Bilateral Inferior Turbinate Flaps for Salvage Reconstruction after Proton Beam Radiotherapy for Clival Chordoma

Kayva L. Crawford[1] Megana Saripella[2] Adam S. DeConde[1] Thomas L. Beaumont[3]

[1] Department of Otolaryngology – Head & Neck Surgery, University of California San Diego, La Jolla, California, United States
[2] School of Medicine, University of California San Diego, La Jolla, California, United States
[3] Department of Neurological Surgery, University of California San Diego, La Jolla, California, United States

Address for correspondence Thomas L. Beaumont, MD, PhD, Department of Neurological Surgery, University of California, San Diego, 9300 Campus Point Drive MC 7893, La Jolla, CA 92037-1300, United States (e-mail: tbeaumont@health.ucsd.edu).

J Neurol Surg Rep 2023;84:e87–e91.

Abstract

Clival chordoma is a rare, aggressive, notochord-derived tumor primarily managed with surgery via an endoscopic endonasal approach (EEA) and adjuvant proton beam radiotherapy. Reconstruction is commonly performed with a nasoseptal flap (NSF) at the time of initial surgery. While failures of the NSF are rare, they can occur following the initial surgery or in the setting of osteoradionecrosis. Salvage repair typically requires transfer of alternative vascularized tissues outside of the previously radiated field including regional scalp flaps such as pericranial or temporoparietal fascial flaps, or free vascularized tissue transfer. Here we describe the case of a 29-year-old woman with a history of clival chordoma with widespread skull base osteomyelitis secondary to NSF necrosis after proton beam radiotherapy. We describe successful skull base reconstruction with intranasal bilateral inferior turbinate flaps based on the sphenopalatine artery with lateral nasal wall extension, despite prior proton beam therapy and a failed prior vascularized intranasal reconstruction.

Keywords
- chordoma
- inferior turbinate
- flap
- osteoradionecrosis
- proton
- radiotherapy

Introduction

Clival chordomas are rare, aggressive, notochord-derived tumors with an incidence of approximately 8 per 10 million.[1] Management commonly includes surgical resection via an endoscopic endonasal approach (EEA) with consideration for adjuvant proton beam radiotherapy.[2,3] After resection of clival chordoma, vascularized coverage of the ventral skull base is paramount to avoid complications such as cerebrospinal fluid (CSF) leak and meningitis, and this is typically achieved using a nasoseptal flap (NSF) at the time of initial surgery.[4] While the NSF is a popular initial reconstructive option, failure rates increase in the setting of skull base radiation; a study by Thorp and colleagues found that in 37 patients with anterior skull base defects reconstructed with NSF who underwent radiation, 10.8% developed osteoradionecrosis (ORN), 8.1% developed a CSF leak, 8.1% developed pneumocephalus, and 2.7% developed meningitis.[5] This exceeds the previously reported 2% necrosis rate among all NSFs, and highlights the importance of identifying alternative tissue coverage sources for the anterior skull base including the lateral nasal wall, inferior or middle turbinates, temporoparietal fascia (TPF) flaps, or anterolateral thigh (ALT) free tissue transfer.[6–9]

received
February 8, 2023
accepted after revision
June 17, 2023

DOI https://doi.org/10.1055/s-0043-1772200.
ISSN 2193-6366.

Georg Thieme Verlag KG, Rüdigerstraße 14, 70469 Stuttgart, Germany

In this case, we employed bilateral inferior turbinate flaps (ITFs) to reconstruct a persistent clival defect in the setting of acute osteomyelitis, meningitis, and chronic wound dehiscence after prior proton therapy to the ventral skull base. This case highlights the utility of ITFs in reconstructing the ventral skull base for patients in whom skull base reconstruction with an NSF has undergone late failure after radiation therapy.

Case Report

We present the case of a 29-year-old female patient with a history of methamphetamine abuse and chronic rhinosinusitis who presented to our institution with neurological decline in the setting of purulent meningitis 6 years after management of clival chordoma at an outside hospital.

At the time of initial diagnosis, magnetic resonance imaging (MRI) demonstrated a 6.7-cm anteroposterior by 5-cm craniocaudal × 5.2-cm transverse heterogeneously enhancing mass with displacement of the left cavernous carotid artery and bilateral vertebral arteries (►**Fig. 1**). The patient underwent subtotal resection via EEA and reconstruction with an abdominal fat graft and bilateral NSFs given the size of the dural defect. She underwent a second-stage EEA for debulking 2 months later. This was performed jointly by neurosurgery and head and neck surgery. Five months later, she was found to have residual chordoma and underwent a right far-lateral approach with intact canal wall mastoidectomy and decompression of the sigmoid sinus, jugular bulb, and posterior fossa dura. Adjuvant proton beam radiotherapy was then completed at an outside hospital 5 months postoperatively with an isodose of 7,920 cGy to the clivus (►**Fig. 2**). The patient did well posttreatment and remained neurologically intact with Karnofsky Performance Status (KPS) of 90%, although she developed chronic sinusitis after treatment.

On admission to our institution, the patient was found to have widespread skull base dehiscence and osteomyelitis secondary to NSF necrosis in the context of postoperative proton therapy and insufflation of recreational drugs. Given that all blood and CSF cultures showed no growth, the patient was started on broad-spectrum intravenous antibiotics and demonstrated significant neurological improvement except for persistent right cranial nerve (CN) VII palsy, House–Brackmann (HB) grade 4 as well as right tongue deviation, fasciculations, and atrophy. She required revision EEA for anterior skull base debridement and reconstruction of the ventral skull base defect. Given the extent of the expected defect following debridement and history of proton therapy to the skull base, initial reconstruction options considered included temporoparietal facial flaps and free tissue transfer with radial forearm or, alternatively, an ALT flap. However, after a careful review of the proton fields, it became apparent that there was relative sparing of the bilateral pterygopalatine fossa (PPF) (2,600–4,200 cGy), which prompted us to select staged EEA with inferior turbinate reconstruction. This allowed us to preserve additional options for salvage if necessary and spare the morbidity of regional and free flap reconstruction.

Description of Surgical Procedure

General endotracheal anesthesia was initiated, and the head was placed into fixation with Mayfield pins. Intraoperative image guidance was registered to a thin-slice computed tomography (CT) angiogram that was co-registered to a T1 postcontrast MRI. A binarial EEA was initiated with a 0-degree endoscope. Greenish yellow phlegmon was

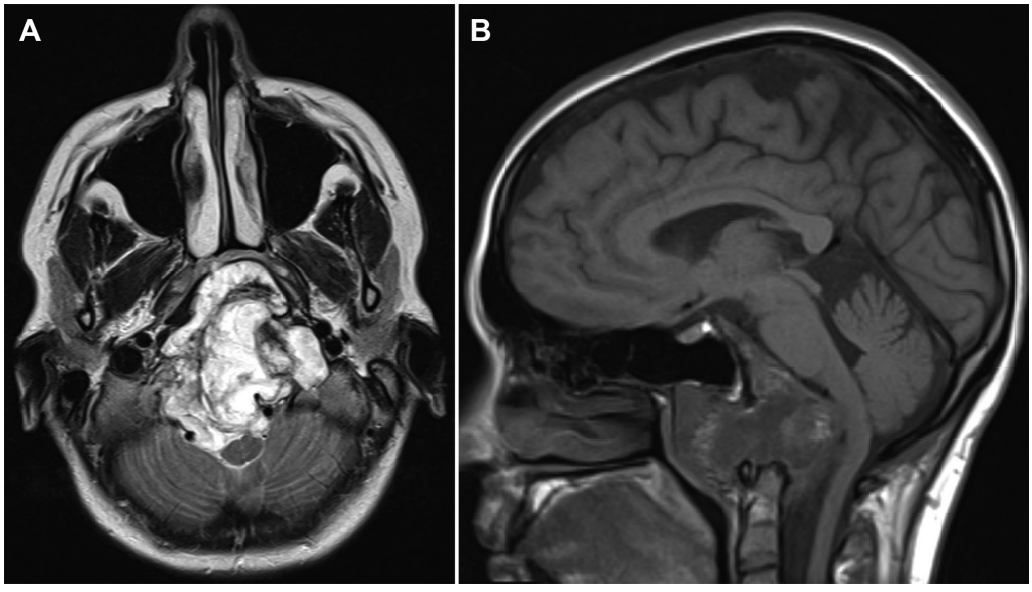

Fig. 1 Magnetic resonance imaging (MRI) demonstrating large clival chordoma at the time of initial diagnosis. (**A**) Axial T2-weighted sequence demonstrates a hyperintense mass extensively involving the clivus and extending inferiorly to C2. (**B**) Sagittal T1-weighted sequence demonstrating obliteration of the nasopharynx with dorsal extension causing marked mass effect and displacement of the cervicomedullary junction.

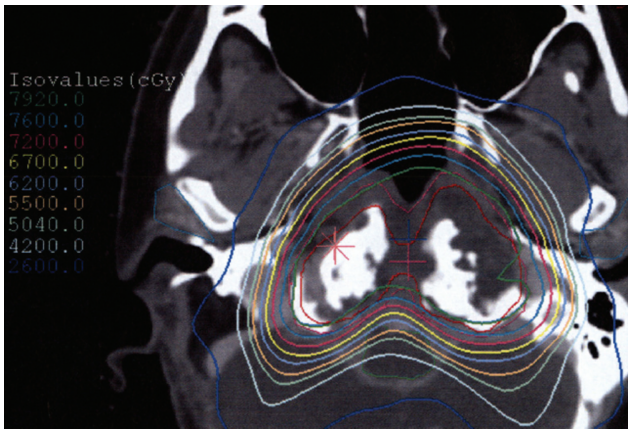

Fig. 2 Proton beam radiation fields demonstrating 7,920 cGy delivered to the clivus at the site of flap dehiscence. Careful review also demonstrates relative sparing of the pterygopalatine fossa (PPF) (2,600–4,200 cGy), which prompted consideration of intranasal reconstructive options.

immediately noted at the site of prior clival resection (►**Fig. 3A**). Egress of CSF was also noted from the site of skull base dehiscence. On closer inspection, several areas of dehiscence of the prior NSF were noted, with fistulous tracks along the right side of the clival resection and extending along the clival dura. Osteonecrosis was noted just inferior and posterior to the foramen lacerum and paraclival carotid, just anterior to the takeoff of the occipital condyle. On the left side, a large portion of residual clivus medial and inferior to the foramen lacerum remained. Given the initial goal was exploration and repair of the CSF leak with vascularized tissue, a high-speed drill was used to debride the necrotic petrous temporal bone on the right until bleeding tissue was encountered.

To reconstruct the skull base defect, a right-sided, vascularized, posteriorly based inferior turbinate mucoperiosteal flap based on the sphenopalatine artery (SPA) with an extended lateral wall was rotated posteriorly over the defect. Local anesthesia and vasoconstriction were achieved with 1% lidocaine with 1:100,000 epinephrine. Monopolar cautery with an extended needle tip and a suction Freer elevator were used to lift a large area of mucosa over the right inferior turbinate with extension superiorly onto the lateral nasal wall. This flap was then rotated posteriorly over the clival defect and secured with a combination of Gelfoam and Merocel spacer.

There was no postoperative CSF leak and the patient continued to improve in hospital with antibiotic treatment. Flap viability was monitored closely in the outpatient setting. She completed a course of prolonged broad-spectrum intravenous (IV) antibiotics under the care of the Infectious Disease service and underwent second-stage surgical debridement 8 weeks later. During debridement, the right ITF was found to be well healed; however, there was a small region of persistently exposed bone over the left petrous apex (►**Fig. 3B**). This was drilled widely, and a left-sided ITF was elevated, rotated posteriorly, and secured as described earlier during stage 1. At the 6-month post-op follow-up, nasal endoscopy revealed full mucosalization without infection, dehiscence, or CSF leak (►**Fig. 3C**). Postoperative MRI demonstrating bilateral inferior turbinate grafts and closure of the CSF fistula can be found in ►**Fig. 4**. The patient's leukocytosis and inflammatory markers had normalized, and she had returned to her prior neurological baseline with trace CN VII weakness and long-standing mild unilateral CN XII weakness. She subsequently developed right hemifacial spasm that was treated with Botox and facial physical therapy.

Discussion

Clival reconstruction is commonly achieved with a pedicled NSF, as this is easily accessible and adds minimal morbidity to an expanded endoscopic approach.[10] While the increased use of the NSFs for reconstruction has significantly improved clival reconstruction outcomes such as CSF leak rates since its initial description in 2006, a systematic review by Lavigne reports a flap necrosis rate of at least 1.3%, with other studies demonstrating higher rates ranging from 5.7 to 8.1% in the setting of adjuvant radiation.[4,5,10–14] The mucosal area from which the NSF is lifted is also left with a large, denuded area that can take up to 3 months to heal, leaving this area with a

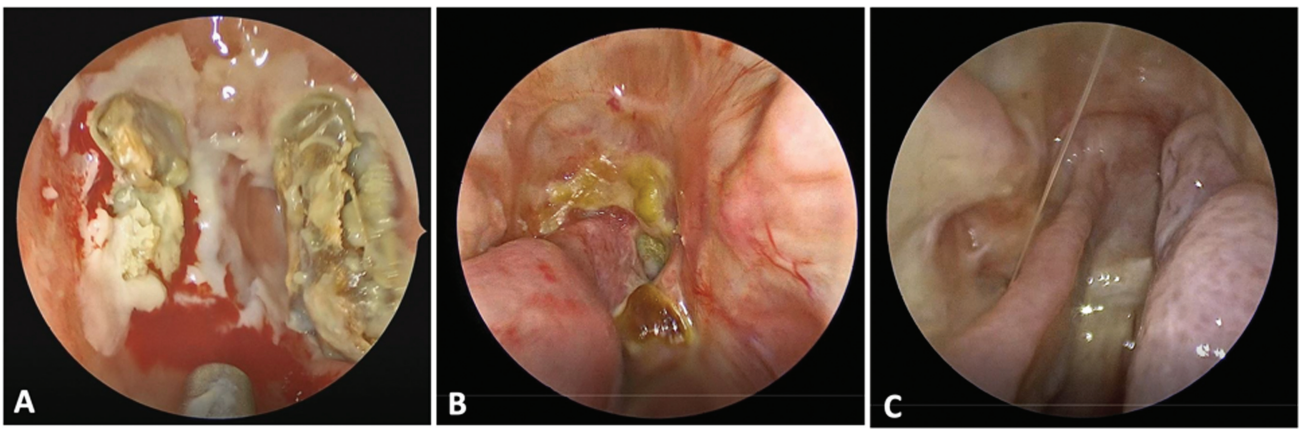

Fig. 3 (**A**) Intraoperative image of the skull base osteoradionecrosis (ORN) prior to debridement. (**B**) Right inferior turbinate flap in place with persistent skull base dehiscence. (**C**) Left inferior turbinate flap with well-healed defect.

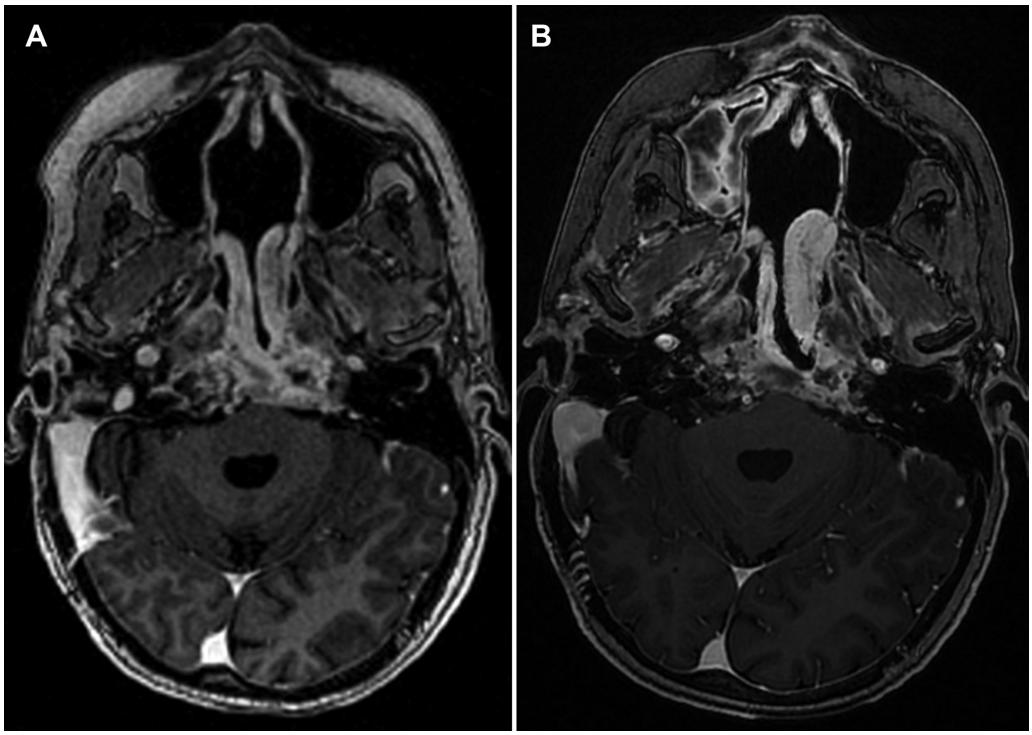

Fig. 4 Postoperative magnetic resonance imaging (MRI) following skull base debridement and bilateral inferior turbinate flap placement. Axial postcontrast T1-weighted MRI (**A**) 3 months following stage 1 and (**B**) 24 months following stage 2 with enhancement of bilateral inferior turbinate grafts and closure of cerebrospinal fluid (CSF) fistula. The patient has asymptomatic mucosal congestion noted in the right maxilla.

lack of functional mucosa that can cause crusting and anosmia.[15] In cases requiring secondary resection, or for patients who undergo septectomy, the NSF may not provide adequate coverage of the skull base defect.[6] Additional complications including necrosis, prolonged crusting, nasal deformity such as saddle nose, nasal perforation, and persistent CSF leakage and/or meningitis may necessitate the use of vascularized mucosal flaps, such as the ITF.[6,13,15]

Prior studies have demonstrated that the posterior ITF has a success rate of approximately 60%, with greater success as surgical experience increased.[16] As surgical manipulation of the inferior turbinates is often avoided during primary resection of clival chordoma, this flap is an available and durable option that has a robust blood supply. However, due to its smaller pedicle, the flap utilizes a more limited arc of rotation and is more technically difficult to place, causing it to be reserved primarily for salvage cases.[6] Unilateral ITFs also may not adequately cover a large petroclival defect, and thus, complete reconstruction may require an additional contralateral ITF to achieve complete coverage. Inclusion of the contralateral inferior turbinate, while carrying low morbidity rates, requires more time and increased mucosa removal.[12] Anterior and posterior ITFs have been described successfully in the literature, proving adequate blood flow in all studied cases.[6,17] Thus, the ITF represents a viable intermediary option, allowing salvage reconstruction without the need for regional or free tissue transfer.

Reconstruction of the skull base in a salvage setting can be a challenging technical problem and often requires extranasal tissue from sources such as TPF or ALT flaps, particularly in the setting of adjuvant radiation and infection.[18,19] While these flaps have the advantage of bringing in robust vascularized tissue from outside the field of radiation, they have significantly higher morbidity than intranasal flaps. Given that our patient presented with chronic dehiscence, meningitis, and osteomyelitis, an initial low morbidity procedure with ITFs was made to reserve complex salvage reconstruction options from extranasal donor sites if poor tissue quality and acute infection impeded wound healing. Importantly, this patient's PPF was spared during proton radiation therapy. We believe that this contributed to the robustness of this flap as the vascular supply to the ITFs was less compromised. Despite the limitations of reporting a single case, we demonstrate proof of concept that bilateral ITFs represent a viable option for reconstruction of large ventral skull base defects for patients in whom NSF reconstruction has failed.

Conflict of Interest
None declared.

References

1 McMaster ML, Goldstein AM, Bromley CM, Ishibe N, Parry DM. Chordoma: incidence and survival patterns in the United States, 1973-1995. Cancer Causes Control 2001;12(01):1–11
2 Walcott BP, Nahed BV, Mohyeldin A, Coumans JV, Kahle KT, Ferreira MJ. Chordoma: current concepts, management, and future directions. Lancet Oncol 2012;13(02):e69–e76
3 Culebras D, Torales J, Reyes LA, et al. Extended endoscopic endonasal surgery for clival chordoma and chondrosarcoma: our experience in 14 cases. Neurocirugia (Astur : Engl Ed) 2018; 29(04):201–208

4 Hadad G, Bassagasteguy L, Carrau RL, et al. A novel reconstructive technique after endoscopic expanded endonasal approaches: vascular pedicle nasoseptal flap. Laryngoscope 2006;116(10):1882–1886
5 Thorp BD, Sreenath SB, Ebert CS, Zanation AM. Endoscopic skull base reconstruction: a review and clinical case series of 152 vascularized flaps used for surgical skull base defects in the setting of intraoperative cerebrospinal fluid leak. Neurosurg Focus 2014;37(04):E4
6 Yip J, Macdonald KI, Lee J, et al. The inferior turbinate flap in skull base reconstruction. J Otolaryngol Head Neck Surg 2013;42(01):6
7 Fortes FS, Carrau RL, Snyderman CH, et al. The posterior pedicle inferior turbinate flap: a new vascularized flap for skull base reconstruction. Laryngoscope 2007;117(08):1329–1332
8 Hanasono MM, Sacks JM, Goel N, Ayad M, Skoracki RJ. The anterolateral thigh free flap for skull base reconstruction. Otolaryngol Head Neck Surg 2009;140(06):855–860
9 Arosio AD, Coden E, Karligkiotis A, et al. Temporoparietal fascia flap endonasal transposition in skull base reconstruction: surgical technique. World Neurosurg 2021;146:118
10 Yamada H, Toda M, Fukumura M, et al. Cerebrospinal fluid leakage due to nasoseptal flap partial necrosis: a pitfall for skull base reconstruction of endoscopic endonasal surgery. Surg Neurol Int 2020;11(121):121
11 Chabot JD, Patel CR, Hughes MA, et al. Nasoseptal flap necrosis: a rare complication of endoscopic endonasal surgery. J Neurosurg 2018;128(05):1463–1472
12 Harvey RJ, Parmar P, Sacks R, Zanation AM. Endoscopic skull base reconstruction of large dural defects: a systematic review of published evidence. Laryngoscope 2012;122(02):452–459
13 Lavigne P, Faden DL, Wang EW, Snyderman CH. Complications of nasoseptal flap reconstruction: a systematic review. J Neurol Surg B Skull Base 2018;79(10, Suppl 4):S291–S299
14 Zanation AM, Carrau RL, Snyderman CH, et al. Nasoseptal flap reconstruction of high flow intraoperative cerebral spinal fluid leaks during endoscopic skull base surgery. Am J Rhinol Allergy 2009;23(05):518–521
15 Ruffner R, Pereira MC, Patel V, Peris-Celda M, Pinheiro-Neto CD. Inferior meatus mucosal flap for septal reconstruction and resurfacing after nasoseptal flap harvest. Laryngoscope 2021;131(05):952–955
16 Lee DH, Yoon TM, Lee JK, et al. Clinical utility of the inferior turbinate flaps in the reconstruction of the nasal septum and skull base. J Craniofac Surg 2012;23(04):e322–e326
17 Gil Z, Margalit N. Anteriorly based inferior turbinate flap for endoscopic skull base reconstruction. Otolaryngol Head Neck Surg 2012;146(05):842–847
18 Kim SM, Park HW, Park MS, et al. Fulminant meningitis after radiotherapy for clival chordoma. Brain Tumor Res Treat 2013;1(02):107–110
19 Moon JH, Kim EH, Kim SH. Various modifications of a vascularized nasoseptal flap for repair of extensive skull base dural defects. J Neurosurg 2019;132(02):371–379