Annular pancreas as a rare cause of gastric outlet obstruction in a 16-year-old male patient: A case report

Bonephace Tresphory¹, Agness Mlawa², Gofrey Giiti³, Andrea Mombo⁴, Giacom Antonini³, Ph³illipo L. Chalya¹³

- ^{1, 2, 5} Department of General Surgery, Ikonda hospital, Njombe, Tanzania
- ³ Department of General Surgery, Catholic University of Health and Allied sciences
- ⁴ Department of Urology, Ikonda hospital, Njombe, Tanzania

Abstract

Annular pancreas (AP) is a rare cause of congenital gastric outlet obstruction that is usually discovered during the neonatal period. Still, clinical severities can vary over a wide range and definite diagnosis could be delayed until late childhood or adulthood. We report here a case of AP detected in a 16-year-old malnourished male patient who was admitted because of non-bilious vomiting and epigastric fullness after intake of food which was relieved after vomiting. A contrast-enhanced CT scan of the abdomen confirmed the diagnosis of AP. At operation, a complete obstruction of the second part of the duodenum was found, caused by an annular pancreas. No other congenital anomaly of the intra-abdominal organs was noted. He successfully underwent retro-colic gastro-jejunostomy with uneventful postoperative recovery. Though a rare finding, AP should be considered as a differential diagnosis in patients presenting with gastric outlet obstruction after excluding common causes. The rarity of this congenital abnormality and its successful correction by surgical means have prompted us to make the following presentation.

Keywords: Annular pancreas, duodenal obstruction, surgery

Introduction

Annular pancreas (AP) is a rare congenital anomaly that causes congenital duodenal obstruction in the neonatal period (Patra *et al.*, 2011). It is characterized by a complete or incomplete ring of pancreatic tissue surrounding the second portion of the duodenum (Patra *et al.*, 2011; Alahmadi & Almuhammadi., 2014; Singh *et al.*, 2016). Several theories have been proposed but the etiology is still unclear. It is thought to originate from failure of the ventral pancreatic bud to rotate with the duodenum, resulting in encirclement of the duodenum (Leeco., 1910; Baldwin., 1910; Dowsett *et al.*, 1989; Shirkhoda., 2000).

AP is usually discovered at the neonatal period. However, the degree of duodenal obstruction and the subsequent obstructive symptoms might be variable, and unrecognized AP has been detected in adolescents or even in adults in some cases (Patra *et al.*, 2011; Alahmadi & Almuhammadi., 2014; Singh *et al.*, 2016). AP is exceptionally rare in the adult population with incidences varying from 0.005% to 0.015%, and presentation is variable and can mimic a wide range of clinical entities like peptic ulcer, pancreatitisand, more rarely, obstructive jaundice, thereby making the diagnosis difficult (Zyromskiet *al.*, 2008; Edman *et al.*, 2019; de la Rosa Rodriguez *et al.*, 2019; Nur *et al.*, 2022).

¹Corresponding authorl: <u>plchalya65@gmail.com</u>

Many imaging studies such as X-ray, upper GI series, abdominal ultrasound, abdominal CT scan, ERCP, MRCP, endoscopic ultrasound can be used to diagnose AP. However, in adults x-rays, upper GI series and abdominal ultrasound may be inconclusive. ERCP is effective but invasiveand can precipitate/exacerbate pancreatitis (Singh *et al.*, 2016). The diagnosis is currently based on abdominal CT scan and MRCP of the pancreasSingh *et al.*, 2016). Surgical bypass leads to fast recovery whenever pancreatic encirclement is associated with duodenal obstruction (Nur *et al.*, 2022). In this report, we present a rare case of a 16-year old male patient who presented with obstructive symptoms of duodenal obstruction due to annular pancreas and successfully treated with retro-colic gastro-jejunostomy.

Case report

A 16-year-old male patient presented to our hospital with a recurrent history of non-bilious vomiting since early childhoodworsening in the past 1 year. The vomitus contained food particles that were taken few minutes back. He also complained of epigastric pain and fullness after intake of food which was relieved after vomiting. He reported to have significant weight loss without loss of appetite. He had been treated symptomatically with the clinical diagnosis of chronic gastroenteritis for his recurrent vomiting.

On physical examination, he was found to have the classical signs of malnutrition: Low body weight for age (32kg), prominent bones, depleted fat and muscle, dry and inelastic skin. He was severely wasted, moderately pale but not icteric. His abdomen was soft, scaphoid withmildly palpable distended stomach at the epigastrium. He was found to have positive succession splash. No masses or hernias were discovered.

Routine biochemical tests revealed hypokalemic hypochloremic metabolic alkalosis, and haematological tests were within normal ranges. Plain abdominal X-Ray and Ultrasound findings were inconclusive for the diagnosis. The arterial phase of an axial multiple detector spiral computed tomography (MDCT) with intravenous and oral contrast only showed a massively dilated stomach full of contrast and a duodenum partially wrapped by the head of the pancreas with the so-called "crocodile jaw" appearance (Figure 1).

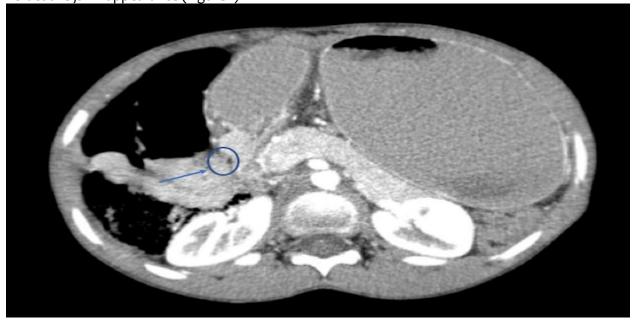


Figure 1: Abdominal computed tomography shows an abrupt narrowing of the duodenal bulb and a massively dilated stomach

Tanzania Journal of Health Research Volume 25, Number 1, January 2024

A laparotomy with the presumption of an obstructive AP confirmed a thick peri-duodenal band of pancreatic tissue, which required to be bypassed by a retro-colic gastro-jejunostomy. No other congenital anomaly of the intra abdominal organs was noted. Oral feeding started on the fourth postoperative day. The recovery was uneventful, and the symptoms disappeared 2 weeks after the operation. He was discharged the 14th postoperative day after intensive nutritional rehabilitation. At the one-year follow-up, the patient was asymptomatic and his body weight was 45 Kg.

Discussion

Since it was first reported by Tieddmann (1818) and named as annular pancreas by Ecker (1862), only few cases of AP have been reported in literature. AP is a rare congenital abnormalitycharacterized by a complete or incomplete ring ofpancreatic tissue surrounding the second part of the duodenum (Patra et al., 2011; Alahmadi & Almuhammadi., 2014; Singh et al., 2016). Several theories have been proposed to explain the development of annular pancreas. Leeco's and Baldwin's theories are the most acceptable among all. Leeco postulated that the tip of the ventral bud fuses abnormally to the duodenal wall and rotates incorrectly around the duodenum and resulting in a band of fibrous or pancreatic parenchymal tissue around the second part of the duodenum (Leeco., 1910; Baldwin., 1910). Baldwin (1910) reported that this condition arose because of the abnormal movement of the ventral pancreatic bud. The third theory explained by Verga (1972) suggests that the primary abnormality is duodenal with the pancreas "filling the space" around a narrowed duodenum. This results in a complete or incomplete stenosis of the duodenal lumen (Shirkhoda., 2000).

Clinical presentation can vary significantly, from asymptomatic cases to complete duodenal obstruction. The age at presentation depends upon the severity of duodenal obstruction (Patra *et al.*, 2011; Alahmadi & Almuhammadi., 2014; Singh *et al.*, 2016). It has been estimated that only about 33% of the cases are symptomatic. 50% of patients present in the pediatric age group, 86% of these present in the neonatal period. AP is exceptionally rare inadults and commonly diagnosed during the investigation of symptoms arising due to its complications(Zyromski et al., 2008; Edman *et al.*, 2019; de la Rosa Rodriguez *et al.*, 2019; Nur *et al.*, 2022). In adults, APusually presents between age 20 and 50 and is most commonly associated with abdominal pain and gastric outlet obstruction, secondary to duodenal stenosis (Edman *et al.*, 2019). Obstructive symptoms presenting in adults may be due to repeated inflammation, edema leading to fibrosis and scarring (Zyromski et al., 2008; Edman et al., 2019). Sandrasegran *et. al.*(2009) in a study of 40 cases of annular pancreas in adult population revealed that majority of cases were asymptomatic with only 5% cases presenting with gastric outlet obstruction /pancreatitis.

A dual-phase clinical manifestation of AP in the same patient, combining partial duodenal obstruction and abdominal pain and vomiting due to chronic pancreatitis at adult age (Cai et al., 2018), as occurred in the patient of the present report, is most unusual. As reported in our patients, vomiting in patients with duodenal obstruction secondary to AP is usually non-bilious in 90% of these cases, mimicking a pyloric obstruction, as the pancreatic encirclement is frequently located proximal to the ampulla of Vater (Patra et al., 2011; Singh et al., 2016). Delay seeking medical advice contributes to long-lasting unrelieved symptoms in adolescents or adults. It was the case with our patient. Late referral to medical attention was due to cultural, economic and logistic factors common to many rural areas where access to specialist care is still problematic.

The diagnosis of AP can be suggested through imaging tests such as X-ray, upper GI series, abdominal CT scan, ERCP, MRCP, endoscopic ultrasound, but the definitive diagnosis is surgery which is considered diagnostic gold standard (Singh *et al.*, 2016). The diagnosis of AP beyond neonatal age requires a high index of suspicion. In adults, ultrasonography, plain abdominal x-ray and upper GI series

may be inconclusive as reflected in our patient. Jadvar and Mindelzun(1999) showed that contrast-enhanced abdominal CT is useful in directly visualizing the completeor partial AP tissue in adults, and Sandrasegaran *et al* (2009) suggested an abdominal CT finding of a crocodile jaw configuration of pancreatic tissue surrounding the second part of the duodenum, which was suggestive of thepresence of AP in adults. This observation is reflected in our patient in whom the definite diagnosis was confirmed by a contrast-enhanced abdominal CT scan.

The treatment of choice in patients with symptomatic AP is surgery. The optimal operationhas been a matter of debate. The goal of surgery is torelieve duodenal or gastric outlet obstruction by abypass surgery of the annulus, which can be achieved viaaduodenoduodenostomy, gastrojejunostomy, or aduodenojejunostomy (Cai et al., 2018). Resection of the annulus is contraindicated since it is associated with serious complications such as pancreatitis, pancreatic fistula formation, andpancreatic insufficiency leading to unacceptablyhigh morbidity(Zyromskiet al., 2008; Cai et al., 2018; Edman et al., 2019; de la Rosa Rodriguez et al., 2019; Nur et al., 2022). The results of previously reported operations are satisfactory, especially the gastrojejunostomy duodenoduodenostomy, which are simple operations performed frequently and have the best results (Nur et al., 2022). In this study, our patient successfully underwent retrocolic gastrojejunostomy with uneventful postoperative recovery. At one year follow up, the patient had gained weight and was doing well.

Conclusion

In this study, we successfully diagnosed and treated an AP in a 16-yearold male, who suffered from a long standing history of duodenal obstruction without definite diagnosis. The definite diagnosis was confirmed by a contrast-enhanced abdominal CT scan and successfully underwent retrocolic gastrojejunostomy with uneventful postoperative recovery. An annular pancreas, although rare, should be considered as differential diagnosis in patient with unresolved symptoms of gastric outlet obstruction after excluding common causes.

Ethical considerations

The permission to conduct this case study was obtained from Ikonda Hospital authority (Hospital Management Team) before the commencement of the study. Informed consent was sought from the parents of the patient.

References

Alahmadi, R., Almuhammadi, S. (2014) Annular pancreas: a cause of gastric outlet obstruction in a 20-year-old patient. *American Journal of Case Reports* 15, 437–440.

Baldwin, W.M. (1910) A specimen of annular pancreas. Anatomical Records4, 299-304.

Cai, H., Wang, X., Cai, Y.Q., Li, Y.B., Meng, L.W., Peng, B. (2018) Laparoscopic Roux-en-Y duodenojejunostomy for annular pancreas in adults: Case Report and Literature Review. *Annals of Translational Medicine*6, 211.

de la Rosa Rodriguez, R., Fogarty, A., Israel, G.M., Sanchez, M.J. (2019) Annular pancreas in a 24-year-old woman with persistent abdominal pain. *BMJ Case Reports* 20, 12, e229223.

Dowsett, J.F., Rode, J., Russell, R.C.G. (1989) Annular pancreas: a clinical, endoscopic and immunohistochemical study. Gut 30, 130-135.

Ecker, A. (1862) Bildungsfehier des Pankreas und des Herzens. Z Art Med. 14, 354.

Edman, J., Turaihi, H., Patel, B., Timmerman G, Thambi-Pillai T. (2019) Annular pancreas in Adults: Case Report and Literature Review. *South Dakota Journal of Medicine* 72, 54-57.

- Jadvar, H., Mindelzun, R.E. (1999) Annular pancreas in adults: imaging features in seven patients. Abdominal Imaging 24, 174–177.
- Leeco, T.M. (1910) Zur Morphologie des Pancrease. Sitzungsb Akad Wiss Math-Nat Ki Wien 119,391-406. Nur, N.M., Artan, A., Omar, A.A., Ahmed, M.R. (2022) Annular pancreas causing duodenal obstruction in 23 year old women managed surgically for gastrojejunostomy; a case report. International Journal of Surgical Case Reports101, 107804.
- Patra, D.P., Basu, A., Chanduka, A., Roy, A. (2011) Annular pancreas: a rare cause of duodenal obstruction in adults. *Indian Journal of Surgery* 73, 163–165.
- Sandrasegaran, K., Patel, A., Fogel, E.L., Zyromski, N.J., Pitt, H.A. (2009) Annular pancreas in adults. *American Journal of Roentgenology* 193, 455–460.
- Shirkhoda, A. (2000) Anomalies and anatomic variants of pancreas. In Gore RM, Levine MS, editors. Textbook of gastrointestinal radiology. Philadelphia: WB Saunders; 1754-66.
- Singh, S.K., Agrawal, D.K., Suganita, et al. (2016) Annular pancreas causing duodenal obstruction: a case report. *Journal of Evolution of Medical and Dental Sciences* 2016;5(5):296-300,
- Tieddmann, F. (1818) Uber die Verschiedenheiten des Ausfuhrungsgangs der Bauchpeicheldruse bei dem Menshen und Sauugetiern. Dtsch Arch Physiol 4, 403.
- Zyromski, Nicholas, J., Sandoval, John, A., Pitt, Henry, A., Ladd, Alan P. Fogel, Evan L. Wissam E. Sandrasegaran, Kumar Amrhein, David W. Rescorla, Fredrick J. Howard, Thomas J. Lillemoe, Keith, D., Grosfeld, Jay, L. (2008)Annular Pancreas: Dramatic Differences Between Children and Adults Journal of American College of Surgeons 206, 1019-1025