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## CASE REPORT

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# CLOSURE OF TRACHEOESOPHAGEAL FISTULA WITH PREFABRICATED REVASCULARIZED BILAMINAR RADIAL FOREARM FREE FLAP

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**Abstract:** *Background.* Tracheoesophageal fistula (TEF) is a rare but serious complication associated with high mortality rates. Traditional management of TEF includes primary closure with or without interposition of regional tissue flaps but is associated with a significant recurrence risk, especially in case of larger fistulas. Application of microvascular free flap reconstruction is an emerging alternative in the surgical management of large TEFs, but may be limited by issues of flap bulkiness and requirement for neoeptelialization across the large inner flap surface.

*Methods and Results.* Here, we report prefabrication of a bilaminar radial forearm free flap to successfully close a large recurrent TEF that occurred years after tracheoesophageal puncture-based voice rehabilitation in a laryngectomized patient.

*Conclusion.* The bilaminar radial forearm free flap may prove to be an important adjunct to the closure of large TEFs.

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**T**racheoesophageal fistula (TEF) is a rare but life-threatening condition due to chronic aspiration, pneumonia, and tracheal stenosis.<sup>1</sup> The overall incidence of TEF is 0.05%, but may be as high as 3% in mechanically ventilated patients.<sup>2</sup> Besides ventilatory cuff injury, additional causes of acquired TEF include local infection, caustic or foreign body ingestion, penetrating or nonpenetrating trauma, tracheal or esophageal malignancy, and iatrogenic surgical injury. An example of the latter includes iatrogenic fistula after surgical prosthetic voice rehabilitation in laryngectomized patients. Moreover, an important proportion of TEF occurs in a congenital fashion in which it is typically associated with esophageal atresia.<sup>3</sup> Regardless of the etiologic background, TEF mortality is high (20–30%) and dictates surgical intervention in the overwhelming majority of viable cases. Although various surgical approaches have been described, successful TEF closure remains an item of concern with a high proportion of recurrence.<sup>4</sup>

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Traditional surgical closure of TEF includes fistula tract division and 3-layer closure of esophageal and tracheal linings with or without interposition of a pedicled regional skin or muscle flap.<sup>5-7</sup> This approach is successful in the majority of small uncomplicated TEFs but has been disappointing in case of larger fistulas with or without circumferential tracheal damage.<sup>4,8</sup> The addition of segmental tracheal resection and anastomotic reconstruction may successfully manage a proportion of the latter but is not possible in all patients.<sup>4,8</sup> Overall, TEF recurrence occurs in 10% of cases, and besides, large fistula size has been associated with a number of variables, including ongoing need for ventilatory support, prior irradiation, diabetes, steroid use, immunocompromised status, malnutrition, and local infection.<sup>4,9</sup> Depending on the presence or absence of these factors, application of extracervical tissue transfer approaches such as pedicled pectoral major flap or microvascular free flap reconstruction is an emerging alternative to the closure of large (recurrent) TEFs but is limited by issues of flap bulkiness and/or the requirement of neoepithelialization across a large surface.<sup>10-12</sup>

Here, we report the successful closure of a large, recurrent TEF with a radial forearm free flap (RFFF), prefabricated to a bilaminar flap with the use of a split skin grafting.

#### CASE REPORT

A 52-year-old man from Germany and with a persistent tracheoesophageal fistula was referred to the Department of Head and Neck Surgery and Oncology of the Netherlands Cancer Institute. The patient had been diagnosed with a T4N0M0 glottic squamous cell carcinoma 5 years earlier for which he was treated with total laryngectomy, bilateral modified radical neck dissection, and postoperative intensity-modulated radiation therapy (70 Gy). At the time of surgery, voice rehabilitation was achieved by tracheoesophageal puncture and implantation of a Provox voice prosthesis.<sup>13</sup> Over the course of the postoperative years, the patient experienced fistula widening with persistent leakage around the prosthesis, which was only temporarily relieved by prosthesis removal and augmentation of fistular margins with injected abdominal fat. The patient underwent repeated ( $n = 6$ ) surgical interventions including primary closure in 3 layers with and without interposition of local skin and muscle flaps. A final intervention prior to referral included separation

of esophagus and airway, with cleavage of the fistular tract and inverted suture of local skin flaps into the esophageal and airway defects after interposition of a sternocleidomastoid muscle flap. Fistula recurrence occurred partly as a result of mechanical damage by a persistent nasal feeding tube on 1 side (the patient refused a percutaneous endoscopic gastrostomy [PEG]) and a rigid stomal canula on the other. At presentation to The Netherlands Cancer Institute, the nasal feeding tube had been replaced by a PEG and the remaining defect measured 2 by 4 cm (Figure 1). At this point, the patient had been voiceless for more than 2 years.

To close the defect, an RFFF was prefabricated on the left arm. Routine preoperative application of Allens test and duplex sonography indicated sufficient perfusion of the ulnar artery and palmar arch on both sides. The cephalic vein, radial artery, and concomitant vein were identified and freed from their surroundings. The vascular pedicle and skin island were raised. In the same setting, a 0.5-mm thick split skin graft was taken from the left upper leg. The split skin graft was sutured top-down to the inner soft tissue lining of the RFFF using dissolvable sutures. After primary closure, the prefabricated flap was left in situ for 2 weeks.

In a second procedure, an incision was made in the mucocutaneous junction of the tracheal stoma from the 3-o'clock position to the 9-o'clock position. The posterior wall of the trachea was undermined inferiorly and separated from the esophageal wall down to and beyond the fistula. After wide excision of the TEF tract, the transverse cervical artery and internal jugular vein were identified on the left side of the neck. As the internal jugular vein appeared heavily fibrotic and obliterated cranially, focus was directed to the right side. Here, the internal jugular vein appeared intact but the transverse cervical artery was absent.

Subsequently, the prefabricated RFFF with the well-healed skin graft was raised and transferred to the defect (Figures 2A and 2B). The donor site was closed with a split skin graft of the opposite thigh. The full thickness skin side of the RFFF was sutured to the esophageal mucosa and neopharynx. Subsequently, the tracheal mucosa was sutured around the split thickness side of the RFFF skin island (Figure 3).

Based on both the absence of viable internal jugular vein on the left side and absence of transverse cervical artery on the right side, the vascular pedicle was longitudinally split to allow anas-



**FIGURE 1.** Preoperative image of tracheoesophageal fistula defect measuring 2 by 4 cm.

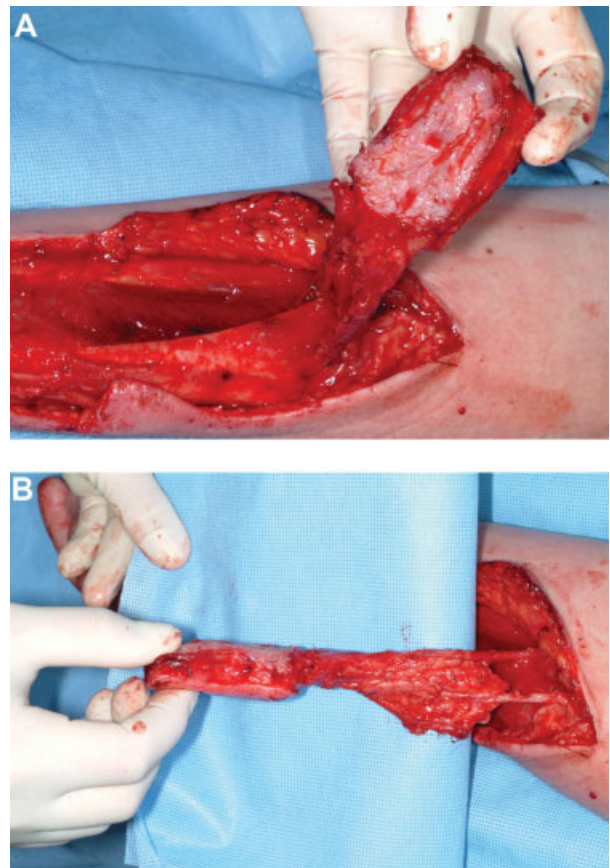
tomosis of the radial artery to the left transverse cervical artery and the cephalic vein to the right internal jugular vein. Postoperatively, the flap remained vital and the defect closed, except for a pinhole fistula at the caudal border. Here, a small round fistula remained, measuring 4 mm in diameter. This fistula was used for Provox prosthesis implantation (Figure 4). Although positioned quite low in the tracheal tract, the prosthesis was easy to reach for regular replacement and cleaning. With a follow-up of 1 year, the prosthesis has remained in situ without fistula widening. The patient readily developed an excellent voice and fluent speech and tolerated a normal oral diet.

#### DISCUSSION

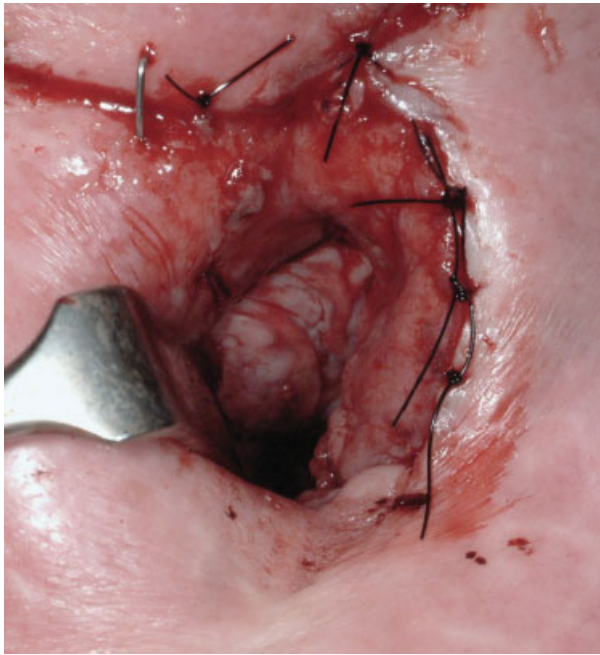
The introduction of voice rehabilitation by tracheoesophageal puncture and voice prosthesis insertion has demonstrated tremendous improvement of speech in laryngectomized patients.<sup>14</sup> Prosthetic voice rehabilitation results in self-reported fair-to-excellent voice quality in 88% of laryngectomized patients.<sup>15</sup> However, a number of patients (20%) experience gradual fistular widening with associated chronic aspiration. Fistular shrinkage may be achieved by downsizing the

prosthesis or by temporary prosthesis removal and nasogastric feeding tube placement in the majority of patients. Alternatively, conservative management may include placement of submucosal purse string sutures around the fistular tract, or marginal augmentation with collagen or abdominal fat injections.<sup>16–18</sup> Despite these measures, fistular widening is progressive, and development of a pathologic TEF ensues in a small number of patients.<sup>17</sup> These patients require surgical closure at the (temporary) expense of their voice.<sup>18</sup> Several studies have demonstrated that primary 3-layer closure of tracheoesophageal puncture sites (with or without interposition of regional muscle/skin grafts) is associated with low recurrence rates and allows for delayed prosthesis replacement in the majority of cases.<sup>5,7,17</sup>

This report describes an extreme case of tracheoesophageal puncture site widening, which resulted in a large pathologic TEF that was persistent despite repeated attempts at conventional



**FIGURE 2.** (A) and (B): Images of prefabricated bilaminar radial forearm free flap with vital, well-healed split skin graft on the inner surface after 3 weeks of incubation.



**FIGURE 3.** Direct postoperative image of bilaminar radial forearm free flap in situ.

closure. Reasons for the recurrent surgical failure remain unknown, but may be related to a combination of prior irradiation, scarring, and mechanical compromise of surgical wound healing between the indwelling nasogastric feeding tube and a rigid stomal canula. Regardless of its causes, the repeated failure of conventional closure efforts, the large size of the TEF, and the impossibility of segmental tracheal resection in this laryngectomized patient led us to explore alternative avenues of surgical reconstruction. The application of pedicled regional flaps or microvascular free flaps to the reconstruction of enlarged tracheoesophageal puncture sites is well-documented but remains largely anecdotal.<sup>10–12,19–21</sup> Although several flaps have been described including the pedicled deltopectoral and pectoralis major flap and the revascularized rectus abdominis free flap, potential issues of flap bulkiness led to the selection of the radial forearm flap in the present case. Advantages of the radial forearm flap in this setting with anticipated dense scarring after multiple surgical failures and a previously irradiated surgical field include its thin and pliable nature, dense vascularization, and long vascular pedicle, besides our routine use of and favorable experience with the RFFF in oromandibular reconstruction. We chose to prefabricate the RFFF with a split-skin graft in an attempt to

obtain 2-sided epithelial coverage of the large tracheoesophageal defect and circumvent possible postoperative issues of reepithelialization compromise. Although the procedure was technically challenging by a vascular-anatomical anomaly and a pinhole fistula opening remained at the transoperative difficult to reach inferior border of the defect, these issues were successfully managed by longitudinal splitting of the vascular pedicle and prosthetic re-implantation respectively. The success of the procedure is best illustrated by the vitality of the flap, closure of the defect, and excellent voice quality of the patient at 1 year of follow-up.

This report is the first of its kind describing the potential value of a prefabricated bilaminar RFFF in the surgical management of TEF. Previously, Wein et al<sup>20</sup> and Delaere et al<sup>11,22</sup> reported successful reconstruction of recurrent TEF after failed conventional closure with a RFFF. However, the described TEFs were small, and the flaps did not require bilaminar prefabrication. Bilaminar prefabrication of a RFFF with a split skin graft was described previously in the reconstruction of large orocutaneous defects after ablative head and neck surgery.<sup>23</sup> Also, Hallock<sup>24</sup> described successful reconstruction of an untreated adult case



**FIGURE 4.** Late postoperative image of covered tracheoesophageal fistula with Provox voice prosthesis inserted within caudal margin.

of cleft palate with a bilaminar RFFF prefabricated with a skin graft. Altogether, the available data suggest that the versatility of the RFFF can be significantly improved by prefabrication with a split skin graft without compromising thickness and pliability. This approach is attractive when the defect to be covered involves 2 large opposing epithelial surfaces as illustrated by the present case. The true viability of the approach described herein will depend on confirmation in larger studies.

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