Surgical Treatment of Intractable Cutaneous Fistula with Osteomyelitis in the Neck Developed after Esophagectomy

TAKESHI SHIMAKAWA1, YOSHIHIKO NARITAKA1, SHINICHI ASAKA1, NORIYUKI ISOHATA1, MINORU MURAYAMA1, SOICHI KONNO1, TAKAO KATSUBE1, KENJI OGAWA1 and HIROKO IDE2

1Department of Surgery, Tokyo Women’s Medical University Medical Center East, 2-1-10, Nishiogu, Arakawa-ku, Tokyo;
2Hamacho Center Building Clinic, 2-13, Hamacho, Chuo-ku, Tokyo, Japan

Abstract. A case of intractable cutaneous fistula in an esophagogastrostomized region complicated by osteomyelitis is reported. A three-stage operation was performed, and gratifying results were obtained. The patient was a 74-year-old man who received a subtotal esophagectomy and a cervical esophagogastrostomy through a retrosternal approach for advanced esophageal cancer. Following subsequent radiotherapy, the anastomosed region in the neck was found to have been stenosed. Endoscopic balloon dilatation was then performed and perforation of the stenosed region occurred to form an intractable fistula. A chest computed tomography scan revealed osteolysis of the sternum, clavicular head and left first rib. Treatment comprised an initial control of the infective foci including osteomyelitis and, after achieving stabilization of the wounds, the subsequent step of reconstruction. The patient’s postoperative course was satisfactory without involvement of any leakage or stenosis of the anastomosed regions, or wound infection. It is considered feasible to cure even a cutaneous fistula in the neck complicated by osteomyelitis, as in the present case, by sufficient control of infection and procedural contrivance.

Using conservative treatment, it is extremely difficult to cure an intractable cutaneous fistula in the neck that has developed after esophageal cancer resection (1-4). We encountered a patient with intractable cutaneous fistula developed in a esophagogastrostomized region complicated by osteomyelitis. The fistula was treated by a three-stage operation comprising control of infective foci, reconstruction by free jejunal interposition and wound covering with a musculocutaneous pedicle flap, with gratifying results.

Case Report

The patient was a 74-year-old man with chief complaints of salivary defluxion from a cervical fistula and precordial chest pain. In September 1997, for the treatment of Mt type 3 esophageal cancer, a subtotal esophagectomy through a right thoracoabdominal approach and a cervical esophagogastrostomy via a retrosternal approach were performed at another hospital. A histopathological diagnosis of well-differentiated squamous cell carcinoma (pT2, pN0, M0) was made. In April 2000, cervical lymph node metastasis was noted and the patient received radiotherapy, to which there was a complete response. Subsequently, as the anastomosed region in the neck was noted to have stenosed, an endoscopic balloon dilatation was performed. In May 2005, the anastomosed region in the neck perforated and formed an intractable fistula; therefore, the patient was seen at our department upon referral from the other hospital.

On admission, a cutaneous fistula was noted in the esophagogastrostomized region in the neck, with an extensive redness of the skin area surrounding the fistula. A gastrostomy tube had been placed in the upper abdomen for enteral hyperalimentation, and granulation tissues surrounded the tube. Hematology revealed no adverse findings except abnormality in glucose tolerance.

An esophagogram was taken through the balloon catheter placed in the cervical cutaneous fistula; the results revealed complete stenosis at the level of the upper sternal margin and dilatation of the esophagus rostral to that lesion. Retrograde esophagograms obtained via the gastrostomy tube failed to show evidence of passage of the contrast medium through the stenosed lumen (Figure 1). An esophagoscopy revealed a fistula at a site 20 cm from the anterior dentition and there was a marked stenosis at the anal side of the esophagus. A chest computed tomography scan showed osteolysis of the sternum, clavicular head and left first rib, and an abscess in the left anterior chest (Figure 2).
The patient was thus diagnosed with an intractable cutaneous fistula in the esophagogastrostomized region, which had developed after radiotherapy following esophagectomy, complicated by extensive osteomyelitis. The patient had been maintained on enteral hyperalimentation at the previous hospital and he was relatively well nourished. However, surgical treatment was indicated for the patient’s condition because of the concurrent osteomyelitis making the fistula extremely intractable and conservative treatment infeasible to cure the fistula. As a single-stage operation was considered impracticable because of the presence of osteomyelitis in the sternum, clavicular head and rib, the first step comprised control of the infective foci and, after achieving stabilization of the wounds, the subsequent step was reconstruction by free jejunal interposition.

In the first operation, conducted in November 2005, an external esophagostomy was performed along with curettage of the granulation tissues of the cervical to precordial perifistular regions. Granulation tissues surrounding the upper abdominal gastrostomy tube were also curetted, followed by reinsertion of a gastrostomy tube for decompression and an enterostomy tube for enteral hyperalimentation (Figure 3). In the second operation, performed in March 2006, the esophagagastrotomized region was resected together with the fistula because the previously curetted anterior chest osteomyelitis had not improved; the sternum, rib and clavicular head were partially resected, followed by filling the bone stumps with an antibiotic-containing cement. The tissue defect resulting from the resection was filled with a pectoralis major muscle musculocutaneous pedicle flap (Figure 4). The third operation carried out in May 2006, consisted of alimentary tract reconstruction with the free jejunum interposed between the cervical esophagus and the stomach, and by covering the wound with a pectoralis major muscle musculocutaneous pedicle flap (Figure 5). No leakage or stenosis of the anastomosed regions nor wound infection was noted postoperatively and the results of postoperative esophagogram were gratifying (Figure 6).

The patient gradually became capable of oral intake and was discharged. There has been no recurrence of osteomyelitis and the food ingestion status is satisfactory in the patient, who is currently treated on an outpatient basis.

Discussion

Cutaneous fistulation after esophageal cancer resection has been reported to occur due to leakage in the anastomosed region and perforation of an ulcer in the reconstructed gastric tract (1-4). A simple cutaneous fistula due to leakage in the anastomosed region may often be treated with conservative treatment (5). Nevertheless, the cause of fistulation was uncommon in our patient in that perforation occurred at a site of stenosis of the anastomosed region that had developed after radiotherapy. Histopathological examination of the resected fistula specimen revealed fibrosis in the gastric tube, causing a stenosis which eventually became perforated and led to the fistulation. Generally, gastric tube tissue displays circulatory disturbance in the gastric mucosa after radical radiotherapy, resulting in depression of mucosal protective function and repair function (6). This may account for the intractability and refractoriness to conservative therapy of the fistula in our case.

Organs that may become fistulated in such a situation would include the skin, chest wall, aorta, heart, trachea (bronchus) and mediastinum, depending on the reconstruction route of the gastric tube. As cases of fistulation with the aorta, heart or trachea (bronchus) may constitute a fatal complication, appropriate treatment should be instituted as soon as possible (7-9). On the other hand, fistulations in the skin, chest wall and mediastinum are not immediately life-threatening but may involve an intercurrent osteomyelitis if inflammation spreads to a
severed or resected bone during operation. In particular, reconstruction through a retrosternal approach is prone to involve an intercurrent osteomyelitis, as bone resection is often required due to the narrow space behind the sternoclavicular joint. Once developed, osteomyelitis is intractable and may result in a fatal complication if it advances. Cases reported in which such extensive osteomyelitis was cured as in the present case have been extremely few in number; an extensive search of the related literature has revealed only 2 cases in Japan (1, 2). Yet there might have been a number of cases where lives could not be saved due to advanced osteomyelitis for the reason mentioned above and hence it is important to perform early exploration of surgical treatments in such cases.

Essentials of the treatment for intractable osteomyelitis consist in sufficient curettement of infective foci, and filling

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**Figure 2.** The chest computed tomography scan showed the balloon catheter placed in the cervical cutaneous fistula (a) and osteolysis of the sternum, clavicular head and left first rib, and abscess formation (b).

**Figure 3.** The first operation: A preoperative finding showed the cutaneous fistula in the esophagogastrotomized region of the neck, with an extensive redness of the skin area (a). An external esophagostomy was performed along with curettage of the granulation tissues of the cervical to precordial perifistular regions (b).
and reconstruction of the tissue defects resultant from the curettage. A pectoralis major muscle musculocutaneous pedicle flaps have widely been used for filling tissue defects in the face, neck and precordial region since being described by Ariyan (10) in 1978. This procedure represents a musculocutaneous flap method whereby a tissue defect is repaired in one stage by shifting the pectoralis major muscle nourished via the thoracoacromial artery and vein with islands of covering skin remaining on the muscle. In the present case, deranged granulation tissues were curetted in the initial operation and a full curettment of the regions of osteomyelitis was carried out by a fistulotomy and an

Figure 4. The second operation: The esophagogastrostomized region was resected together with the fistula, and the sternum, rib and clavicular head were partially resected (a). The tissue defect resulting from the resection was filled with a pectoralis major muscle musculocutaneous pedicle flap (b).

Figure 5. The third operation: Reconstruction with the free jejunum interposed between the cervical esophagus and the stomach was performed (a). The tissue defect resulting from the resection was filled with a pectoralis major muscle musculocutaneous pedicle flap (b).
osteotomy in the subsequent operation. The bone stumps were filled with an antibiotic-containing cement and the tissue defect that resulted from the resections was filled with a pectoralis major muscle musculocutaneous pedicle flap. By these two operation steps, a cure and the prevention of recurrence of osteomyelitis were accomplished in our patient.

Reconstruction by free jejunum interposition has commonly been performed for reconstructing the defective laryngopharynx and cervical esophagus (11-14). This is reportedly based on the following rationales: this portion of the intestine is cleaner and involves less leakage than the lower portions of the intestine; its vascular pedicle is longer and diameter larger; its caliber is closer to that of the esophageal stump; and this procedure is associated with lesser post-enterectomy impairment and sacrifice. However, this procedure involves vascular anastomoses; hence, a sufficient control of infection over the surgical field is imperative for its indication. In the present case, therefore, we conducted the third operation, comprising reconstruction by free jejunum interposition and covering of the wound using a pectoralis major muscle musculocutaneous pedicle flap, after a sufficient control of infection over the surgical field was achieved in the preceding two steps. There has been no postoperative problem concerning the vascular anastomoses, nor leakage in any new anastomosis in this case, possibly as a result of the surgical operation being performed in these three steps.

The present case showed that it is considered feasible to cure even a highly intractable cutaneous fistula developed in a cervical esophagogastrectomized region accompanied with osteomyelitis by prudent measures (a three-step operation) for sufficient control of infection and procedural contrivance such as free jejunum interposition and wound covering with a pectoralis major muscle musculocutaneous pedicle flap.

References


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