Case Report

Annular pancreas: the failure of side-to-side gastrojejunostomy anastomosis and the success of repair with Roux-en-Y gastrojejunostomy

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ABSTRAK

Pankreas annular adalah kelainan kongenital yang khas dengan terbentuknya cincin pankreas yang mengelilingi duodenum secara utuh atau sebagian. Kami melaporkan sebuah kasus pankreas annular pada pasien anak berusia 15 hari yang datang ke rumah sakit dengan keluhan muntah berulang dengan penurunan berat badan yang bermakna. Foto rontgen abdomen menunjukkan pembesaran lambung dengan pasase gas yang terkumpul pada duodenum proksimal. Laparotomi terbuka dilakukan dan ditemukan adanya pankreas annular yang mengelilingi duodenum proksimal yang menyebabkan hambatan pasase melewati duodenum. Pasien ini dilakukan side-to-side gastrojejunostomy *dan stoma dibuat melewati obstruksi, dan didapatkan pasasenya memuaskan. Empat hari setelah operasi, pasien kembali muntah-muntah dengan abdomen yang membesar. Foto abdomen menggunakan kontras menunjukkan tidak adanya pasase kontras melalui stoma yang dibuat. Terjadi striktur atau re-stenosis pada stoma yang pertama dibuat. Hasil yang memuaskan tampak dengan teknik* Roux-en-Y gastrojejunostomy bypass anastomosis. *Pasien diperbolehkan pulang setelah 24 hari perawatan di rumah sakit tanpa sekuele yang tidak diinginkan.*

ABSTRACT

Annular pancreas is a rare congenital anomaly characterized by a partial or complete encirclement of ectopic pancreas tissue around the duodenum. We report a case of annular pancreas in a 15 day-old infant admitted to the hospital with complaints of profuse and recurrent vomiting and loss of body weight. Non-contrast abdominal X-ray showed a dilated stomach with bubbles formation around the upper abdomen. An obstruction was noted and open laparotomy was performed. Upon laparotomy, pancreatic ring encircled the proximal duodenum causing an obstruction. Side-to-side gastrojejunostomy was performed and passage through the bypass was satisfactory. Four days after the operation, vomiting and bulging abdomen ensued. Contrast abdominal X-ray demonstrated filling defects at the level of obstruction and the anastomosis made. Anastomotic stricture was suspected and thus was corrected using Roux-en-Ygastrojejunostomy procedure. Postoperative course in this patient was satisfactory and patient was discharged after 24 days of hospitalization.

Keywords: *annular pancreas, infant, Roux-en-Y gastrojejunostomy, side-to-side gastrojejunostomy*

pISSN: 0853-1773 • eISSN: 2252-8083 • http://dx.doi.org/10.13181/mji.v24i2.1207 • Med J Indones. 2015;24:115-9
• Received 17 Feb 2015 • Accepted 17 Jun 2015

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Annular pancreas is one of the rarest causes of acute duodenal obstruction in both children and adults. It is characterized by the presence of a ring of pancreatic tissue that completely or partially surrounds the duodenum. Approximately one to three out of 20,000 babies were born with this congenital disorder with higher predisposition towards male.[1]

Defects during the embryologic development of the foregut are responsible in the development of annular pancreas. The formation of pancreas begins during the 5th week of gestation. Primitive foregut develops into one dorsal and two ventral buds, in which during the 7th week of gestation ventral buds will rotate with gut as the axis to unite with dorsal bud after rotating behind the duodenum. During this embryologic process, it was postulated that annular pancreas was formed by the abnormal union of the edge of ventral bud with duodenum resulting in fibrous tissue formation around the duodenum.[2-4]

Annular pancreas causes complete or incomplete duodenal obstruction where this may lead to recurrent profuse vomiting and poor feeding. This will manifest as malnutrition and failure to thrive if there is a delay in hospital admission.[5,6] Abdominal distension is commonly found and may prompt immediate nasogastric decompression. Diagnosis of annular pancreas is usually confirmed upon laparotomy, but radiographic images with the help of contrast may aid in displaying evidences of the presence of duodenal obstruction. Immediate management of annular pancreas in children should include fluid resuscitation to prevent hypovolemic shock, correction of any electrolyte abnormalities, gastric decompression and surgical management to relieve the obstruction. Surgical options include duodenoduodenostomy, duodenojejunostomy, gastrojejunostomy or very rarely pancreatic resection.[7]

CASE REPORT

A 15 days old infant was admitted into Tarakan General Hospital (Rumah Sakit Umum Daerah Tarakan, North Kalimantan) with chief complaint of persistent vomiting. The first episode of vomiting occurs one hour after breastfeeding. The vomit was projectile and not bile-stained. From the date of delivery until hospital admission patient had been vomiting repetitively, look more exhausted and lost a lot of body weight.

The baby was delivered full-term through normal birth delivery and assisted by midwife with the birth weight of 4,000 grams. He immediately cried after delivery and passed meconium on small amount within 24 hours after delivery. Breastfeeding was initiated as soon as possible.

During physical examination, the baby looked severely ill and dehydrated. The infant's heart rate was 160 x/minute, respiratory rate 48 x/minute, and temperature 36.6°C. The baby's weight upon admission was 2,800 grams and his body length was 68 cm. The upper abdomen was moderately distended, no mass was palpable and peristalsis was prominently seen in the upper abdomen. There was no abnormal peristalsis heard upon auscultation.

Laboratory examination showed no significant abnormalities. However, abdominal x-ray photo revealed distended stomach with bubbles of gas in the right upper quadrant of abdomen. Distended abdomen and bubbles of gas raised suspicion of the presence of obstruction although the level of obstruction still could not be determined as of this moment. Barium meal was not given at this point, because the diagnosis of obstruction was already appropriately established to start surgical exploration without further investigation.

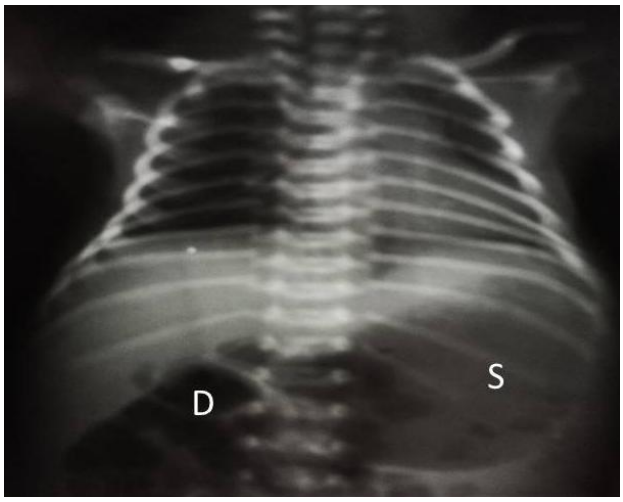

Figure 1. Plain X-ray of the abdomen shows distended stomach(S) and passage of gas in the proximal duodenum (D). Presence of bowel gas distal to the proximal duodenum is hardly portrayed in this photo

Operation was carried out after both the state of dehydration and malnutrition had been resolved. After general anesthesia was performed, supraumbilical transverse incision was made. Upon opening the peritoneum, a dilated stomach approximately as large as eight centimeter was exposed containing fluid and no passage of fluid was apparent. The second part of the duodenum was apparently constricted by a ring of pancreatic tissue that completely surrounded the duodenum and was continued with the head of pancreas. Upon identifying the obstruction, it was decided to bypass the obstruction by performing side-to-side gastrojejunostomy. Upon the completion of side-to-side gastrojejunostomy, passage through the by-pass was tested and seemed to be patently working well. After that, abdominal cavity was adequately irrigated with normal saline and surgical wound was closed.

Postoperatively the nasogastric tube was inserted and remained in place for the next three days. The patient was programed to start breastfeeding again three days after the operation in which during that period of time he received parenteral nutrition under the close supervision of pediatrician. After three days, nasogastric tube was taken off because of production of vomit through nasogastric tube was minimal. Unfortunately, the patient started to vomit repetitively on the next day after the nasogastric tube was taken off. The vomit was again projectile and bilious; therefore initiatives were taken to re-examine the patient. Barium meal examination was performed. It was clear that the barium contrast only going to fill the stomach and proximal part of duodenum. No contrast was clearly seen entering the by-pass that has been made during the operation.

Consequently, another laparotomy was performed to correct anastomotic stenosis. With the confirmation of anastomotic stenosis, another bypass was constructed using Roux-en-Y gastrojejunostomy technique. Postoperatively, patient showed considerable progress in recovery, no vomiting was observed after nasogastric tube was taken out. The patient was discharged after 24 days of hospitalization. During the follow-up at the patient's age of 11 months old, events of profuse vomiting were no longer observed. The patient's body weight was also increased steeply as it was reached 7.5 kg on the follow-up examination.

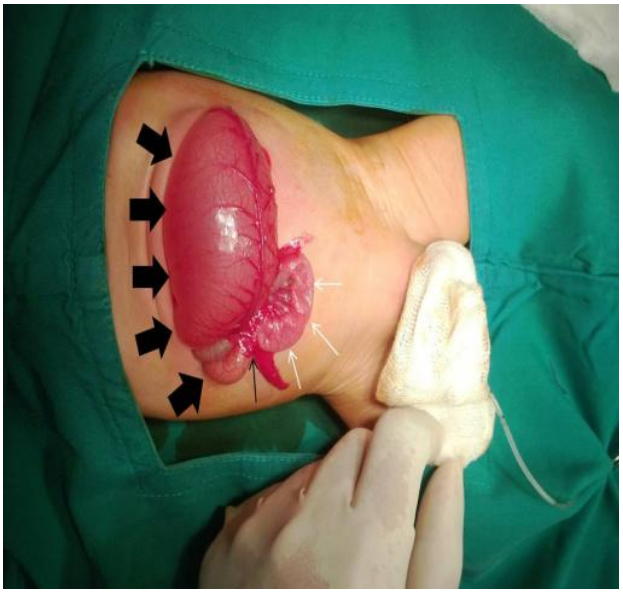

Figure 2. Intraoperative: dilation of the stomach and proximal duodenum is clearly visualized (black arrowheads) due to a complete annular band of pancreatic tissue that constricted the second part of the duodenum (black arrow). Distal to site of obstruction, the distal or third part of was collapsed (white arrows)

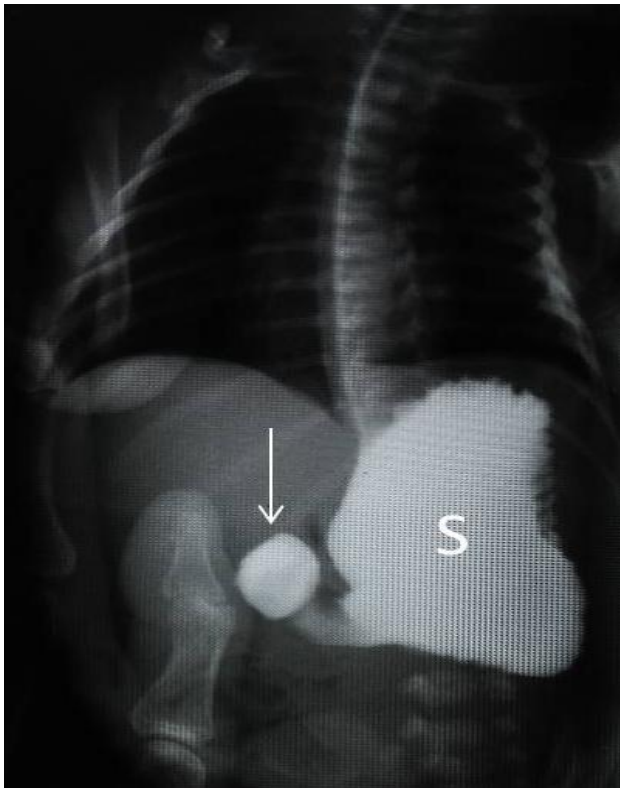

Figure 3. Barium Study taken after side-to-side gastrojejunostomy. Barium contrast only fills the dilated stomach (S) and duodenum proximal to the obstruction (white arrow)

DISCUSSION

Annular pancreas is a congenital abnormality that is characterized by the formation of pancreatic tissue that partially or fully encircles the duodenum. Annular pancreas, with its rare incidence, may be symptomatic in the early stage of life or can go unnoticed until adulthood depending on the degree of obstruction.[8,9]

Symptoms of annular pancreas in adults are commonly associated with complications of peptic ulcer, pancreatitis, duodenal, and biliary obstruction.[9,10] In infants, on the other hand, clinical manifestations revolve around duodenal obstruction in which leads to abdominal distention and profuse vomiting that leads to failure to thrive if left unresolved. Annular pancreas sometimes associates with other congenital abnormalities such as Down syndrome, esotracheal fistula, anal imperforation, Hirschsprung disease[11] and Smith-Lemli-Opitz syndrome.[12]

The diagnosis of annular pancreas can be suspected prenatally with the findings of double-bubble signs accompanied with hyperechogenic band around the duodenum during ultrasound examination.[13] After birth, simple radiograph of the abdomen may signify the presence of duodenal obstruction. These signs, however, are also commonly observed in other conditions such as duodenal web, duodenal atresia, and midgut volvulus.[6] Despite its role as pathognomonic finding, double bubble signs may not be present in all cases of duodenal obstruction[14] as with the present case. A barium enema can also be performed to further confirm the presence as well the level of obstruction. Nevertheless, in 40% of annular pancreas cases, the diagnosis was made by visual confirmation during laparotomy.[14] Other imaging modalities such as CT-scan, endoscopic retrograde cholangiopancreatography (ERCP) and magnetic resonance cholangiopancreatography (MRCP) may provide rapid anatomic depiction of pancreatic ring encircling the duodenum in a number of adult patients.[3,15]

The relief of duodenal obstruction surgically is the main goal in the management of patients with annular pancreas. The relief of obstruction is preferably achieved by creating anastomosis that will bypass the obstruction. The procedures that may be used include gastrojejunostomy, duodenojejunostomy or duodenoduodenostomy.[4,14,15] The choice of which procedure to be performed has been an argument among different authors. Fu, et al[1] stated that each procedure is proper for certain type of patient. In younger patients, duodenoduodenostomy or duodenojejunostomy is preferred because the need of vagotomy may be minimized. Up until today, however, there has not been enough data to conclude which procedure provides better outcome and it requires further research. Attempts to resect or divide the annulus can be extremely difficult to achieve as the pancreatic tissue can lie intramurally with no dissection plane.[14] Furthermore separation of annulus from the duodenum has been associated with many complications, including duodenal leakage, pancreatitis and pancreatic fistula.[1,4,14,15]

In our patient, side-to-side gastrojejunostomy was initially performed. This procedure, however, was complicated with incidental anastomotic strictures or re-stenosis. Therefore, this necessitates the repair of by-pass with Roux-en-Y gastrojejunostomy. In a study conducted by Ezomike, et al[16] repair of anastomosis is as high as 17.4% (out of 23 patients) which is due to anastomotic leakage and strictures. They also stated that repair of anastomosis or reoperation is related to higher mortality.[16] In our case, nevertheless, result from post-repair has been satisfactory and uneventful until the date of the most recent follow-up. This is probably attributable to the time of presentation to the hospital and meticulous pre-operative resuscitation with the appropriate parenteral nutrition.[16] A number of studies also report high mortality rate associated with prolonged gastric stasis and sepsis[16,17] which is in contrast to our patient.

In conclusion, symptomatic presentation of annular pancreas in infants should prompt immediate relief of obstruction by the means of surgical by-pass. Symptomatic patients may present with profuse vomiting (bilious or non-bilious), abdominal distension as well as failure to thrive. Supporting diagnostic tools such as plain abdominal radiograph and barium enema may confirm the presence of obstruction but not the real cause as 40% of annular pancreas are actually visually confirmed during laparotomy.

Adequate pre-operative resuscitation without delaying definitive surgical by-pass is important to achieve better prognosis for the patient with lower mortality. The patency of the anastomosis must be also monitored carefully before patient is discharged. The presence of anastomotic stricture prompts immediate repair of anastomosis.

Conflicts of interest

The authors affirm no conflict of interest in this study.

Acknowledgment

Many thanks is given to Tarakan General Hospital in Tarakan, North Kalimantan and the close relatives of the patient in giving their consent in regard to the publication of this article.

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