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CASE REPORTS

AORTO-ESOPHAGEAL FISTULA: AN UNUSUAL COMPLICATION OF ESOPHAGO-GASTROSTOMY, FOLLOWING RESECTION FOR CARCINOMA OF THE ESOPHAGUS

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AORTO-ESOPHAGEAL fistula is a rather uncommon condition.[1] A study of the previously reported cases of aorto-esophageal fistulae shows that the majority are the result of swallowing foreign bodies,[2, 3] most of which are fish, poultry or rabbit bones. There are many cases also in which the cause of the perforation into the aorta was fungating carcinoma of the esophagus.[4] In one instance an esophageal diverticulum was responsible for the perforation. One case has been reported in which an esophago-pleural communication was aggravated by monilial infection producing severe mycotic tension pyopneumothorax and a concomitant aorto-esophageal fistula.[5] Isolated cases of perforation of the thoracic aorta by penetrating peptic ulcers of the esophagus have been reported by several authors.[6, 7] Bullet wounds causing similar fistulae have also been noted.

In reviewing the literature, we found reported only four cases of esophageal perforation into the aorta following esophago-gastrostomy; one resection was performed for esophageal stricture due to ingestion of lye[8] and three resections were carried out for esophageal carcinoma.[9, 10] In one of the latter three cases, the perforation was caused by a misplaced suture passing through the media of the aorta and the esophago-gastric anastomosis. In another case, leakage at the suture line on the 17th postoperative day was followed by an esophago-pleural fistula, which was sealed by the juxtapositioned aorta. The third case was ascribed to the necrotizing digestive effect of acid gastric juice on the tissues of the aortic wall. In our case, the presence of yeast-like organisms (most likely Candida) in the deep tissues of the fistulous tract and their apparent invasion of the aortic wall suggest that these organisms were a probable contributory factor—if not a causative one—in the development of the fistula. The unique nature of this probable pathogenesis makes this case of interest.

A.R., a 68-year-old white man, was first admitted to the Henry Ford Hospital, Detroit, Michigan, on September 24, 1960.

He had been in good health until a month prior to admission, when he developed difficulty in swallowing. At the time of admission the patient was able to swallow clear fluids only. In the last four weeks, he lost 10 lb. Radiographic examination of the upper gastrointestinal tract was reported to show a filling defect three inches long, in the midportion of the esophagus,

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with moderate dilatation of the upper half of the esophagus.

Physical examination was essentially negative. Laboratory findings revealed a normal blood picture and negative urinalysis. His serology was non-reactive. Values for urea nitrogen, alkaline phosphatase, cephalin-cholesterol flocculation, thymol turbidity and prothrombin time were all within normal limits. Esophagoscopy showed a fungating firm lesion, 13 inches below the upper teeth. Biopsy taken from this region was reported as epidermoid carcinoma.

On October 3, 1960, a left, low posterolateral thoracotomy was performed, with resection of the seventh rib. A mass the size of a golf ball was demonstrated in the esophagus just below the left main stem bronchus. The diaphragm was opened and the lesser curvature was cleaned off from the pylorus toward the stomach, removing all the lymphatic and adipose tissue. The stomach was resected from the fundus down to a point approximately half way on the lesser curvature, which converted the stomach into a tubular structure utilizing the greater length of the greater curvature. The esophagus was transected first below the aortic arch, but since tumour cells were found in the submucosa at the line of resection, another segment of proximal esophagus was excised. After the anastomosis was completed, the stomach was tacked to the left pleura for support. Diaphragmatic closure was then completed and the stomach was tacked to the margin of the hiatus to prevent herniation of the abdominal contents into the chest.

The resected portion of esophagus was 18.5 cm. long and 5.8 cm. in circumference at the proximal line of resection. In the proximal part of the esophagus there was a large tumour measuring 5.5 cm. in diameter extending almost to the edge of the resection. The rest of the esophageal mucosa was pale pink and slightly thickened.

Microscopic examination showed well-differentiated epidermoid carcinoma extending deep into the submucosal and muscular layers. In one area, the tumour almost reached the serosa but did not penetrate it. No invasion of the perineural or perivascular lymphatics was noted. Sections taken from the separate segment of the esophagus and from 14 regional lymph nodes showed no evidence of tumour.

The patient had a violent postoperative course. An emergency tracheostomy had to be carried out on October 5 because the patient developed acute respiratory distress due to retained secretions. He became very dyspneic and apprehensive. Moist rales were heard in both lung bases and the neck veins were markedly distended. Early left ventricular failure was diagnosed and the patient was digitalized. His temperature rose to 105° F. but gradually subsided on antibiotic therapy. An electrocardiogram taken on October 10 showed atrial flutter. After a few days of intravenous feeding he was given liquids by mouth. From that time on his general condition rapidly improved and on October 27 he was ambulatory and taking five meals a day. On November 1, he was discharged from the hospital.

The patient was asymptomatic after discharge, except for some weakness. On November 10 he had an acute episode of hematemesis which soon stopped spontaneously. He had no complaints until late afternoon, when he experienced moderate epigastric discomfort, but had no more bleeding. He was seen in the hospital emergency room, where he was found to be pale, cold and clammy, and was readmitted. His blood pressure was 94/60 mm. Hg and his hemoglobin value 8.8 g. %. The hematocrit was 28%, and the white blood cell count 7600/c.mm. Physical examination showed epigastric tenderness with some muscular guarding; bowel sounds were hyperactive. On November 11 at 6 a.m., the patient had profuse hematemesis. A venous cutdown was performed on both the right and the left legs and blood was administered but no blood pressure reading was ever obtained. He vomited again, a large amount of blood, and continued bleeding until 9.35 a.m., when his heart stopped.

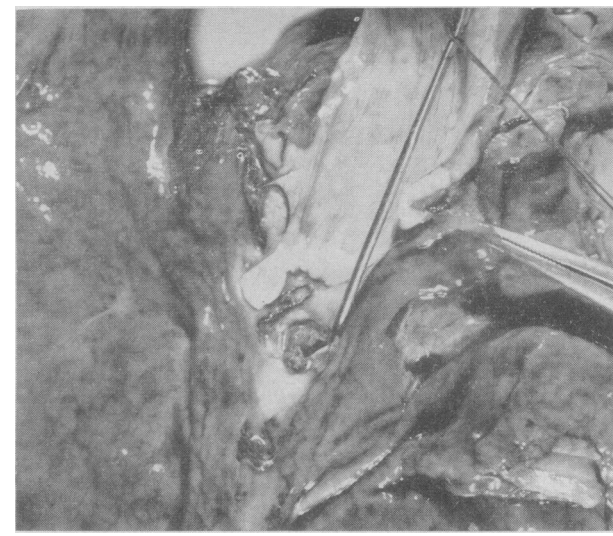

Fig. 1.—Aorto-esophageal fistula.

Postmortem Findings

Examination of the chest at autopsy showed that the esophagus had been resected at the junction of its proximal and middle thirds. A tube formed from the greater curvature of the stomach was anastomosed to the esophagus. The suture lines of this esophago-

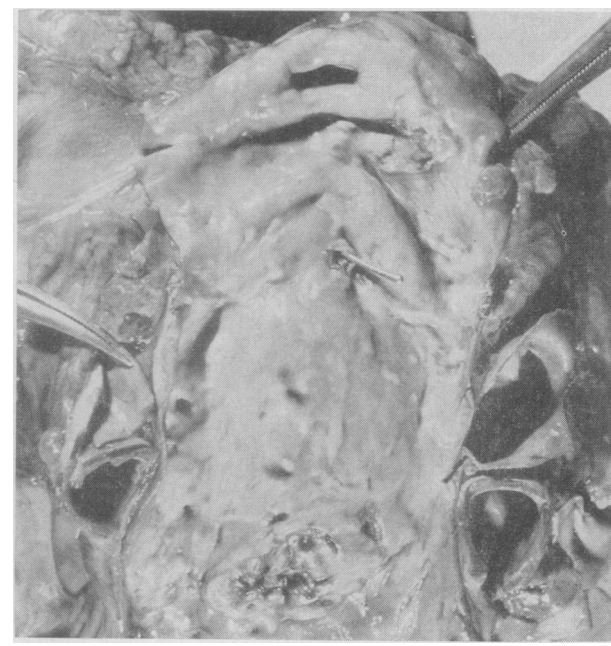

Fig. 2.—Aorto-esophageal fistula.

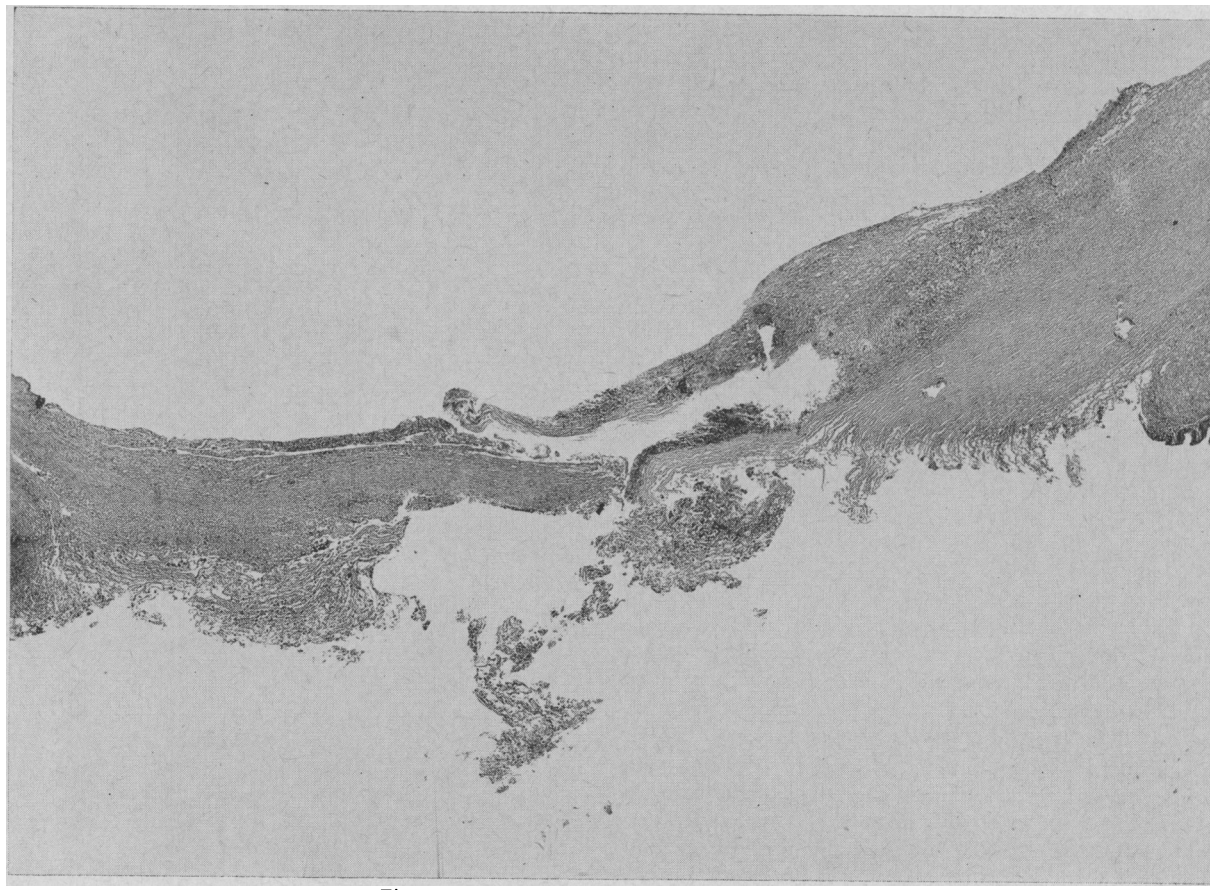

Fig. 3.—Aorto-esophageal fistula, × 22, H & E.

Fig. 4.—Aorto-esophageal fistula, × 14, H & E.

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gastrostomy were easily visible and the anastomosis appeared leak-proof. The posterior surfaces of the esophagus and the gastric tube were firmly adherent to the anterior portion of the aortic wall. The entire gastrointestinal tract was completely filled with bright red clotted blood. There was a small ulcerated area approximately 3 mm. above the gastro-esophageal anastomosis on the posterior surface of the esophagus. This ulcerated area measured 5 mm. in diameter (Fig. 1). The edges of this defect were firmly attached to the arch, forming a fistulous tract. This fistula extended to and actually penetrated the entire thickness of the anterior wall of the thoracic aorta just below the aortic wall. A probe was passed from the esophageal opening of the fistula and the aorta was opened on its posterior aspect. The probe entered the aorta approximately 2 cm. below the aortic arch. The aortic opening of this fistula measured 3 mm. in diameter (Fig. 2). There were several yellowish atheromatous plaques and foci of calcification in the aorta, but no ulceration was found. No significant plaques were present at the site of the fistulous opening. Examination of the surrounding organs, lymph nodes, and abdominal viscera failed to show evidence of residual carcinoma. Further autopsy findings were: left pleural adhesions and adhesions of the pericardial sac to both pleurae. Moderate coronary arteriosclerosis was present.

Microscopic examination of sections through the fistulous tract (Fig. 3) showed the edges of the ulcer to be necrotic, surrounded by plasma cells, histiocytes and proliferating fibroblasts. The deeper tissues around the fistula were infiltrated by yeast-like organisms invading the adventitia and media of the aorta. In this area, the fibres of the media of the aorta were broken up and contained micro-organisms (Fig. 4). Foreign-body giant cells, some containing suture material, were also present some distance from the necrotic area (Fig. 5). These areas showed the typical picture of foreign-body granulomas. Subintimal atheromatous plaques and foci of calcification were seen in the aorta. At the site of the anastomosis, the serosa of the gastric tube showed a marked granulomatous reaction with fibroblastic proliferation and numerous newly formed capillaries. Many macrophages were present, loaded with blood pigment. Some displaced esophageal epithelium was found in the serosa of the stomach with surrounding granulomatous reaction. The presence of the yeast-like organisms was confirmed by special stains (Grocott and Gridley). Since the organisms were not cultured, positive identification was not possible. Basing their identification on histological appearance only, they most likely belonged to the genus Candida (Figs. 6 and 7).

DISCUSSION

All three previously reported cases of aorto-esophageal fistula following esophago-gastrostomy for carcinoma of the esophagus have been preceded by a stormy febrile postoperative course. This applies also to our case. The time intervals between operation and the fatal hemorrhage in the three cases were 12, 81 and 40 days, respectively. Our patient survived the operation by 39 days. The causes of the perforation in the three reported cases have been noted above. As to the cause of the perforation in our patient, we can only speculate.

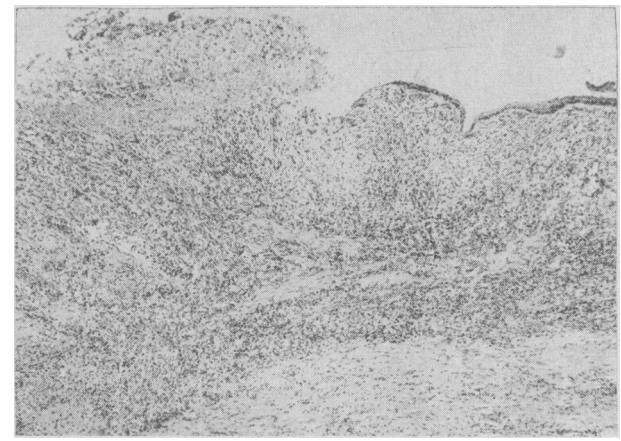

Fig. 5.—Aorto-esophageal fistula, × 77, H & E.

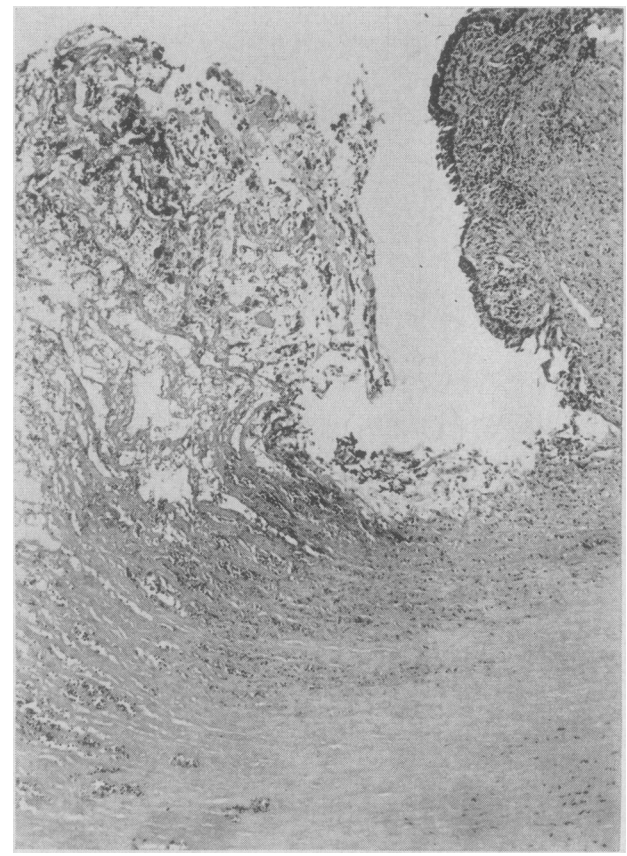

Fig. 6.—Aorto-esophageal fistula, × 90, H & E.

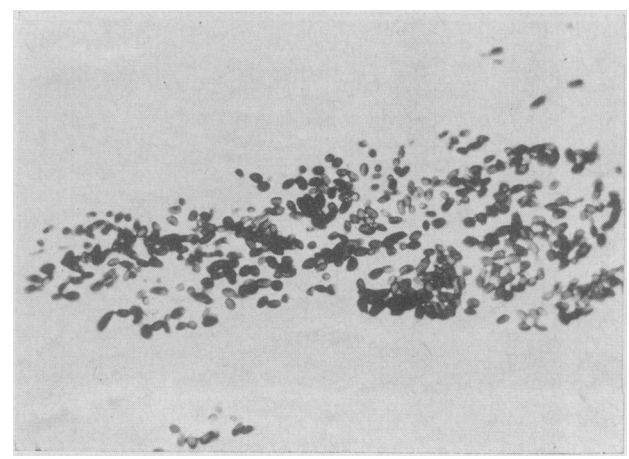

Fig. 7.—Aorto-esophageal fistula, × 865, Grocott.

Although there were a number of foreign body giant cells, some of them containing suture material, in the microscopic sections, associated with foreign-body granulomas, these were at some distance from the perforation. The granulomatous character of the inflammatory reaction at the site of perforation was more suggestive of mycotic infection than of an uncomplicated peptic ulcer. The integrity of the esophagus at the site of the perforation may have been compromised by impairment of the blood supply and thus made more readily subject to the effects of the acid gastric juice. We believe that this primarily peptic ulcer was secondarily invaded by yeast-like organisms, probably Candida. The presence of the latter, unfortunately, was not suspected at the time of the autopsy and no culture was taken. The presence of these organisms in the wall of the fistulous tract penetrating deeply into the wall of the aorta and the absence of the same organisms in the surrounding areas lead us to suspect that they probably played an important part in the causation of this perforation.

SUMMARY

A case of aorto-esophageal fistula is reported resulting in the death of the patient, who previously underwent esophago-gastrostomy for carcinoma of the esophagus. Primary peptic ulcer, due to the digestive effect of gastric juice at a site of lessened resistance, and secondary invasion by yeast-like organisms as a contributory factor, are proposed as causes of this unusual complication of esophago-gastrostomy.

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ADDISON'S DISEASE WITH PSYCHOSIS

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SINCE Engel and Margolin[6, 7] in 1942 first clearly described the frequency of abnormal psychic states in Addison's disease, a rising interest in this association has been manifested. Prior to this, reports had been published from time to time, since Addison[1] first described the disease in 1849, of deviant mental states associated with Addison's disease, but not until 1942 was interest re-stimulated when it was suggested that emotional changes were closely related to physical crises. Engel and Margolin were impressed by the evidence of disturbed carbohydrate metabolism in Addison's disease and especially by the fact that hypoglycemic symptoms may develop at higher glucose levels than in normal persons. Further work has been carried out by Cleghorn,[2-5] who in 1951 obtained results strikingly similar to those reported by Engel and Margolin, with the exception that in his series of 25 cases he noted that these patients were under treatment with desoxycorticosterone acetate and salt most of the time, and that psychological deviations persisted despite such therapy. Smith[8] has recently reviewed the literature relative to this subject and concluded "the incidence of emotional disturbance in Addison's disease is high . . . [those] reaching psychotic proportions is much smaller and in most of these there is evidence of metabolic disturbance . . . [but] in some the relationship to biochemical change is not obvious." The following is a report of a patient with Addison's disease who was observed over a continuous two-year period at the Provincial Mental Hospital, Essondale, B.C.

E.L. was first admitted to the Provincial Mental Health Services on March 3, 1958, having been transferred from a general hospital after treatment for acute adrenal insufficiency. The diagnosis of Addison's disease was established in 1949 when he suddenly began to feel weak and lost the use of his legs. At that time he noticed that when he sat in the sun for a while, or in any warm place, it made him extremely weak. Also at that time he had no appetite and lost a great deal of weight, following which he began to vomit. Treatment with desoxycorticosterone acetate (DOCA) was then commenced. From that time until his crisis in February 1958 he required implantation of DOCA pellets every nine to 16 months. In February of 1958 he again went into addisonian crisis and was admitted to a general hospital in a semicomatose state. He was treated for his acute adrenal insufficiency, and as his body chemistry began to return to normal the patient became acutely psychotic. He stated that while in the general hospital he felt that he was interfering with hospital regulations in some way and kept hearing his own voice and other people's voices, especially voices of other members of his family. The voices told him to pull down the blinds, to keep the doors closed, and that his hair was standing on end. These things made his teeth chatter and he was very much afraid. He was particularly bothered by sounds and felt that people were deliberately making noises in order to annoy him.

This psychotic episode was brief in nature with the exception of a persistence of his auditory hallucinations,

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