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CASE REPORT
Craniofacial/Pediatric

Staged Intracranial Free Tissue Transfer and Cranioplasty for a Refractory Nasal-cranial Base Fistula

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Summary: After a transnasal endoscopic resection of a high-grade adenoid cystic carcinoma that underwent adjuvant chemoradiation, there was delayed recurrence managed by en bloc resection through an open craniofacial approach. Subsequently, the patient developed a chronic nasocranial fistula with secondary infection and bone flap resorption. This resulted in infectious episodes with secondary scalp incisional dehiscence and hardware exposure which required multiple bone debridement procedures, hardware removal, prolonged IV antibiotics, and hyperbaric oxygen treatment. The nasocranial fistula and chronic frontal bone osteomyelitis persisted despite the previous interventions. The patient underwent a frontal bone removal and obliteration of the anterior cranial base fistula with a free vastus lateralis muscle flap. At 4 weeks postoperatively, the intranasal portion of the muscle flap had completely mucosalized. After a 6-week course of IV antibiotics, a secondary cranioplasty using a custom-made poly-ether-ether-ketone implant was performed. The patient remained disease- and infection-free for the duration of follow-up (17 months). *(Plast Reconstr Surg Glob Open 2023; 11:e5392; doi: 10.1097/GOX.0000000000005392; Published online 9 November 2023.)*

Nasocranial fistulae in the setting of prior radiation and multiple operations represent a unique treatment challenge.[1,2] Reconstruction aims to separate the intracranial contents from the extracranial contents.[3] Radiation delivered to the tumor bed limits the ability to use local and regional flaps for reconstruction.[4] In these situations, free tissue transfer is considered the gold standard in the reconstruction of large skull base defects in irradiated fields.[5]

We describe a staged, multidisciplinary approach for the treatment of a refractory nasocranial fistula in the anterior skull base after prior skull base irradiation and repeat resection of an adenoid cystic carcinoma.

ILLUSTRATIVE CASE

The patient is a 42-year-old woman who was diagnosed with high-grade adenoid cystic carcinoma. Initial imaging demonstrated a heterogeneous mass in the left nasal vault that crossed midline through an eroded nasal septum and left maxillary sinus. She underwent transnasal endoscopic resection of the tumor with trilayer fascia lata duraplasty. Pathologic evaluation showed that perineural infiltration and the margins of one olfactory nerve were positive. She underwent adjuvant proton-beam radiation with a total dose of 6000 cGy in 30 fractions and weekly chemotherapy with cisplatin. The timeline of the surgical interventions is summarized in Table 1.

A focal tumor recurrence was identified on surveillance imaging three and a half years after the index surgery. The patient underwent repeat resection through a bifrontal craniotomy and duraplasty with fascia lata graft with a pericranial flap.

Three months after her bifrontal craniotomy, she developed a nonhealing wound at the apex of her bicoronal incision that required two surgical debridements and subsequent hardware removal. She continued to have progressive resorption of the frontal bone and a persistent open wound at the apex of the craniotomy incision that did not respond to suppressive antibiotics and hyperbaric oxygen therapy.

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Table 1. Timeline of Surgical Interventions from August 2016 to March 2022

Date	Procedure
August 2016	Transnasal endoscopic tumor resection, trilayer fascia lata duraplasty with postoperative proton-beam radiation
August 2020	Open resection of recurrent tumor, trilayeral fascia lata duraplasty with pericranial flap
November 2020	Irrigation and debridement of scalp wound with primary closure
December 2020	Irrigation and debridement of scalp wound, hardware removal, primary closure
January 2022	Hardware removal, right free vastus lateralis flap
March 2022	Cranioplasty with custom PEEK implant

Repeat imaging demonstrated disintegration of the outer table of the frontal bone flap with evidence of fistulous communication between the nasal cavity and site of the open wound. Due to the aggressive nature of adenoid cystic carcinoma, a major reconstruction was delayed until this point in time to ensure that there was no early tumor recurrence. A decision was made to approach the problem in a staged fashion.

FIRST STAGE

The primary purpose of the first stage was to remove all remaining hardware and nonviable resorbed frontal craniotomy flap and to separate the intracranial cavity from the nasal cavity. After removal of the resorbed frontal bone flap, an anterior cranial base fistula measuring 1.5 by 2.0 cm was identified. (**See figure 1, Supplemental Digital Content 1,** which displays a photograph showing the nasocranial fistula measuring 1 × 2 cm after removal of the frontal bone flap. **http://links.lww.com/PRSGO/C851**.)

A free vastus lateralis muscle flap was used to separate the anterior cranial base from the extracranial cavity and obliterate the dead space (Fig. 1) with anastomosis to the left superficial temporal vessels. A postoperative CT scan demonstrated successful obliteration of the nasocranial fistula. Intraoperative cultures revealed polymicrobial growth. Broad-spectrum antibiotics were continued for 6 weeks. At 4 weeks postoperatively, transnasal endoscopy revealed complete mucosalization of the intranasal portion of the muscle flap. Immediate skull reconstruction was not performed because there would likely be a need for long term antibiotic suppression. Furthermore, with free tissue transfer, there is often postoperative edema that limits skull reconstruction. The staged approach with short-term skull reconstruction as a second stage was designed to minimize long-term antibiotic use and allow immediate flap swelling for a better cosmetic result.

SECOND STAGE

After ensuring an infection-free interval of 8 weeks, the patient underwent delayed cranioplasty using a custom-made poly-ether-ether-ketone (PEEK) implant to cover a frontal bone defect, taking care to include a 1 cm^2 gap, which allowed passage of the extracranial pedicle to the intracranial muscle flap (Fig. 2). There were no complications of cerebrospinal fluid leakage, flap loss, or infection in the immediate postoperative period.

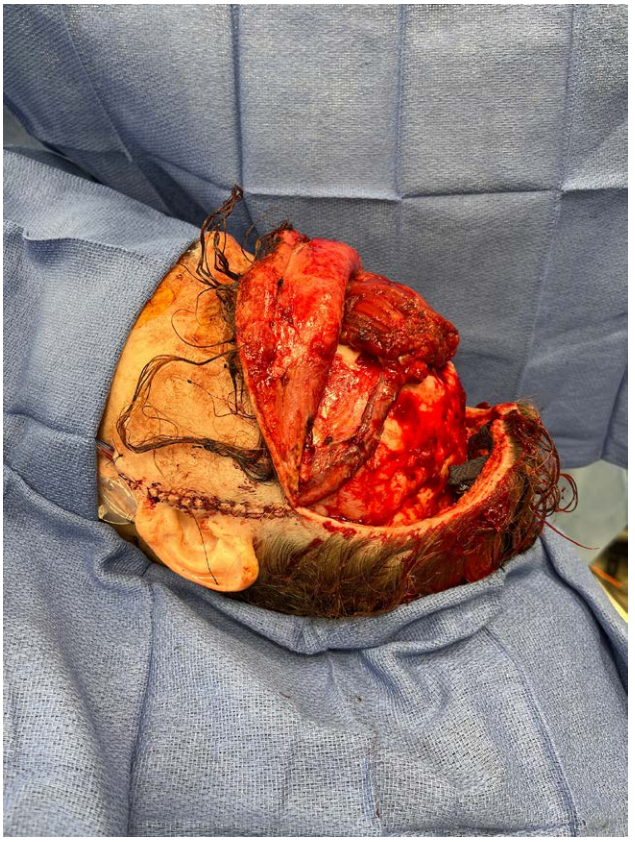

Fig. 1. A photograph showing the free vastus lateralis flap with microvascular anastomosis to the superficial temporal vessels. The flap is secured to the surrounding bone and inset in a way that ensures complete coverage of the anterior cranial base fistula.

The patient had no evidence of clinical of radiographic evidence of infection or tumor recurrence during the follow-up duration and reports good functional and aesthetic outcomes (Fig. 3). (**See figure 2, Supplemental Digital Content 2,** which displays the most recent magnetic resonance imaging demonstrating the PEEK cranioplasty with complete obliteration of the dead space and no evidence of osteomyelitis or tumor recurrence. **http://links.lww.com/PRSGO/C852**.)

DISCUSSION

Conventionally, the cranioplasty in a previously infected surgical bed is delayed for 1 year or more.

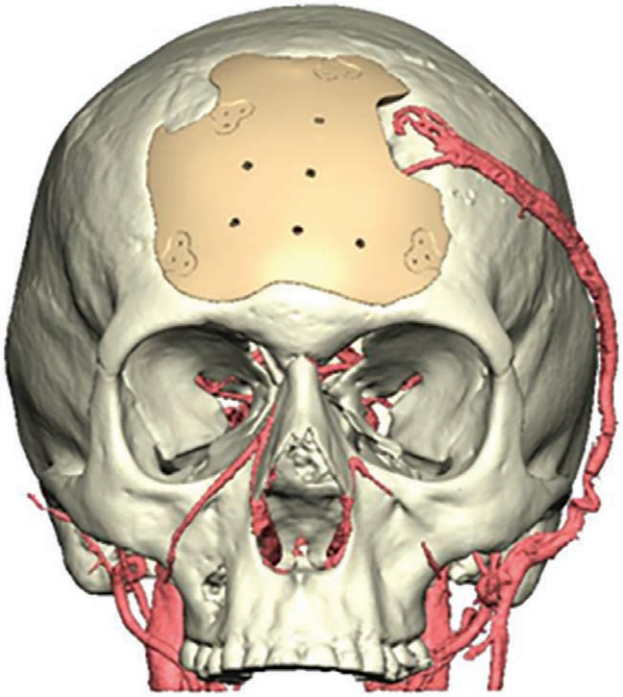

Fig. 2. Custom PEEK implant with a 1 square cm gap for passage of the extracranial pedicle to the intracranial muscle flap.

However, prolonged loss of protection to the brain, neurological deterioration in trephined syndrome, and socioeconomic burden of an altered appearance has led to attempts to shorten these intervals to a matter of weeks.[6] Immediate alloplastic reconstruction in the context of infection has a higher complication rate.[7] Kwiecien et al analyzed the use of alloplastic material in the setting of prior skull bone osteomyelitis and noted that the risk of reinfection decreases by approximately 10% with each month of delay, reaching the level similar to that of a cranioplasty in a noninfected patient after 12 months.[7]

In this patient, we performed a delayed, alloplastic cranioplasty in a much shorter period (2 months) than what has been described in other studies.[7] There are many reports of delayed calvarial reconstruction after free tissue transfer; however, each of these cases delayed the second stage for at least 5–12 months.[8–10] Additionally, there have been reports of immediate calvarial reconstruction with free tissue transfer; however, these cases did not use custom PEEK implants.[11] We believe that the staged approach combined with the free tissue transfer allowed us to proceed with secondary cranioplasty in a more expedited fashion.

A PEEK implant was chosen over other materials because the PEEK implant is rigid, radiolucent, does not resorb, provides strength similar to the surrounding bone,[12] allows for ease of oncologic surveillance,[9] and does not incorporate into the overlying soft tissue, allowing for ease of revision.

Given the aggressive nature of adenoid cystic carcinoma, regular surveillance and follow-up is essential to detect early recurrence.[13] The patient continues to be seen by a multidisciplinary team for routine oncologic surveillance.

Fig. 3. A photograph showing postoperative forehead contour.

CONCLUSIONS

We describe a case of refractory nasocranial fistulae in a contaminated and irradiated field, successfully managed in a staged fashion. Removal of nonviable bone flaps and obliteration of the fistulous communication with free tissue transfer is mandatory for successful treatment. Alloplastic cranioplasty can be safely performed in a delayed (8 weeks) fashion after completion of culture-directed antimicrobial treatment.

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DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

PATIENT CONSENT

The patient provided written consent for the use of her images.

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