The Evolution of Double Pylorus Associated With NSAIDs-Induced Gastric Ulcer

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ABSTRACT

Double pylorus is an uncommon condition where two communicating channels between the gastric antrum and duodenal bulb exist and found incidentally by esophagogastroduodenoscopy. This case report presented a 77-year old Timorese man with long-term frequent use of nonsteroidal anti-inflammatory drugs (NSAIDs) who came to hospital because of hematemesis and melena. The initial endoscopy showed gastric ulcer and the double pylorus. Nine months later, the double pylorus structure fused into one large opening. This elderly patient with a history of NSAIDs abuse was diagnosed with acquired double pylorus due to a gastric ulcer complication. The fusion of the two channels occurred as a result of septum destruction, likely caused by fistulous tract ulceration or mechanical damage from food bolus. This structure predisposes the patient to bile regurgitation and ulcer recurrence, emphasizing the need for continued monitoring and avoidance of ulcerogenic factors.

Keywords : *Double pylorus, gastroduodenal fistula, NSAIDs, gastric ulcer, follow-up*

ABSTRAK

Pilorus ganda adalah suatu kondisi yang cukup jarang, dimana terdapat dua kanal antara antrum gaster dan bulbus duodenum yang secara kebetulan ditemukan pada pemeriksaan esofagogastroduodenoskopi. Laporan kasus ini mendeskripsikan seorang laki-laki bersuku Timor berusia 77 tahun dengan riwayat penggunaan obat anti inflamasi non steroid (OAINS) yang datang ke rumah sakit dengan hematemesis dan melena. Pada pemeriksaan endoskopi inisial ditemukan ulkus gaster dan pylorus ganda. Sembilan bulan kemudian, struktur pylorus ganda tersebut menyatu menjadi suatu orifisium yang besar. Pasien yang berusia lanjut dengan riwayat penyalahgunaan OAINS ini didiagnosis dengan pylorus ganda akuisata sebagai akibat komplikasi ulkus gaster. Fusi antara kedua kanal tersebut terjadi akibat destruksi septum, biasanya disebabkan oleh ulserasi traktus fistula atau kerusakan mekanik akibat bolus makanan. Struktur ini meningkatkan risiko pasien untuk mengalami regurgitasi cairan empedu dan rekurensi ulkus, menekankan perlunya pemantauan berkala dan penghindaran dari faktor-faktor ulserogenik.

Kata kunci : *Pylorus ganda, fistula gastroduodenum, OAINS, ulkus gaster, pemantauan*

INTRODUCTION

Double pylorus (DP) is an uncommon condition when two communicating channels between the gastric antrum and duodenal bulb exist. The two separated orifices of the pylorus can be seen endoscopically leading to the duodenal bulb.[1,2] The prevalence of DP is reported about 0.001-0.4%.[3] Patients with DP usually have no symptom related to the structure, but they often present with indications of EGD such as hematemesis melena or chronic anemia.[4]

According to the etiology, DP can be categorized as acquired and congenital. Acquired DP is preceded by ulcer associated by chronic use of NSAIDs or Helicobacter pylori infection.[2,3,5–7] The congenital form of DP is less often than the acquired one and can be presumed when the patient has another congenital anomaly or without history of peptic ulcer.[8] The outcome of the DP over time can be variable. The two channels later can be unchanged, fused into one single hole, or very rarely can be complicated into three channels.[7,9,10] This case report aimed to provide insight about this rare condition especially to new endoscopists that hopefully can help them in differentiating it with the congenital type and knowing factors that trigger the formation of such structure.

CASE ILLUSTRATION

A 77-year old male patient was admitted to Marianum Halilulik Catholic Hospital, Belu Regency, East Nusa Tenggara, with the chief complaint of having black stool and vomit. Six hours before admission, the patient had coffee ground colored and foul smelling stool. In the previous morning, patient vomited black colored liquid of total about 400 mL. He also felt epigastric pain. Patient had history of frequent NSAIDs consumption since 3 years ago because of painful swelling in his feet. The swelling decreased and pain improved after he took mefenamic acid or ibuprofen, sometimes both drugs were consumed together. There was no history of using the anti-thrombotic drugs and the patient didn't have significant weight loss. He was an occasional alcohol drinker and totally stopped the consumption 4 years ago. This is the first episode of hematemesis melena and he never felt epigastric pain or the history of stomach acid medication before.

Patient was full alert when admitted to emergency room. His blood pressure was 76/52 mm Hg with the heart rate 102×/minute. Initial assesment was hemorrhagic shock and after fluid rescusication of 500 mL of ringer's lactate, the vital sign was back to normal. Because of unavailability of endoscopy unit at that hospital, the patient was managed conservatively. Patient was fasted and given omeprazole 40 mg i.v. twice daily. Physical examination showed normal abdomen with no stigmata of liver cirrhosis. The blood examination showed anemia with hemoglobin was 7,1 g/dL and platelet was 224.000/µL. Serial measurement of hemoglobin showed stable value indicated that the bleeding was already stopped. Transfusion of three bags of packed red cell was given with the expected increased hemoglobin. Patient then started clear liquid feeding at day two of the treatment with careful observation of rebleeding. He was informed about the next diagnostic step and referred to Atambua as the referral center of the regency, but he refused because he felt the improvement already. Finally after 4 days of treatment, patient was discharged with double dose omeprazole and strictly prohibited to consume NSAIDs.

The patient then had the esophagogastroduodenoscopy (EGD) examination in an outpatient care in Mgr. Gabriel Manek General Hospital Atambua, 1 week after previous hospital discharge. The EGD documented mild hyperemic mucosa in gastric body and antrum and then diagnosed histopathologically as chronic gastritis with no evidence of Helicobacter pylori. There were two openings separated by bridging tissue at the end of the antrum created double pylorus appearance. There was peristaltic opened and closed movement in the first opening suggested the true pylorus. The second opening showed no contraction and when the scope entered, it was led to duodenal bulb confirming it was not a gastric diverticulum. There was also a clean white based ulcer (class III according to Forrest Classification) at small curvature side of pre-pyloric region of the antrum, which explained the cause of hematemesis and melena. The first EGD findings can be seen in **figure 1A & 1B.**

The PPI treatment was continued and after one month of PPI treatment, the patient was scheduled for endoscopic evaluation. It showed the unchanged double pylorus structure with healed previous ulcer and some raised erosions around the fistula **(figure 1C).** To rule out the possibility of false negative result, the H. pylori Urea Breath Test (UBT) was done after PPI washout for two weeks after the PPI was stopped and showed negative result. The PPI treatment then continued until total of 12 weeks to complete the healing and prevent the ulcer recurrence. The swelling of his feet was diagnosed as acute gouty arthritis and treated with colchicine. The arthritis improved without signs or symptoms of gastric ulcer's recurrence.

Nine months after the initial EGD, the patient came to the hospital with general weakness. He denied having black stool or vomit like before. The laboratory showed anemia with hemoglobin 6.4 g/dL with normal erythrocyte index. To make sure there is no recurrent bleeding from the ulcer, we did the third EGD to the patient. The EGD showed antral gastritis and one large opening connecting the antrum and duodenal bulb suggested as fusion between the two previous channels. There was also sign of perforated mucosal bridge and the remnant mucosa of the bridge looked erythematous **(figure 1D).**

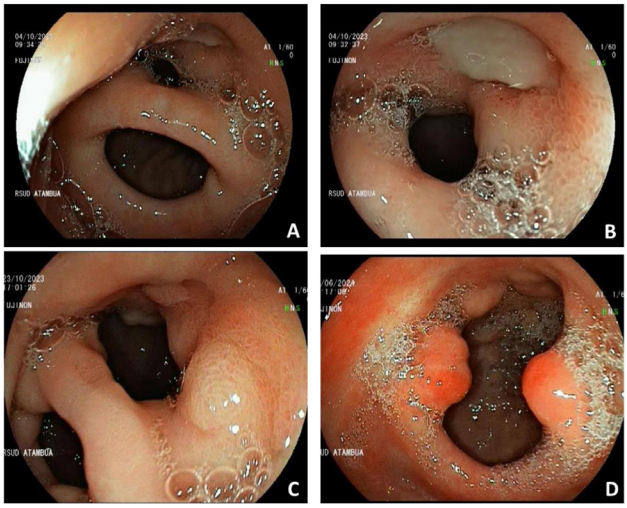

Figure 1. The evolution of double pylorus. A, Initial EGD showed true pylorus (lower opening) separated by mucosal bridge with fistula (upper opening). There is also marked erosion in the center of mucosal bridge and the ulcer along the fistula tract. B, Close up view of clean based ulcer along the fistula tract. C, One month after PPI treatment the ulcer was healed with raised erosions around the fistula. D, Nine months after the initial EGD fusion between the fistula and pylorus occured with sign of perforated mucosal bridge, the remnant mucosa of the bridge looked erythematous.

We also did the colonoscopy to the patient which showed normal findings. The urinalysis showed microscopic hematuria which can be the cause of anemia. The abdominal ultrasonography then revealed grade I right hydronephrosis with proximal ureteral stone, grade IV left hydronephrosis with mid ureteral stone, and multiple cysts in the right kidney. The patient then was refered to urologist.

DISCUSSION

Double pylorus also known as gastroduodenal fistula is a rare condition with a prevalence of 0,001-0,4%.[3] By on line searching, we found only one case of DP from Indonesia which was reported by Oktaricha and Miftahussurur.[3] As found in our case, most of the fistulas in DP occur on the lesser curve aspect of the gastric antrum.[3,6,7] It is thought that the route of penetration on the lesser curve is the shortest so the fistula is easier to be formed although the fistula also reported to be found on the posterior wall, greater curvature, and anterior wall of the stomach.[1,11]

The acquired type of DP is possibly formed by the advanced erosion between the two ulcers, one a distal gastric ulcer and the other a duodenal ulcer. It then creates a communication between the two areas.[1] Another proposed mechanism is the effect of gastric scarring which can attract the lesser curvature wall which is weakened because of reccurent ulcer healing and later perforates into the duodenal bulb.[2,12] Hattori et al. reported the false pylorus formed by perforated prepyloric ulcer which is possiblly caused by increased antral pressure in hypertrophic pyloric stenosis in an adult.[13]

Desmukh et al. in 2020 documented the development of pre-pyloric ulcer into gastroduodenal fistula in a cirrhotic patient. In two serial endoscopic examination over the period of 7 years, the initial antral ulcer later fistulized into duodenal bulb and created the double pylorus appearance.[2]

The congenital type of DP is less often than the acquired one.[2] The etiology of congenital double pylorus has been attributed to a tubular duplication of the pylorus which is very rare and reported in only 1 out of 281 of all gastrointestinal tract duplication cases published in English.[8] The possibility of congenital double pylorus is greater if patient is young with no history of peptic ulcer disease, and has other gastrointestinal congenital abnormalities, for example pancreas divisum or heterotropic pancreas.[7,14]

Double pylorus itself has no specific signs or symptoms. Incidentally the lesion is found during EGD examination.[3] The symptoms like vomiting and epigastric pain are more related to gastric ulcer which has a role as a trigger of a DP formation.[7] Thick bridging tissue between the orifices can cause symptoms of gastric outlet obstruction.[15] Because of the pyloric incompetence, bile reflux can happen and it can contribute to a persistent ulcer that can not be healed by usual medication.[7] The risk of recurrent ulceration can also be increased due to poor epithelization of the fistulous tract.[16]

In this case, the most likely diagnosis is acquired DP because the patient was quite old, with the evidence of gastric ulcer. No peristaltic contraction in the second orifice also supports the presumption of the acquired nature of the orifice. The data about congenital DP is

limited. Sufian et al. reported the congenital double pylorus by surgical exploration and they confirmed the lesion by histological findings.[14]

The frequent use of NSAIDs and the presence of Helicobacter pylori infection will infuence the course of the gastric ulcer that leading to development of fistula.[17] In this patient, there was history of NSAIDs abuse and the role of H. pylori was eliminated by the negative UBT result. Besides ulcerogenic agent like NSAIDs itself, double pylorus is also associated with chronic diseases like diabetes mellitus, chronic lung disease, and several autoimmune diseases. They are related with the poor healing of the ulcer and initiating the formation of double pylorus.[3,11]

After a month of PPI treatment, second EGD showed the persistence of the double pylorus and the ulcer was healed. Nine months after the initial endoscopy the DP evolved into a single large opening due to septum destruction between the two holes. It was reported that about 27% of the patients with acquired double pylorus will have fusion between the fistula and pylorus.[10]

Fousekis et al reported a case with such an evolution after 2 years of the initial diagnosis.[7] The possible mechanisms of septal destruction may be related to fistulous tract ulceration or mechanical damage by food bolus.[18] Anticipating the ulcer recurrence in this patient is important because bile regurgitation by this new non-physiologic large opening predisposes the ulcer formation.[10] Not only being a single large opening, the outcome of DP is variable. About 60% of DP is unchanged throughout patient's lifetime. The other 25% of the fistula will be closed.[17] Very rarely, it also can be complicated from two into three openings.[9]

To date, no standardized guidelines exist regarding optimal follow-up intervals in patients with a double pylorus. This report provides follow-up data from an endoscopy performed at a 9-month interval due to anemia, which was confirmed not to be caused by recurrent ulcer bleeding. Consistent with other studies, reassessment should be considered when patients develop new symptoms or complications.

The treatment of acquired DP is focused on factors that influence the healing process of mucosa in gastric ulcer.[19] Drugs like NSAIDs and corticosteroids should be avoided. If present, H. pylori infection must be eradicated. PPI therapy should be given as indicated in peptic ulcer. Controlling that factors gives advantages in relieving the symptoms, prevent the ulcer recurrence, and closing the fistula.[7]

Surgery is extremely rare to be performed except in some conditions such as free perforation, obstruction that are refractory to endoscopic treatment, refractory bleeding, and persistent symptoms after optimal medical therapy.[6] In procedure using side view endoscopy, the presence of double pylorus can contribute to complication, so the endoscopists must take proper anticipation before the scope passes through this kind of anomaly.[7]

CONCLUSION

Double pylorus is a rare condition, often acquired as a complication of gastric ulcer associated with NSAIDs use or Helicobacter pylori infection, and is typically discovered incidentally during endoscopy. There is no special treatment of DP but monitoring the lesion endoscopically can be useful to assess the potential complication like bile regurgitation and ulcer recurence.

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