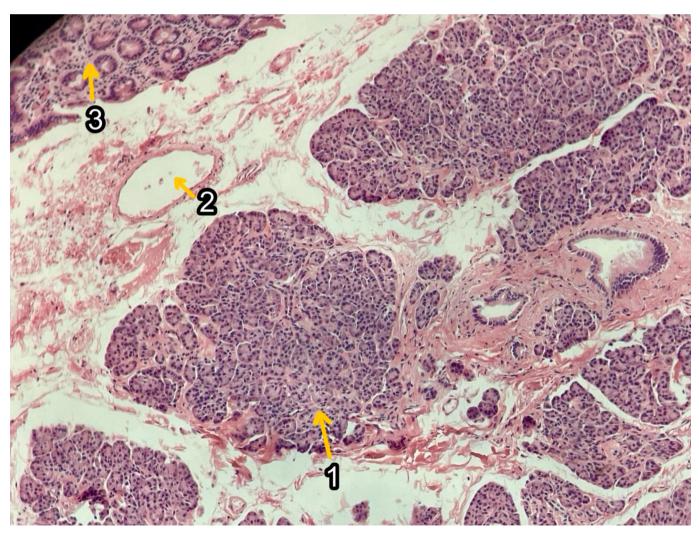


Fig. 2. Histological section of stomach segment at the perforation site.

adolescents are occasionally seen, so I believe we have little experience with the disease in this population; this exemplifies the diagnostic difficulty of the presence of heterotopic pancreas in the stomach, where until a serious complication such as perforation occurred, it also shows the reason why no other study was performed on this patient, no abdominal tomography was performed, due to the need to perform emergency surgery and not delay treatment, except for simple x-rays of the abdomen, where a large amount of subdiaphragmatic free air was observed, a consequence of the perforation of the stomach, which the patient was urgently transferred to the operating room. He never had esophagogastroduodenoscopy with endoscopic ultrasound performed, which was indicated according to what the literature mentions, with which this nosological entity called heterotopic pancreas would have been identified. Before causing an abdominal catastrophe that could cause death, however in this case the diagnosis was made late, in the piece surgery that was sent to pathology, which was the resected stomach segment; In general, this disease is evident until some complication occurs, such as: inflammation, bleeding, obstruction or malignant transformation. The difficulty of nosological diagnosis, in patients who only manifest few symptoms, as in this case: pain in the epigastrium, has already been described in several manuscripts by other authors [8,9,10] and they even describe it as the only constant symptom; and they agree that identifying this ectopic tissue becomes a real challenge [11]. We recommend that patients with these characteristics also think about this possibility and rule out the presence of heterotopic pancreas, although the most common congenital alteration is pancreas divisum. When the biopsy is taken from the correct site, histopathologicall, it is not a diagnostic problem when pancreatic acini, ducts, islets of Langerhans and intervening connective tissue are present; There are cases in which the biopsy was so complex that it was necessary to repeat it several times to diagnose the disease [12]. The most characteristic gross feature is a central ductal orifice [13] the way in which the endoscopic appearance is usually a small, centrally umbilicated, submucosal mass and the surface biopsies are usually normal because of the frequent submucosal localization of the lesion [14,15], so taking the biopsy is decisive. Specifically in the stomach, involvement of the submucosa layer has been described in 73 %, muscular layer in 17 % and subserosa in 10 % of cases respectively [16], in this case presented it was found in the least frequent, up to the serosa. This patient even received treatment with antibiotics for gastric symptoms and there was no evidence that this treatment was necessary. We once again urge you to prescribe antibiotics judiciously and only when they are strictly indicated. At the moment, antibiotics are no longer sold without a prescription like in other countries.

The heterotopic pancreas in this patient caused gastric perforation, and foci of acute and chronic pancreatitis were identified, and due to the history of his symptoms, it is probable that he had several recurrences, which could have caused it was considered that necrotizing inflammation in the heterotopic pancreas. Tissue resulted in ulceration and then perforation, as described in 2009, described as perforation sealed by omentum [17] and which is the only case reported in the last 20 years in the adolescent and adult population. In the patient we are reporting, the perforation was of dimensions older and did not seal, which caused generalized peritonitis; Let us remember that in this entity all the diseases suffered by the normotopic pancreas can also occur, because it has a genetic structure, function and local environmental exposure similar to that of the pancreas; then it can present from acute pancreatitis and pseudocysts [15] could result from retention of exocrine secretions in the absence of a communication between the glandular epithelium and the gastric lumen resulting in obstruction of pancreatic secretion, phenomena of recurrent acute pancreatitis [1], elevation of pancreatic enzymes at the site where the tissue is found. Like to intestinal intussusception [18,19] and even malignant diseases, which are derived from any pancreatic cell.

We emphasize the importance of carefully studying the symptoms and signs that the patient presents, which is most frequently abdominal pain, alone or with vague associated symptoms, since this is what allows us to reach a correct diagnosis, without underestimating the importance of the histological studies that give us the definitive diagnosis, endoscopic, laboratory and cabinet studies.

#### 4. Conclusion

The majority of patients with heterotic pancreas are asymptomatic or present non-specific symptoms, it is necessary to rule out this diagnosis in patients with chronic epigastric pain, and it is essential to avoid serious complications such as perforation, diffuse peritonitis and even added nosocomial infections, like this case, early esophagogastroduodenoscopy, with endoscopic ultrasound it is indicated in these patients, when taking the biopsy the most frequent location of this pathology, which occurs in the submucosa, must be considered.

#### Consent

The patient's mother signed the consent and accepted the disclosure of her son's clinical case for scientific purposes, as well as respect for her privacy.

#### Ethical approval

Reviewed the case as well as the informed consent and allowed us to publish it.

#### **Funding**

All authors declare that we did not obtain any type of financing for the preparation of this work.

#### **Author contribution**

All authors participated in the concept of this report, as well as the treatment of the patient, as well as in the writing of this manuscript.

#### Guarantor

María Norma Gómez Herrera. PhD.

#### Research registration number

None.

#### **Conflict of interest statement**

All authors declare that we did not have any conflict of interest.

#### References

- K.C. Mulholland, W.D. Wallace, E. Epanomeritakis, S.R. Hall, Pseudocyst formation in gastric ectopic pancreas, JOP 5 (2004) 498–501.
- [2] C. Ozcan, A. Celik, C. Guclu, E. Balik, A rare cause of gastric outlet obstruction in the newborn: pyloric ectopic pancreas, J. Pediatr. Surg. 37 (2002) 119–120 (PMID 11782002).
- [3] M. Kaneda, T. Yano, T. Yamamoto, T. Suzuki, K. Fujimori, H. Itoh, et al., Ectopic pancreas in the stomach presenting as an inflammatory abdominal mass, Am. J. Gastroenterol. 84 (1989) 663–666 (PMID 2729238).
- [4] R.F. Thoeni, R.K. Gedgaudas, Ectopic pancreas: usual and unusual features, Gastrointest. Radiol. 5 (1980) 37–42 (PMID 6965644).
- [5] S. Kobayashi, Y. Okayama, K. Hayashi, et al., Heterotopic pancreas in the stomach wich caused obstructive stenosis in the duodenum, Intern. Med. 45 (2006) 1137–1141.
- [6] R.V. Dolan, W.H. ReMine, M.B. Dockerty, The fate of heterotopic pancreatic tissue. A study of 212 cases, Arch. Surg. Chic. Ill 1960 109 (6) (1974) 762–765.
- [7] C. Sohrabi, G. Mathew, M. Nicola, A. Kerwan, T. Franchi, Riaz A. Agha, The SCARE 2023 guideline: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg. 109 (5) (May 2023) 1136–1140, https://doi.org/10.1097/JS9.000000000000373.

- [8] C De Ponthaud, E Daire, M Pioche, B Napoléon, M Fillon, A Sauvanet. Cystic Dystrophy in Heterotopic Pancreas.
- [9] G.H. Sakorafas, M.G. Sarr, Ectopic gastric submucosal pancreatic tissue, JOP. J. Pancreas (Online) 4 (2003) 214–215 (PMID 14614202).
- [10] O.T. Ormarsson, I. Gudmundsdottir, R. Mårvik, Diagnosis and treatment of gastric heterotopic pancreas, World J. Surg. 30 (9) (Sep 2006) 1682–1689, https://doi. org/10.1007/s00268-005-0669-6.
- [11] S.A. Sathyanarayana, G.B. Deutsch, J. Bajaj, B. Friedman, R. Bansal, E. Molmenti, J.M. Nicastro, G.F. Coppa, Ectopic pancreas: a diagnostic dilemma, Int. J. Angiol. 21 (3) (Sep 2012) 177–180, https://doi.org/10.1055/s-0032-1,325,119.
- [12] Zhang, Xiaohan, Peng, Lihua, Wang, Zikai, Extensive heterotopic pancreas in a rare site: a case report and a review of literature, Medicine 102 (9) (March 03, 2023) e32241, https://doi.org/10.1097/MD.000000000032241.
- [13] Anca Trifan, Eugen Târcoveanu, Mihai Danciu, Cătălin Huţanaşu, Camelia Cojocariu, Carol Stanciu, Gastric heterotopic pancreas: an unusual case and review of the literature, J. Gastrointestin. Liver Dis. 21 (2) (June 2012) 209–212.

- [14] C.Y. Hsia, C.W. Wu, W.Y. Lui, Heterotopic pancreas: a difficult diagnosis, J. Clin. Gastroenterol. 28 (1999) 144–147.
- [15] K. Yamashita, K. Yamazaki, A. Ueno, Y. Arimura, T. Endo, K. Imai, Image of the month. A gastric heterotopic pancreas with cystic change, Gastroenterology 129 (5) (2005 Nov) 1374, https://doi.org/10.1053/j.gastro.2005.01.062, 1809.
- [16] J.R. DeBord, J.D. Majarakis, L.M. Nyhus, An unusual case of heterotopic pancreas of the stomach, Am. J. Surg. 141 (1981) 269–273.
- [17] B. Gurocak, H.S. Gokturk, S. Kayacetin, S. Bakdik, A rare case of heterotopic pancreas in the stomach which caused closed perforation, J. Med. 67 (7) (julyaugust 2009) 285–287.
- [18] K. Ratan, M. Singh, B. Rani, Tina. Heterotopic pancreas leading to ileo-ileal intussusception, APSP J. Case Rep. 3 (2) (2012 May) 12. Epub 2012 Jun 1.
- [19] S.H. Asfaw, N. Kundu, X. Liu, H. Abdel-Aziz, M.A. Samotowka, M.M. Loor, Heterotopic pancreas as a lead point for ileoileal intussusception, Am. Surg. 78 (3) (Mar 2012) E187–E189.