Aortic-Esophageal Fistula: Attempted Surgical Repair*

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A ORTIC-ESOPHAGEAL FISTULA IS A catastrope due usually to impaction of a foreign body in the esophagus and subsequent perforation of the aorta. Surprisingly there is often no significant bleeding at the time of initial perforation and the fatal hemorrhage may be delayed for several days and rarely for years. As early as 1867, Adelman1 was able to collect 14 cases of perforation of the aorta among 314 instances of perforated esophagus, but it was not until 1914 that Chiari2 recognized and first described an aortic-esophageal syndrome. Classically the discomfort at the time of swallowing the foreign body is followed by an asymptomatic latent period. There is then reappearance of pain and a warning or "signal" hemorrhage which accurately forecasts the inevitable lethal episode. Often the interval between the signal hemorrhage and death from exsanguination is sufficiently long to allow operative intervention if the diagnosis has been made.

The sequence of events in this syndrome has not been appreciated in the past and diagnosis has been difficult. Since the disease has been accepted universally as fatal, attention has been centered chiefly on pathologic features. Thus, it is well known that most of the perforations occur in the region of the aortic arch, which is a common site of foreign body impaction due to the presence of the normal "aortic constriction." The gross pathology usually consists of a small cavity, a few centimeters in diameter, connecting with the aorta through a minute hole, a few millimeters in diameter, and with the esophagus through a larger fistula that may be a centimeter or two in length. The aortic fistula is sometimes filled with clot, and at the time of post-mortem examination there may be neither gross nor microscopic evidence of infection. It has been postulated, therefore, that pressure necrosis alone can cause the perforation. While this would seem probable when the foreign body is permitted to reside in the esophagus until the esophageal and aortic walls erode, hemorrhage occurring subsequent to foreign body removal suggests that a continuing localized necrotizing infection may be the determining factor in aortic fistulization.

In the following case, the site of bleeding was suspected preoperatively and thoracotomy was performed with the intent of arresting the hemorrhage. We believe that this is the first reported instance in which a direct operative attack on the source of bleeding has been attempted. The unsuccessful outcome suggests, if anything, an even more aggressive approach to this lethal, but potentially remediable condition.

CASE REPORT

A 34-year-old parking lot attendant was admitted to the Thoracic Surgery Service of Boston City Hospital on the evening of October 8, 1959, with a two-day history of severe crampy substernal pain. He stated he had been in excellent health until two days prior to admission when he experienced the sudden onset of substernal pain while eating chicken for his supper. The pain was intensified by further attempts to swallow the food, which "seemed stuck" in his chest, and it became almost intolerable when he swallowed more solid or fluid material. Although he felt nauseated, he did not vomit at any time. After a day and a half of unrelenting discomfort, he came to the Medical Clinic for help. A barium study on the afternoon of October 8 showed obstruction in the middle third of the esophagus. With this finding and the history suggestive of esophageal obstruction by a foreign

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body, he was admitted to the hospital for appropriate treatment.

Except for the fact that he had undergone complete dental extractions some years prior to admission, the patient's past history was non-contributory. He had always enjoyed good health and was not an alcoholic. He had worn both upper and lower dentures since shortly after the time of his extractions.

Physical examination on admission showed a well-developed, well-nourished, cooperative young man who was considerably distressed by severe substernal pain. The blood pressure was 140/70; the pulse was 84 per minute and regular; and the temperature was normal. There were no abnormal physical findings except for absent teeth.

Laboratory data obtained at the time of admission showed the hematocrit to be 47 percent and the white-cell count to be 13,700. Routine urinalysis was normal and a guaiac test of the stool for occult blood was negative.

On the evening of admission, esophagoscopy was attempted under topical anesthesia but because the patient was extremely agitated the esophagoscope could not be passed safely. Therefore, the procedure was postponed until the following morning when it was carried out under general anesthesia. At that time, two and a half days after the onset of pain, the upper esophagus beyond the cricopharyngeal muscle contained many fragments of unchanged chicken meat and carrot. When most of the food had been removed through the esophagoscope, a flat triangular bone, later identified as a chicken scapula, was found impacted 30 cm. below the level of the upper gum margin. The bone was removed by grasping its proximal angle with forceps and trailing it behind the esophagoscope as the latter was slowly withdrawn. The esophagoscope was then reinserted and the site where the bone had lodged was inspected. There was a short longitudinal mucosal rent in the left posterior aspect of the esophageal wall from which small amounts of serous fluid could be expressed by pressure on the surrounding area. Beyond this level the esophagus was normal as far as the stomach.

In order to minimize the danger of mediastinitis parenteral antibiotic therapy was instituted consisting of chloramphenicol and erythromycin, 2 grams each in divided daily doses, and no oral intake was permitted for two days. Upon recovering from anesthesia, the patient reported that the substernal pain was completely relieved. It did not return until the third day after esophagoscopy when he was allowed a fluid diet and then experienced two episodes of substernal pain. On the fourth day after esophagoscopy, a soft solid diet was begun and he experienced one episode of substernal pain in the morning. At 1:15 p.m. on this day, while lying quietly in bed, he suddenly vomited between 500 and 1000 mL of bright red blood. An infusion was started and he was immediately taken to the operating room where blood transfusion was administered and a portable chest x-ray film was secured. Meanwhile, his vital signs were stable and no further blood loss was apparent. The roentgenogram was unremarkable except for a small right pneumothorax. When esophagoscopy was carried out at 3 p.m. under general anesthesia, it revealed an 8 mm tear in the left posterior esophageal wall 30 cm. below the upper gum margin. As there was no active bleeding the tear was lightly packed with oxidized cellulose (Oxycel) gauze and the esophagoscope was removed. At 4 p.m. when he was reactive and had received 2 units of blood in transfusion he was transferred to the recovery room. His blood pressure on arrival was 100/70 and his pulse 95 per minute. At 4:10 p.m. he was "awake, restful, color flushed." At 4:20 p.m., three hours after the first hematemesis, he vomited an estimated 2000 mL of blood and went into shock. Without being moved from the recovery room he was intubated, transfused rapidly, and again esophagoscoped. There was now definite, continuous hemorrhage in the mid-esophageal region which was partially controlled by packing with many feet of 1-inch gauze roll. He was taken to the operating room where right thoracotomy was performed.

At operation, there was no evidence of bleeding in the right pleural space. When the mediastinal pleura was opened and the posterior aspect of the esophagus was inspected, a moderate-sized hematoma was found at the level of the azygos vein. As the esophagus was further mobilized and the exposure was improved by dividing the azygos vein, brisk bleeding occurred which seemed to stem either from the aorta or from a right intercostal artery where it crossed the vertebral column behind the esophagus. Many suture ligatures of 2-0 black silk were placed without obtaining more than temporary control of the bleeding; the angle between the posterior aspect of the esophagus and the vertebral column was firmly packed with 3 inch wide gauze roll on several occasions, but again without effective control of the bleeding. Finally, after he had received 39 units of blood in transfusion, a reasonable degree of hemostasis was obtained and the right chest was closed with a large pack and a chest tube in place. One hour postoperatively, he expired while in shock from continued blood loss.

Necropsy disclosed 1850 mL blood in the right pleural cavity. There was a 1.5 cm. longitudinal rent in the left posterior wall of the esophagus at the level of the carina (Fig. 1). At a corresponding level in the arch of the aorta, precisely midway between the first and second right intercostal ostia, there was a 1 mm. intramural defect

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filled with a fibrin plug which extended into the lumen of the aorta. It was possible to pass a probe directly through this defect into the esophageal rent. The stomach and entire small bowel contained clotted blood.

Histologic study of the aortic perforation demonstrated whorls of fibrin with inflammatory cells in the adventitial margins of the defect. These in turn were surrounded by proliferating adventitial connective tissue within which were occasional foci suggestive of bacterial inflammation. Microscopic examination of the esophageal rent showed that the mucosa was intact about its periphery, but was covered with fibrin. There was interstitial edema and a subacute inflammatory response manifested by proliferating connective tissue and a predominantly polymorphonuclear leukocytic infiltration.

Discussion

That the initial substernal discomfort and dysphagia were the first indications to the patient of having swallowed a foreign body is not surprising since he was wearing an upper denture at the time and thus was unable to feel the bone. Ingested foreign bodies occur more frequently in young children, the mentally disturbed, and those adults who wear dentures or prosthetic palates. It has been suggested that one method of prevention would be the use of fenestrated dentures to permit the contents of the mouth to contact the sensitive palate and thereby allow detection of intra-oral foreign bodies.

The four-day interval following the initial discomfort was relatively asymptomatic and corresponded to Chiari's latent period, i.e., the time between the initial discomfort and the onset of bleeding. During this period, the impacted foreign body was demonstrated by a chest x-ray film and barium swallow, and endoscopic removal was accomplished. For two days after esophagoscopy, however, he pursued a moderately febrile course. Since the foreign body had been removed and since there had been no bleeding, it is difficult to establish accurately the time of formation of the aortoesophageal fistula. A possible esophageal perforation was noted when the foreign body was removed. The precise role of infection and pressure necrosis has never been defined clearly, although it is likely that they act in concert. While sharp bones, pins, wires, and needles have been the chief offenders, coins, dentures, and other blunt objects have occasionally been responsible. It has been suggested that pulsations of the

![Figure 1: Necropsy specimen showing longitudinally opened esophagus with the rent clearly evident in the mid-portion of the specimen. The proximal mucosa is blood-stained due to submucosal infiltration of blood. To the right is the carina and to the left the opened aorta. In the upper left portion of the photograph is seen part of the extensive clot. The offending foreign body, previously removed endoscopically, has been placed in its assumed original position. (Photograph courtesy Dr. E. A. Gaensler).](image-url)
aorta and contractions of the esophagus facilitate penetration of the aortic wall, and that the latent period may be pro-
longed when a blunt object is involved because a longer interval is required for erosion. Nevertheless, the latent period in
some of the cases involving blunt objects has been relatively short and in some of
the cases involving sharp objects, relatively long. An occasional patient with aortic fistulization has died of sepsis and not of hemorrhage. In some cases mediastinal infection has resulted in aortic perforation in the absence of a foreign body. Henry and Miscall have recently observed an aortic perforation which did not bleed at the time of thoracotomy for removal of the arrested foreign body, but which subsequently resulted in fatal hemorrhage. Therefore, it seems highly probable that penetration of the aorta and sealing may occur early and that during the latent period clotting may effect temporary occlusion of the aortic perforation. This is substantiated by the occasional discovery of clot within the aortic fistula at the time of postmortem examination. Disturbance or dissolution of this clot by infection in the mediastinum, or by intra-aortic pressure, or by both, may precipitate bleeding (the “signal” hemorrhage) and pain. Resealing of the aortic fistula may be responsible for intermittent sublethal hemorrhage which has been known to continue for as long as four years. The situation is somewhat analogous to the type of bleeding seen with infected arterial grafts or with progressive infections involving arteries.

From the outset, our attention in this case was focused on the foreign body in the esophagus and with the first emesis of bright red blood, involvement of the aorta or other large artery was suspected. However, in the absence of a clear history of foreign body ingestion, the appearance of dark red blood or tarry stools may be misleading. In one reported case, an unrevealing laparotomy was performed and in others, extensive radiologic and laboratory studies were employed in a vain effort to determine the site of bleeding. In our case, approximately two hours elapsed between the signal hemorrhage and the final bleeding episode. During this time a second esophagoscopy revealed a rent in the posterior wall of the esophagus. The fact that the original bleeding had ceased spontaneously led to the erroneous conclusion that the aorta could not have been the source. When hemorrhage recurred massively, however, it was thought to originate either from the aorta or from one of the intercostal vessels arching anterior to the vertebral column. Operative approach via a right thoracotomy was selected in order to obtain optimum exposure of the esophagus and because it was suggested by the pneumothorax on this side. The bleeding appeared to originate from the area between the esophagus and vertebral column, but because of its massive nature it was not possible to locate its exact source. Packing the area with gauze was a hopeful but futile gesture.

In the event exploratory thoracotomy is performed after the “signal” hemorrhage and discloses no active bleeding, it would seem logical to interpose soft tissues between aorta and esophagus, as well as to foster transpleural drainage of the area of possible infection.

A retrospective analysis of the management directed to our patient suggests several areas of possible improvement. Since there is little doubt that any lengthening of the period of residence of such a foreign body increases the chance of aortic fistulization, it was unwise to postpone endoscopic intervention overnight when topical anesthetization proved unsatisfactory. Actually, there are many who believe that endoscopic procedures done for this purpose are preferably accomplished with the aid of general anesthesia. Visualization of the mucosa at the site of impaction after removal of the offending bone, complete restriction of oral intake, and the prophylactic use of broad-spectrum antibiotics appeared indicated and were carried out. Following endoscopic manipulation, a care-
ful roentgenologic study of the site of impaction by means of a water-soluble contrast medium other than barium may have disclosed a small abscess cavity and thus served as a warning of impending disaster.

In the present case, the timing of the surgical intervention requires comment, since the optimum time was immediately after the "signal" hemorrhage. Prompt esophagoscopy showed absence of active bleeding from any site in the esophagus, however, and engendered a false sense of security. With the subsequent onset of massive hemorrhage, esophagoscopic packing of the entire lumen was performed and this proved an effective temporizing measure. An error in emphasis was responsible for selection of the right-sided operative approach. Since the source of hemorrhage was not the esophagus, but the aorta a left-sided thoracotomy should have been undertaken and would have afforded better exposure. Furthermore, once the selected approach proved inadequate in this desperate situation, full visualization of the thoracic aorta should have been attempted by extending the posterolateral incision transternally or by employing a separate longitudinal sternum-splitting incision.

The ideal technic for repair of the aortic fistula must remain speculative since the unfortunate outcome in our hands lends little authority to any statement. Certainly suture-ligatures placed without benefit of clear visualization, as here, offer little hope of arresting hemorrhage. Furthermore, because the aortic wall in the vicinity of the fistula may have been weakened by infection or pressure necrosis, sutures placed in this area may invite further difficulties. However, on theoretic grounds a free graft of muscle applied to the site of hemorrhage and maintained in position by sutures placed in the aorta at a distance from the fistula might prove effective. Since the actual aortic fistula may be quite small, tamponade by means of gauze or other material suggests itself, but the likelihood of success is undoubtedly slim. Hypothermia or cardiopulmonary bypass would undoubtedly facilitate surgical management of the lesion, but the emergent nature of the problem probably would preclude their use.

REFERENCES