Resection of an upper cervical aneurysmal bone cyst and spinal reconstruction using a midline mandibular osteotomy in a pediatric patient

Case report


Departments of †Neurological Surgery and ‡Otolaryngology–Head and Neck Surgery; and §Section of Hospital Dentistry, Division of Oral and Maxillofacial Surgery, Columbia University, New York, New York

The authors report on the surgical management of an extensive lesion of the upper cervical spine that required an uncommon transmandibular approach to facilitate exposure, resection, and stabilization in a pediatric patient. A 6-year-old boy with a large aneurysmal bone cyst of the C-2 vertebra presented with progressive weakness and right-sided neck pain. The lesion extended laterally into the soft tissue of the neck, inferiorly to C-4, and posteriorly around the spinal cord. A transmandibular osteotomy was performed to provide adequate exposure for complete resection of the mass and anterior C1–3 instrumentation and fusion. Subsequently, the patient underwent occiput to C-4 posterior instrumentation and fusion. The patient tolerated the operation well and had regained all function at 3 and 11 months’ follow-up. No neurological complications or problems of speech, swallowing, or respiration occurred. Even in pediatric patients, the transmandibular approach for the treatment of upper cervical spine lesions is an effective method of maximizing exposure for complex lesions requiring resection and stabilization. (http://thejns.org/doi/abs/10.3171/2014.3.PEDS13511)

Key Words • transmandibular approach • aneurysmal vertebral cyst • osteotomy • anterior fusion • pediatric • spine

Transmandibular approaches to the cervical spine are highly uncommon surgeries requiring extensive surgical collaboration. With the growing experience in endoscopic transnasal and transoral techniques for upper cervical lesions, even fewer cases necessitate jeopardizing the pharyngeal and lingual architecture and the neurovasculature, which is inherent to the transmandibular approach. In some circumstances, the exposure required for complete resection and stabilization of a high cervical lesion cannot be accomplished using an endoscopic transnasal, an endoscopic transoral, or a high cervical retropharyngeal approach. In this report, we describe the use of a transmandibular approach for the resection of an extensive aneurysmal bone cyst of the C-2 vertebra with spinal reconstruction.

Case Report

History and Examination. In April 2012, a 6-year-old boy presented to the Morgan Stanley Children’s Hospital, Columbia University Medical Center, New York, for evaluation after a several-month history of progressive right-sided neck pain and torticollis. He was previously healthy and had no remarkable medical history, although over the past 2 months he had begun to lose weight and demonstrate right-sided weakness, a deepening of his voice, and snoring. Computed tomography and MRI studies revealed a lytic C-2 lesion with an adjacent soft-tissue mass compressing the posterior pharynx (Fig. 1). A skeletal survey did not demonstrate additional metastasis, and a CT-guided biopsy of the lesion was histologically consistent with an aneurysmal bone cyst. After a multidisciplinary discussion, a transmandibular approach was selected to provide the necessary exposure to resect the entire mass and reconstruct the spine.

Operation. The patient was brought to the operating room and was positioned supine with his neck in mild extension. After inducing anesthesia, we performed a tracheostomy to maintain the airway. A lip-splitting incision down the level of the thyroid notch with a transverse extension was marked out. An initial transcervical approach

* Mr. McDowell and Dr. Hanft contributed equally to this paper.
Midline mandibular osteotomy and anterior fusion

was taken where a superior subplatysmal flap was raised to the inferior border of the submandibular gland and the thyroid notch (Figs. 2A and 3). The medial borders of the carotid sheath were exposed but not opened. A window was created between the larynx and the carotid sheath into the retropharyngeal space. Although the tumor was palpable with this approach, the exposure was insufficient for a safe resection and subsequent reconstruction; therefore, the superior flap was further raised along the pre-marked lip-splitting incision. The hypoglossal nerve was identified and isolated. The lower lip was then completely split at the midline, and a sulcular gingival incision was made from the right primary canine to the left; a vertical incision was made in the gingiva at the right primary canine into the vestibule and anteriorly toward the midline down to the bone. The created flap was elevated laterally from the central incisors. The anterior symphyseal region was then exposed, a 2-mm fracture plate was adapted, and screw holes were placed for later attachment to the inferior border of the mandible. After the mentalis muscle was divided, a midline mandibulotomy was performed downward between the midline incisors using a reciprocating saw and a spatula osteotome (Fig. 4).

The anterior belly of the digastric muscle was released from the mandible, and the geniohyoid, geniglosus, and mylohyoid muscles were divided to allow for additional lateral dissection. The submandibular gland was reflected laterally with the mandible, and the submandibular duct was transected, cannulated, and laterally implanted after the lingual nerve was separated from it. Posterior pharyngeal exposure was achieved by dividing the styloglossus and palatoglossus muscles (Figs. 5 and 6). The mandibular swing was performed with careful
preservation of the neurovasculature. A final incision in the posterior oropharynx was performed to allow for exposure up to the top of C-1.

Dissection continued through the posterior oropharyngeal tissue from the top of C-1 to the bottom of C-3, with confirmation of position via fluoroscopy. The longus coli muscles were dissected bilaterally to expose the anterior vertebral bodies and the tumor. The tumor was removed, and the remaining normal cortical bone was drilled and removed via curettage. The additional soft-tissue mass was resected laterally, extending past the spinal cord, venous plexus, and vertebral artery.

The C2–3 disc and inferior dens were removed and a 50 × 10–mm cage was cut to fit the C1–3 space. The cage was filled with allograft and demineralized bone matrix prior to insertion. One 10-mm screw was placed in the anterior tubercle of C-1, and two 10-mm screws were placed in the anterior body of C-3 (Fig. 7). Fluoroscopy confirmed placement and alignment. Hemostasis was achieved and the mandible was replated. A drain was placed and the incision was closed in layers. An occiput to C-4 posterior segmental instrumentation and fusion with rib autograft was performed 1 week later to maximize stabilization.

**Postoperative Course.** The patient tolerated both procedures well without any complications. He was advanced to a regular diet, and his tracheostomy tube was removed 3 weeks postoperatively. The final pathology demonstrated an aneurysmal bone cyst. Postoperative imaging demonstrated gross-total resection of the lesion. The patient was placed in a hard collar for 3 months. At the 3 and 11 months’ follow-up, he was neurologically intact and pain free with the expected reduction in neck mobility. Imaging at that time demonstrated excellent alignment with no evidence of abnormal motion or instability (Fig. 8).

**Discussion**

Aneurysmal bone cysts are expansile lesions of vascular origin that are characterized by rapid expansion, adjacent bone destruction, and recurrence. In this case report, we present a primary bone cyst discovered in a patient with a classic constellation of symptoms that included progressive right-sided neck pain and weakness due to impingement on the spinal cord. Among children and adolescents, the spinal column is among the most common locations in which aneurysmal bone cysts develop. To our knowledge, this report documents the first time such a lesion required mandibular osteotomy as a result of significant lateral extension of the tumor in the

---

**Fig. 5.** Dissection of the sublingual space with preservation of the neurovasculature.

**Fig. 6.** Diversion of the tongue via a single suture, allowing for maximal visualization of the posterior pharynx on the right side.

**Fig. 7.** Successful placement of the cage in the C1–3 space occurs after successful dissection of the posterior pharynx and removal of the mass.

**Fig. 8.** Extension (A) and flexion (B) lateral standing radiographs of the cervical spine obtained at 11 months postoperatively. Stable alignment is seen without evidence of instability on flexion or extension views.
Midline mandibular osteotomy and anterior fusion

upper cervical spine. Given the high rate of recurrence after partial resection of these tumors and the need for anterior spinal reconstruction, we believed that the additional invasiveness of this procedure was justified.

In many cases, transoral and high cervical retropharyngeal approaches are sufficient for the treatment of high cervical lesions, and recent advances in endoscopic procedures have expanded the procedural armamentarium.2,5,7,12 Transoral and endoscopic techniques are typically favored given the accessibility of both intradural and extradural spaces from the clivus to typically the C-3 vertebra. However, a major disadvantage with these techniques is the limited exposure in the lateral direction, exposure that was necessary in this case. Furthermore, the range of access varies according to the patient’s age as well as the nasal and oral anatomy.1 In addition, spinal instrumentation and reconstruction are difficult through current endoscopic approaches. The high cervical retropharyngeal approach also has a reduced range of access, particularly to the upper cervical spine at and beyond the midline.8,11 On the other hand, the transmandibular approach has the greatest and widest exposure, which is critical in cases in which gross-total resection and anterior spinal reconstruction are needed. The predominant disadvantages of this approach are infection due to violation of the oral mucosa, extensive dissection and osteotomy of the mandible, and the need for tracheostomy to protect the airway. The vagus, accessory, and hypoglossal nerves as well as the trachea and major vessels are of particular concern, but as demonstrated in the current report, the risk of complications can be minimized by isolating each structure.9 Another alternative for a lesion with less lateral extension is a direct midline approach using a midline mandibulotomy and a midline incision to split the tongue.

Hall and colleagues initially described the midline mandibulotomy for spine surgery. It is typically reserved for lesions for which less invasive approaches do not provide sufficient exposure for resection and/or anterior spinal reconstruction.6 Its application has been recently reported for a similar procedure in an adult with sarcoma.3 To our knowledge, however, it has not been reported in a pediatric patient with an aneurysmal bone cyst of the upper cervical spine.

Conclusions

In summary, even in pediatric patients, the transmandibular approach for the treatment of upper cervical spine lesions is an effective method of maximizing exposure for complex lesions in which maximal resection and spinal reconstruction are needed.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper. Michael McDowell is a recipient of a Clinical Research Fellowship from the Doris Duke Charitable Foundation.

Author contributions to the study and manuscript preparation include the following. Conception and design: Anderson. Acquisition of data: Anderson. Drafting the article: Anderson, McDowell, Hanft. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Anderson. Administrative/technical/material support: Anderson.

References


Accepted March 3, 2014.

Please include this information when citing this paper: published online April 4, 2014; DOI: 10.3171/2014.3.PEDS13511.

Address correspondence to: Richard C. E. Anderson, M.D., The Neurological Institute, 710 W. 168th St., Rm. 213, New York, NY 10032. email: rca24@columbia.edu.