Primary aortoduodenal fistula due to a swallowed sewing needle: a rare cause of gastrointestinal bleeding

Dikiş iğnesi yutulması sonucu oluşmuş primer aortoduodenal fistüle bağlı nadir görülen bir masif üst gastrointestinal sistem kanaması

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A primary fistula between the abdominal aorta and the duodenum is rare and usually fatal. Atherosclerosis remains the most common etiologic factor, accounting for more than two-thirds of the cases reported. Other etiologies include carcinoma, ulcers, radiation, aortitis and foreign bodies including sewing needle, cocktail stick, open safety pin and fishbone. We report a case of a 17 year-old girl who underwent surgical treatment because of severe upper gastrointestinal bleeding which was related to an aortoduodenal fistula caused by a swallowed sewing needle. At operation, a chronic aortoduodenal fistula that contained the sewing needle was found and repaired. This is the fourth case in the literature in which a needle was found to be associated with the development of an aortoenteric fistula.

Key Words: Primary aortoduodenal fistula; swallowed sewing needle; gastrointestinal bleeding.

Abdominal aorta ve duodenum arasında primer fistül oluşumu nadir görülen ve yüksek mortaliteye sahip bir durumdur. Ateroskleroz en sık görülen fistül nedenidir; bildirilen olguların üçte ikisinden fazlasında sorumludur. Kanserler, ülser, radyasyon, aortit ve yabancı cisimler diğer etyolojik nedenlerdir. Burada dikiş iğnesi yutulması sonucu oluşan aortoduodenal fistüle bağlı masif üst gastrointestinal sistem kanaması nedeniyle ameliyat ettiğimiz 17 yaşındaki olgu sunuldu. Ameliyatta dikiş iğnesi içeren kronik aortoduodenal fistül bulunarak onarıldı. Olgumuz iğne yutma nedeniyle İngilizce literatürde sunulan dördüncü aortoenterik fistül olgusudur.

Anahtar Sözcükler: Primer aortoduodenal fistül; dikiş iğnesi; gastrointestinal kanama.

Primary and secondary aortoenteric fistulas are infrequent causes of gastrointestinal bleeding. Primary aortoenteric fistula (PAEF) is defined as a communication between the aorta and the gastrointestinal tract. PAEF is less common than secondary fistulae which represent a well known complication of surgery for abdominal aortic aneurysms.[1] The incidence of PAEF has been reported to be 0.04-0.07% in large autopsy series, whereas it occurs in 0.69-2.36% of patients with an aneurysm of the abdominal aorta.[2] Atheriosclerosis, leading to formation of an aortic aneurysm, remains the most common etiological factor, accounting for more than two-thirds of the cases reported.[3,4] Other causes of PAEF include radiation, gallstones, carcinoma, mycotic aneurysm, septic aortitis due to salmonella infection, diverticular abscess, duodenal ulcer and foreign bodies.[1,2,5,6]

Recently, we successfully treated a patient with overt upper-gastrointestinal bleeding due to an aortoduodenal fistula caused by a foreign body, sewing needle, at our institution.

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CASE REPORT

A 17 year-old girl was admitted to our gastroenterology department because of melena and abdominal pain in the epigastrium. She had a similar episode with syncope, haematochezia and abdominal pain 15 days before admission and at that time, she was admitted to another hospital, where she underwent an upper-gastrointestinal tract endoscopy and an abdominal ultrasound; both of them were found to be normal. Her hemoglobin levels dropped only slightly from 14.3 to 12.6 g/dl; she remained haemodynamically stable and was discharged one week later.

The patient was well until four days before her admission to our institution. Upon admission, the patient had an episode with haematochezia, melena and syncope. Her blood pressure was 100/70 mmHg with orthostatic hypotension, regular heart rate of 80 /min, and respiratory rate of 18/min. Rectal examination revealed melena. There was no other finding at the physical examination.

Initial laboratory examination revealed hemoglobin 11.2 g/dl, white blood count 11200/mm^3, urea 24 mg/dl, creatinine 0.3 mg/ dl, glucose 119 mg/dl, total bilirubin 0.44 mg/dl, alanine aminotransferase (ALT) 73 IU/I, aspartate aminotransferase (AST) 75 IU/I, alkaline phosphatase 250 U/I, gamma glutamyltranspeptidase (GGT) 126 U/I, lactic dehydrogenase (LDH) 403 U/I and prothrombine time 16.2 s. Chest radiography and electrocardiogram were normal, while plain abdominal radiograph revealed a metallic foreign body localized paravertebrally (Fig. 1); abdominal ultrasound and upper gastrointestinal endoscopy assessed to be 'normal'.

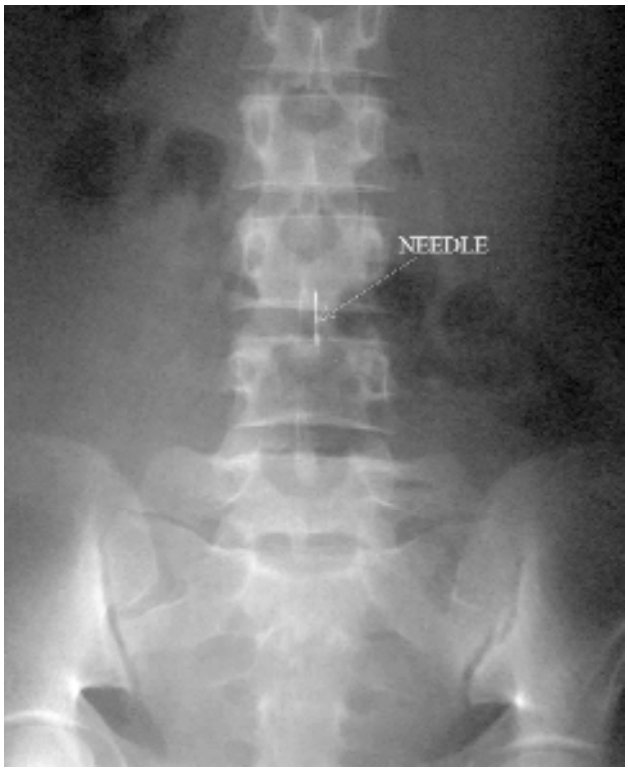

Fig. 1. Plain abdominal radiograph showing the needle.

The patient received only intravenous fluids administration and remained haemodynamically stable. However, three days after admission, she had a new, more severe bleeding episode with epigastric pain, haematemesis, recurrent haematochezia and circulatory collapse. Her blood pressure dropped to 50/0 mmHg, but she was bradycardic at that time with a sinus rate of 50/min. Hemoglobin levels dropped to 3.8 g/dl. The patient received four units of whole bloods and intravenous colloid (Gelofusine) and crystalloid fluids.

The patient was rushed to the operating theatre. At emergency laparotomy a vast amount of blood was found in the intestinal tract. The retroperitoneal part of the duodenum was adherent to the aorta. After mobilization of the duodenum, a gastroduodenotomy was performed and a chronic aortoduodenal fistula that contained the sewing needle was visualized. The duodenal and aortic openings were closed after debridement of the inflamated tissues both on the duodenum and the aorta (Fig. 2). Samples of the fistula and the duodenum were sent to the pathology department. Histological examination of sections taken from the region of the fistula revealed chronic ulcerative inflammatory lesions with formation of granular tissue and presence of granulomatous reaction with dense fibrosis around the sewing needle.

Her hospital stay was uncomplicated and the patient was discharged from hospital at postoperative 12th day. She is currently doing well at 18th month of follow-up.

DISCUSSION

PAEF is a rare condition. The most frequent cause is arteriosclerosis. The pathogenesis is usually based on direct adhesion of a segment of the gastrointestinal tract to an aortoiliac aneurysm, followed by progressive erosion through the bowel wall and

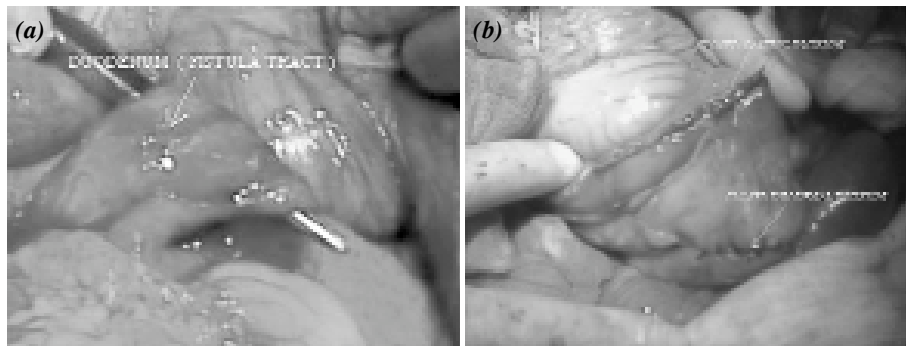

Fig. 2. **(a)** The fistulous opening on the duodenum incision. **(b)** Closed gastric and duodenal incisions.

secondary contamination by intestinal flora. The ensuing infection further contributes to necrosis of the adjacent aortic wall with gastrointestinal hemorrhage as a consequence of necrosis. Any part of the gastrointestinal tract may be implicated,[7] but the most common site of involvement is the third part of the duodenum (70%). It is presumed that this is the result of a relatively fixed position of the third portion of the duodenum to the superior mesenteric vessels anteriorly and the aorta posteriorly.[2,8,9] In our case the fistula tract was between the third portion of the duodenum and the abdominal aorta.

Bleeding due to an aortoenteric fistula has rarely been reported to be associated with foreign bodies, including sewing needle, cocktail stick, open safety pin and fishbone.[1,10,11]

Grande et al., studied 28 autopsied cases of aortoenteric fistulas and found that the most common predisposing factor was an aortic prosthetic graft. The second most common setting for the development of an aortoenteric fistula was gastrointestinal carcinoma. Surgically untreated aortic aneurysms, infective aortitis, idiopathic hypertrophy of the esophagus that was associated with spontaneous mucosal lacerations extending into the muscularis propria layer were other rare causes of AEF. One of the these 28 patients, a severely mentally retarded 19 year old woman swallowed an opened safety pin, which subsequently perforated both the midesophagus and the distal portion of the aortic arch. The fistula that resulted with death by exsanguination. Histological examination of the fistula revealed extensive suppurative inflammation and young granulation tissue.[10] A similar case, in which aortoenteric fistula was due to ingestion of a seamstress needle had been reported by Hambrick.[12] The third case has been reported by Voorhoeve et al.[11] In this case fistula was due to a cocktail pin. The present case is the fourth case that has been reported.

Clinical manifestations of an aortoenteric fistula include abdominal pain, upper gastrointestinal bleeding and a pulsatile mass in the epigastrium. In a systematic review of 118 patients with primary aortoenteric fistulae in the world's literature, abdominal pain was reported in 32%, abdominal mass in less than 25% and gastrointestinal bleeding as an initial symptom in 64% of the patients.[3] The initial bleeding commonly known as the 'herald bleeding' is often transient and self-limiting owing to the formation of a thrombus. Bouts of bleeding recur over a period of hours, days or weeks, eventually culminating in massive hemorrhage and hypovolemic shock.[8] Most experts recommend particular attention to the herald or sentinel bleeding episode, which is usually followed by massive bleeding a few hours or days later.[1,2,4,8] It is noteworthy that the time interval between the herald bleed and the catastrophic event exceeds one week in more than 40% of cases as occurred in our patient.[1-3] Although one mild or moderate herald bleed has been reported in the literature, our patient experienced three herald bleeds of moderate severity before the massive catastrophic bleeding episode. Bleeding arising from a point distal to the second portion of the duodenum in the absence of a proximal lesion should raise the suspicion of PAEF.[8] The precise diagnosis can be very difficult because the bleeding may be intermittent and diagnostic tests often fail to reveal the course. Laparotomy should be performed as soon as the diagnosis is taken into consideration clinically. Many physicians support the need for immediate

exploratory laparotomy in patients with massive upper-gastrointestinal bleeding and normal endoscopy in order to avoid unnecessary delay in establishing the diagnosis.

As our case shows, PAEF is difficult to diagnose and sometimes not diagnosed until laparotomy. It mandates a high index of suspicion and recognition of the herald bleed. Survival is rare and requires aggressive surgical management.

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