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Endoscopic endonasal resection of sinonasal teratocarcinosarcoma with intracranial breakthrough: illustrative case

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BACKGROUND Teratocarcinosarcoma traversing the anterior skull base is rarely reported in literature. The heterogenous and invasive features of the tumor pose challenges for surgical planning. With technological advancements, the endoscopic endonasal approach (EEA) has been emerging as a workhorse of anterior skull base lesions. To date, no case has been reported of EEA totally removing teratocarcinosarcomas with intracranial extensions.

OBSERVATIONS The authors provided an illustrative case of a 50-year-old otherwise healthy man who presented with left-sided epistaxis for a year. Imaging studies revealed a 31 × 60-mm communicating lesion of the anterior skull base. Gross total resection via EEA was achieved, and multilayered skull base reconstruction was performed.

LESSONS The endoscopic approach may be safe and effective for resection of extensive teratocarcinosarcoma of the anterior skull base. To minimize the risk of postoperative cerebrospinal fluid leaks, multilayered skull base reconstruction and placement of lumbar drainage are vitally important.

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KEYWORDS endoscopic; endonasal; teratocarcinosarcoma; transplanum; transtuberculum; sphenoidotomy; ethmoidectomy

Communicating lesions of the anterior skull base consist of a wide range of pathologic entities that pose both diagnostic and therapeutic challenges for neurosurgeons. Patients present with nonspecific symptoms, including nasal congestion, epistaxis, and headache. Imaging findings are also unspecific, with overlapping features. These lesions traverse anatomical compartments and may arise superiorly from the brain (e.g., olfactory groove meningiomas, subfrontal schwannomas), inferiorly from the sinonasal tract (e.g., squamous cell carcinomas, adenocarcinomas), from the bone proper (e.g., chordomas, fibrous dysplasia), or from systemic conditions (e.g., multiple myelomas, Langerhans cell histiocytosis).[1–5] Of note, nonneoplastic lesions such as frontoethmoidal mucoceles and invasive fungal sinusitis may also reveal aggressive features.[1] To date, there are few guidelines regarding this entity of lesions. Therefore, management approaches should be individualized and discussed on a multidisciplinary basis.

Here we present a case of sinonasal teratocarcinosarcoma (SNTCS) with intracranial breakthrough, a rare entity with only about 15 cases (Supplementary Table 1) reported in literature. The tumor had intracranial breakthrough and was completely resected by the senior author (M.Q.L.) via the endoscopic endonasal approach (EEA) with rigorous skull base reconstruction. To our knowledge, this is the first case of teratocarcinosarcoma with intracranial extension to achieve gross total resection and successful skull base reconstruction via EEA.

ABBREVIATIONS CSF = cerebrospinal fluid; CT = computed tomography; EEA = endoscopic endonasal approach; MRI = magnetic resonance imaging; SNTCS = sinonasal teratocarcinosarcoma.

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Illustrative Case

A 50-year-old otherwise healthy man presented at the ear, nose, and throat office with intermittent left-sided epistaxis and dizziness lasting for a year. Computed tomography (CT) at another hospital revealed a left-sided mass located at the anterior cranial fossa. Biopsy indicated malignancy arising from the paranasal sinuses (Supplementary Fig. 1A–E). Smoking and alcohol history were positive. He denied any occupational exposure to toxic or radioactive materials. On admission, the patient was alert and oriented. Vital signs were normal. There was no lymphadenopathy. The patient was further referred to our neurosurgery department.

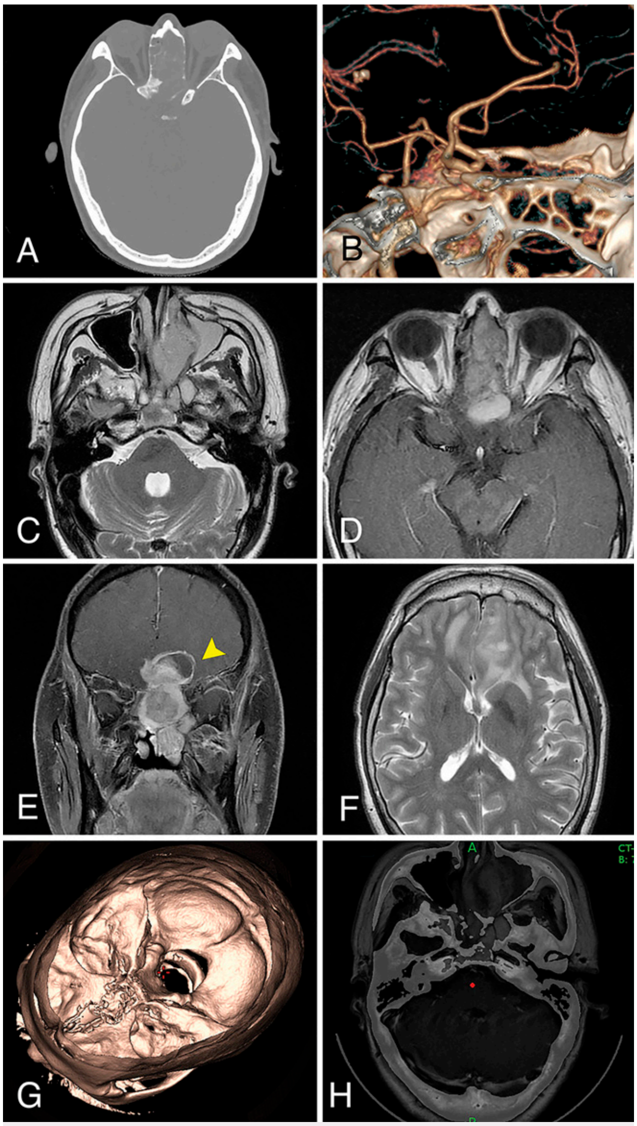

FIG. 1. Preoperative imaging studies and snapshots of the preoperative surgical planning. **A:** CT revealed the lesion at the anterior skull base (low density area). **B:** Further three-dimensional reconstruction confirmed bony destruction of the ethmoidal sinuses while sparing the crista galli. **C:** T2-weighted image (T2WI) revealed fluid retention in the left maxillary sinus due to obstruction by the tumor. **D:** T1-weighted image (T1WI) with contrast showed heterogeneously enhanced lesion of the anterior skull base. **E:** The cystic component (*yellow arrowhead*) of the intracranial portion was a feature like esthesioneuroblastoma. The right-sided tumor margin was not well-defined, which indicated pia mater invasion. This was further confirmed during the resection process. **F:** T2WI also indicated left frontal lobe edema. **G:** Midline structures of the anterior skull base was extensively destructed by the lesion, which was anteriorly limited by the crista galli and posteriorly limited by the tuberculum sellae. **H:** Further merging of CT and MRI confirmed remnants of the cribriform plate and the ethmoidal sinuses.

Preoperative CT with contrast revealed a lesion of the anterior skull base (Fig. 1A and B). MRI showed a well-defined 31 × 60-mm mass of irregular shape with heterogenous enhancement (Fig. 1C–E). The left frontal lobe abutted frank vasogenic edema (Fig. 1F). Preoperative surgical planning (StealthStation S7, Medtronic, Minneapolis, MN) further confirmed cribriform plate destructions (Figs. 1G, 1H, and 2).

Management approaches were discussed on a multidisciplinary basis. Resection (Video 1; Fig. 3) by binostril EEA was decided with the patient's consent after family meeting. Neuronavigation (StealthStation S7) was set up in the preparatory phase. Extensive sphenoidotomy and ethmoidectomy were performed to remove the intranasal portion of the tumor. Via the transplanum-transtuberculum approach, the peritumoral bony margin was drilled and extended. Care was taken to protect the underlying dura as long as it was

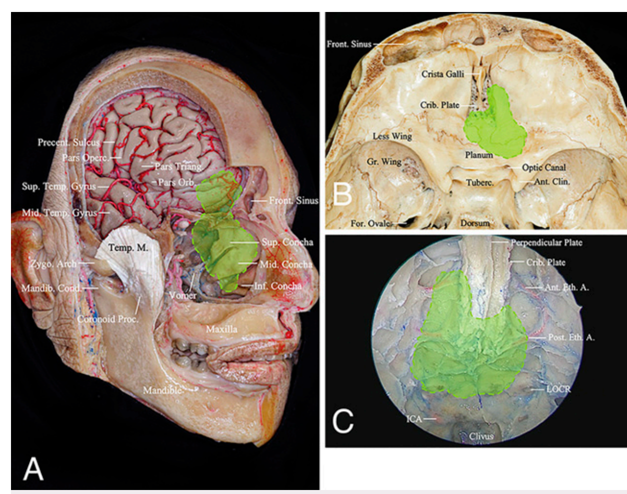

FIG. 2. Surface projections (*green overlay*) of the tumor on different viewing planes of human cadaveric specimens. **A:** The longitudinal extension (*green overlay*) of this tumor was defined superiorly by the left frontal lobe and inferiorly by the left inferior nasal concha. The frontal lobe abutted vasogenic edema, which was revealed on T2WI. The left-sided nasal cavity was occupied by the tumor, making endoscopic resection optimal choice for this portion. **B:** The anteroposterior extension (*green overlay*) was limited anteriorly by the crista galli and posteriorly by the planum sphenoidale. Despite the posterior proximity of the tumor to the optic canal, the right-sided optic nerve was not invaded. The crista galli was also spared. **C:** Endoscopic view of the skull base indicated that the tumor received arterial supplies from the ethmoidal arteries. The internal carotid artery was not involved because the tumor was limited laterally by the optico-carotid recess. A = artery; Ant = anterior; Clin = clinoid; Cond = condyle; Crib = cribriform; Eth = ethmoidal; For = foramen; Front = frontal; Gr = greater; ICA= internal carotid artery; Inf = inferior; LOCR = left optico-carotid recess; M = muscle; Mandib = mandibular; Mid = middle; Orb = orbitalis; Operc = opercularis; Post = posterior; Precent = precentral; Proc = process; Sup = superior; Temp = temporal; Triang = triangularis; Tuberc = tuberculum; Zygo = zygomatic. Used with permission from Yuanzhi Xu.

intact by tumoral invasions. The dura of the tuberculum sellae was incised open, and the tumor was carefully removed in a piecemeal fashion. The skull base was reconstructed using multilayered materials according to institutional protocols (Fig. 4). Lumbar drainage was placed for 5 days.

VIDEO 1. Clip showing endoscopic endonasal resection of a traversing SNTCS with intracranial breakthrough. Click here to view.

The postoperative course was uneventful and without new-onset neurological deficits. Postoperative imaging studies confirmed gross total resection (Fig. 5). Pathologic findings were consistent with teratocarcinosarcoma (Supplementary Table 2; Supplementary Fig. 2;). The patient was referred for further chemoradiotherapy and discharged home. There was no local recurrence of tumor upon 1-year follow-up.

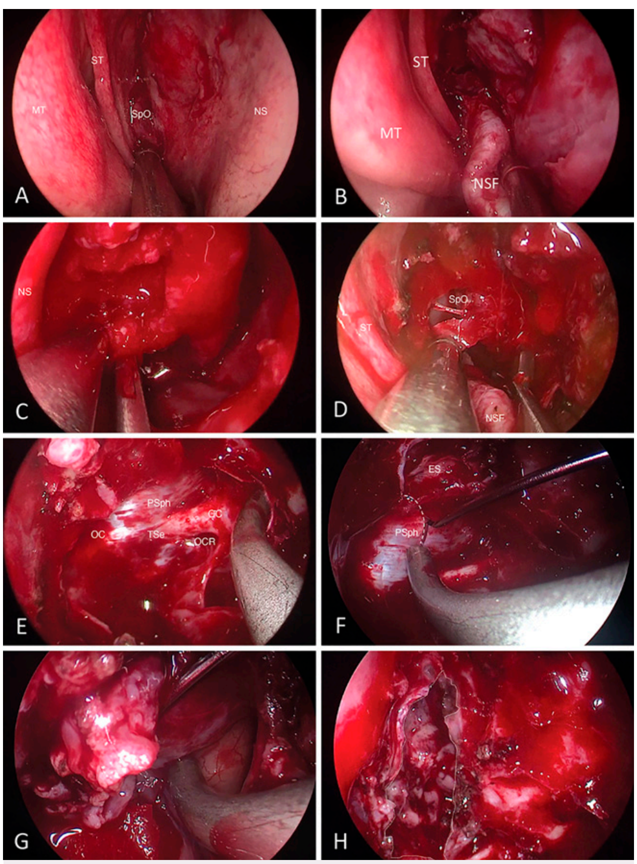

FIG. 3. Intraoperative snapshots of sequential endoscopic resection of the tumor. **A:** First, important anatomical landmarks in the periphery of the sphenoid ostium were identified. **B:** Nasoseptal flap was harvested and placed into the posterior nasopharynx. **C and D:** The sphenoid sinuses were opened, and the rest of the intranasal portion was removed before the extended sphenoidotomy. **E:** After removal of the tumor in the ethmoidal sinuses, important landmarks in the periphery of the planum sphenoidale were identified. **F:** The dura of the planum sphenoidale was incised open, which was brought together with (*dashed line*, incision) the already incised dura of the ethmoid sinuses. **G:** The tumor-brain interface was explored, and the tumor was carefully detached from the brain. **H:** The tumor was fully removed. Part of the underlying brain parenchyma was invaded by the tumor. If left unrepaired, the skull base defect (bounded by the *dotted line*) would give rise to CSF leaks and infections in the postoperative course. ES = ethmoidal sinuses; MT = middle turbinate; NS = nasal septum; NSF = nasoseptal flap; OC = optic canal; OCR = optico-carotid recess; PSph = planum sphenoidale; SpO = sphenoid ostium; ST = superior turbinate; TSe = tuberculum sellae.

Discussion

Observations

Based on history and biopsy results, differential diagnosis in this case includes squamous cell carcinoma, adenocarcinoma, adenoid cyst carcinoma, esthesioneuroblastoma, and rare entities such as carcinosarcoma and teratocarcinosarcoma (Supplementary Table 3).[6] According to the existing literature, multimodality treatment, including

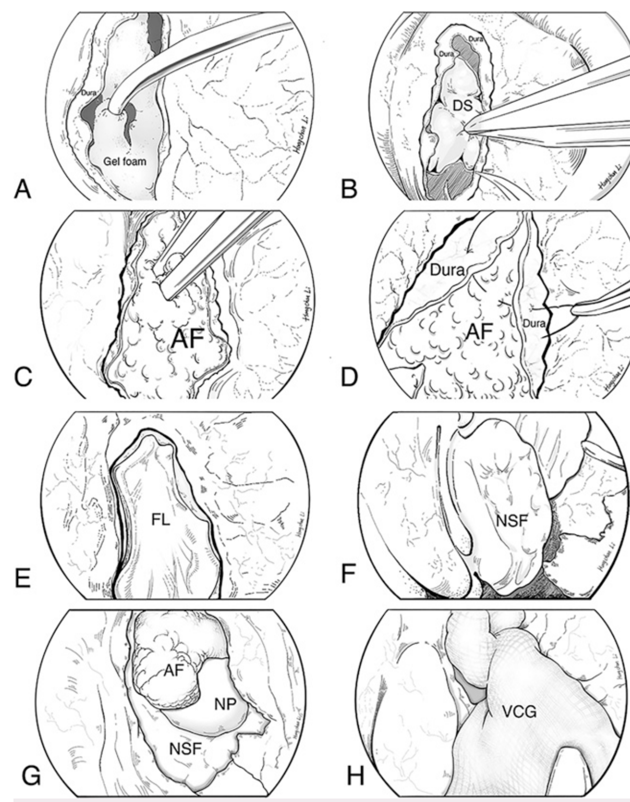

FIG. 4. The artist's (Hongchan Li) illustrations of sequential skull base reconstruction in this patient. **A:** Gel foam was used to cover the underlying brain parenchyma. **B and C:** This was followed by subdural reconstruction composed of the dural substitute (DuraMax, TianXinFu Medical Appliance Co., Ltd., Beijng, China) and autologous fat tissue. **D:** A continuous uninterrupted suture using 6–0 PROLENE polypropylene sutures (Ethicon Inc., Raritan, NJ) was then performed. Given the local tension and the large defect, a watertight suture was impossible. Despite this, dural suturing was still necessary because it would bolster the inlay materials to achieve final watertight closure of the skull base in a multilayer reconstruction setting. **E:** Fascia lata of the thigh was used for epidural overlay. **F:** The pedicled nasoseptal flap was placed to cover the dura. **G:** Autologous fat tissue and Nasopore (Stryker, Kalamazoo, MN) were added for further support as epidural overlay. **H:** We applied a Vaseline-coated gauze strip (Unilever, London, UK) as a final bolster of the above materials. AF = autologous fat; DS = dural substitute; FL = fascia lata; NP = Nasopore; NSF = nasoseptal flap; VCG = Vaseline-coated gauze strip.

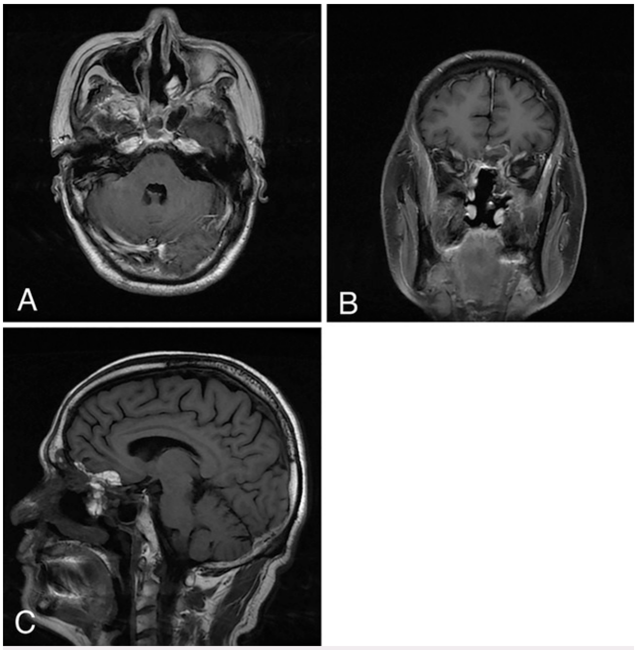

FIG. 5. MRI 1 month postoperatively indicated gross total resection of the tumor. **A:** Axial view of T1 fluid attenuated inversion recovery with contrast indicated complete removal of the tumor within the ethmoidal sinuses. **B:** Coronal view of T1WI revealed complete remission of the vasogenic edema. **C:** T1WI showed intact materials of the skull base reconstructions in place (high signal intensity).

surgery for sinonasal malignancy, can improve survival.[7–12] Therefore, we opted for initial resection followed by adjuvant therapies.

The anterior skull base can be managed surgically by transfacial, transcranial open or keyhole, purely endoscopic, or combined transbasal and transnasal approaches. Selection of these approaches should be based not only on expected optimal outcomes but also on surgical abilities and preferences of the operating surgeon. This especially applies for the endoscopic approach, which involves a steep learning curve. For giant lesions that traverse anatomical compartments, surgical planning is highly individualized.[13] The tumor in this case remained midline, with both intracranial and intranasal portions. Therefore, it could be readily accessed from a combined transnasal and transbasal approach.[14] EEA in this case was another solution. With recent advancements, EEA has proven itself a safe alternative to the open approach for anterior and middle skull base lesions.[13,15–18] The endoscope provided panoramic and close-up views to search for potential tumor remnants. In addition, the tumor was likely to be supplied by the ethmoidal arteries with meningeal branches, which would be more readily cauterized under direct visualization with the endoscope. Of note, postoperative CSF leak (overall rate of 8.5%) remains a major concern for the transnasal endoscopic approach.[19] The tumor here eroded the posterior two-thirds of the cribriform plate. High-flow intraoperative CSF leaks secondary to a large skull defect were a major challenge for watertight reconstruction.[20–22]

Many techniques have been proposed and have evolved through time, but the standard is not well established (Supplementary Table 4). The “gasket-seal” technique decreases the CSF leak rate of extensive endonasal surgeries to 4.3%.[21,23] For larger defects, a nasoseptal pedicled flap with multilayered repairs should suffice, especially in grade 3 leaks.[19,24–26] In addition, dura suturing has been practiced and proven effective.[25,27–35] It was possible that the dura of the anterior skull base was not fully invaded. Otherwise, abdominal fat could be used as the primary reconstruction material.[36] To further reduce postoperative CSF leaks, many studies advocated the placement of lumbar drainage.[37–39] In our center, we achieved an overall 2.4% (4/170, unpublished data) postoperative CSF leak rate for first-onset anterior skull base lesions using a graded reconstruction approach. Therefore, multilayered skull base reconstruction with dura suturing was feasible and justified in this case.

To the best of our knowledge, there are approximately 128 SNTCS cases reported in English literature, with around 15 cases of intracranial breakthrough.[12,40] The natural history of SNTCS is not fully understood. Given its aggressive nature, teratocarcinosarcoma has a 55% mean survival at 2 years, with a recurrence rate of 38%.[12] Resection is still the mainstay of treatment. Although neoadjuvant chemo- and radiotherapy were reported, their effective roles have not been well studied.[10,12,41,42]

Lessons

In the hands of an experienced neurosurgeon, the EEA could be safe and effective for resecting a giant and extensive lesion of the anterior skull base, such as SNTCS. To minimize the risk of postoperative CSF leaks, the importance of meticulous skull base reconstruction and placement of lumbar drainage cannot be overemphasized.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Lou, Ni, Xu. Acquisition of data: Lou, Ni, Xu, X Zhang, Li, Shen, F Wang, Z Wang. Analysis and interpretation of data: Lou, Ni, Xu, X Zhang, Shen, Ren, F Wang, Zhou, J Wang. Drafting the article: Lou, Ni. Critically revising the article: Lou, Ni, Bi. Reviewed submitted version of manuscript: Lou, Ni, Xu, Dong, A Zhang, Zhu. Approved the final version of the manuscript on behalf of all authors: Lou. Statistical analysis: Lou, Ni. Administrative/technical/material support: Lou, Yuan. Study supervision: Lou, Dong, Ren.

Supplemental Information

Video

Video 1. https://vimeo.com/591624150.

Online-Only Content

Supplemental material is available with the online version of the article.

Supplementary Tables and Figures. http://thejns.org/doi/suppl/10.3171/CASE21471.

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