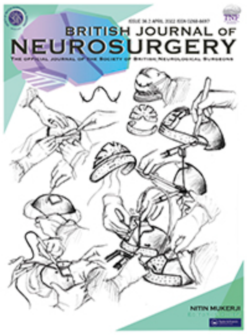

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Anatomical aspects and technical note of a modified retropharyngeal approach and reconstruction of the anterior occipitocervical junction

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TECHNICAL REPORT

Anatomical aspects and technical note of a modified retropharyngeal approach and reconstruction of the anterior occipitocervical junction

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ABSTRACT
Surgery to expose the anterior occiptocervical junction (OCJ) is exacting, and optimal approaches are debatable. The close proximity of vital structures and difficult surgical access present a unique challenge to treat lesions in this area. Routine access to the upper anterior cervical spine remains limited. The authors present a modified retropharyngeal approach and instrumentation in order to resect an exceptionally rare atypical rhabdoid teratoid tumor involving the craniovertebral junction. The technical aspects of this approach in anatomical perspectives are discussed in this article.

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Occipitocervical junction; retropharyngeal approach; atypical rhabdoid teratoid tumors; clivus; neurosurgery; Spine

Introduction

Surgery to expose the anterior upper cervical spine has always been seen as technically demanding and routine access to the upper anterior cervical spine remains limited.

A ventral lesion at the craniovertebral junction caused by tumors, infection or deformity may need surgical treatment. It may demand an anterior support for an anterior defect reconstruction and to restore stability, but because of intricate anatomy and significant neighboring structures, such reconstruction is extremely complex.

The purpose of this article to describe the anatomical aspects of the retropharyngeal approach on the upper anterior cervical spine modified by a mandibular angle osteotomy. A modified anterior occipitocervical fixation through a harm mesh attached to the clival edge for a clival screw placement in the sequence of an extensive resection of the upper cervical spine is also described.

The destructive lesion diagnosed in this young adult patient was an atypical rhabdoid teratoid tumor. These tumors are extremely uncommon in adults and are rarely found in spine. Only nine such cases have been reported in medical literature.

Surgical method

A 17-year-old female presented with a six-month history of neck pain and a one-month history of progressive generalized weakness. She was severely disabled with Karnofsky Performance Scale index (KPS) of 30 and strength on a 2/5 scale. There was no medical or family relevant information. Spinal magnetic resonance imaging (MRI) demonstrated an increasing mass compressing the upper spinal cord at the C1 and C2 level (Figure 1). Computed tomography (CT) images showed the lesion had eroded the clivus in part, as well as the C1 and C2 vertebrae (Figure 2). A metastatic workup was negative. The patient promptly underwent posterior spinal cord decompression and instrumented fusion using Axon occipital screws, lateral mass screws, and roads from the occiput to C5. Five days later, she returned for anterior resection.

Usually the patient is supine with the head rotated 30 degrees away from the surgeon and in extension. This head position is beneficial because it lifts the mandible up and away from the surgeon's line of sight to the field. Once she could not be positioned with the head extended and rotated, a modified extrapharyngeal approach with a mandibular angle osteotomy was planned to enlarge the exposure.

A transverse skin incision was started one fingerbreadth from the midline below the mandible and proceeded laterally and parallel to the lower edge of the mandible, curving around the angle of the mandible posteriorly over the mastoid process (Figure 3). The solution to adequate exposure was sharp dissection of each plane starting with the development of a wide subcutaneous flap on each side of the incision superficial to the platysma muscle. This wide dissection of the fascial planes helps to decrease the danger of neural and vascular injuries because no excessive force is required for retraction. At this point of dissection, steps were taken to avert injury to the mental branch of the facial nerve.

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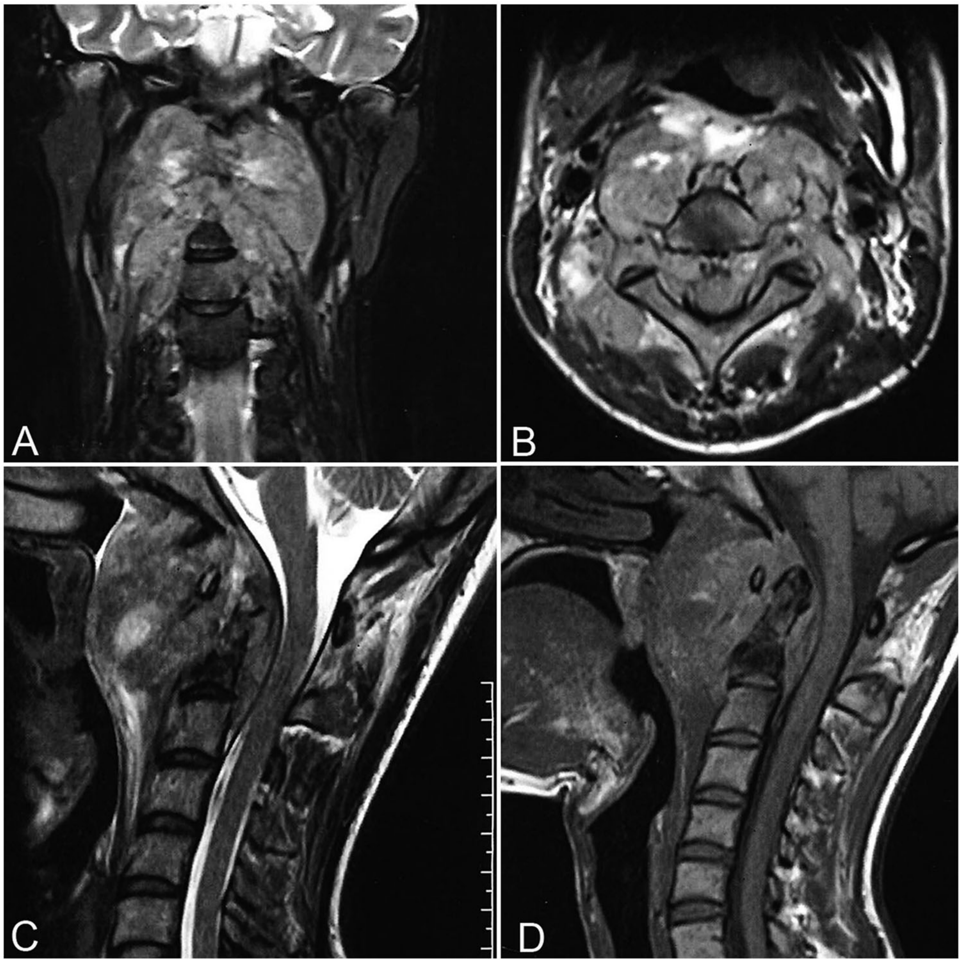

Figure 1. Preoperative coronal (A), axial (B) and sagittal (C) magnetic resonance (MR) T2-weighted images and precontrast MR sagittal T1-weighted image (D) demonstrating a heterogeneous tumor involving the inferior clivus, C1 and C2 vertebrae, the posterior aspect of the pharynx and the vertebral canal.

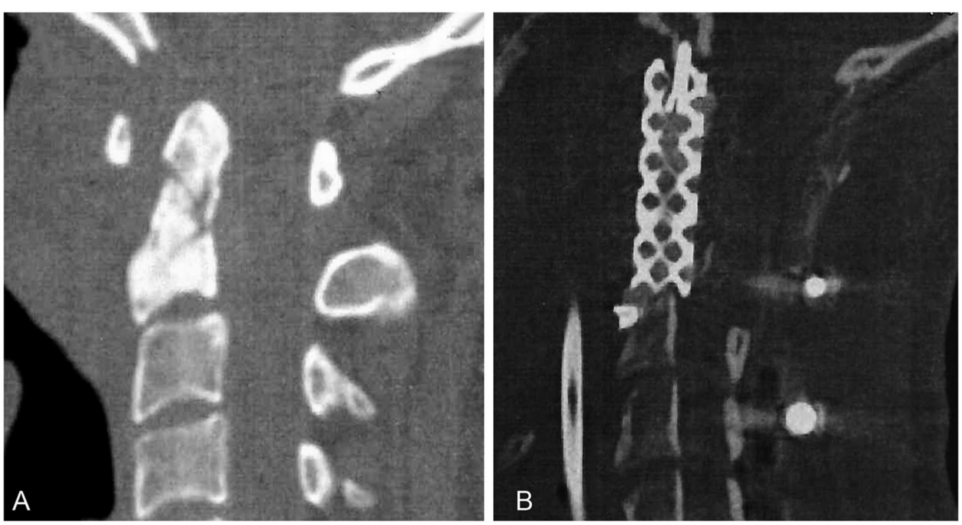

Figure 2. Preoperative sagittal computed tomography (CT) scan image demonstrating C1 and C2 infiltrating bony erosion with C1–C2 instability (A). Postoperative sagittal CT scan image showing replacement of the anterior column of high cervical spine by a titanium mesh cage filled with cement bone and fixed to the clivus (B).

Next, the layer of the platysma muscle was incised, and a retractor inserted into this plane. Then, the region of the mandible angle was exposed. The pterigomasseteric strap was incised and the masseter and medial pterigoyd muscles were separated from their attachments (they form the mandibular band, a muscular strap where the angle of the mandible lies and attaches the mandibular ramus onto the skull). Longitudinal and transversal osteotomies with conic drill were performed. As a reference to the osteotomy and to avoid damage to neurovascular bundle, the *antilingula* was identified. It was located approximately 2 cm from the posterior margin of the mandibular ramus and 1.5 cm from its inferior margin (Figure 4(A)). These maneuvers are not expected to result in postoperative malocclusion but demand posterior muscle reinsertion and bone reconstruction.

Before the osteotomy was complete and the bone fragment removed, two plates were fixed on it to facilitate the posterior reconstruction Then, the inferior aspect of the submandibular gland was exposed, and the fascial capsule was cut and dissected in line with the cutaneous incision. The facial vein was dissected and transected. The facial artery was superiorly retracted with the submandibular gland to uncover the tendon of the digastric muscle. This tendon runs parallel to the incision under the inferior edge of the submandibular gland. It was separated from its fascial sling at the hyoid bone and retracted toward the mandible. Then the hypoglossal nerve was exposed deep and parallel to the anterior digastric muscle and was dissected free. This maneuver facilitated the identification of the hypoglossus muscle and the cornu of the hyoid bone. The fascia over it was incised and the carotid sheath was retracted laterally. The pharyngeal constrictor muscles were retracted medially, and the prevertebral fascia was incised what exposed the retropharyngeal space (Figure 4(B)).

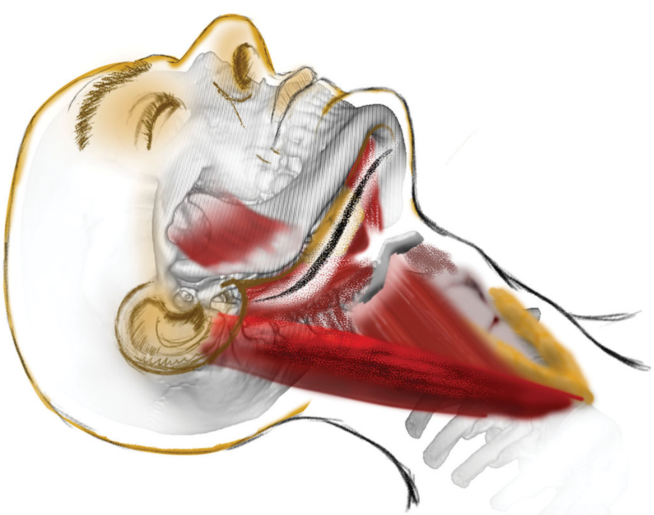

Figure 3. A transverse skin incision performed parallel to the lower limit of the mandible, curving around the angle of the mandible posteriorly across the mastoid process.

Finally, the midline of the spine was determined between the longus colli and longus capitis muscles. These muscles were separated from their medial attachments from C3 up to the pharyngeal tubercle region. The tumor involved the soft tissues and compromised the easy identification of the structures, including the anterior tubercle of C1. Retraction was crucial for exposure and access of the medial half of the C1 and C2 lateral masses and the anterior rim of the foramen magnum. The anterior rim of the foramen magnum was palpated and observed between the attachments of the longus capitis muscles. The C1 arch and odontoid removal were obscured by damages of the disease, and the transverse cervical ligament was a guide after C1 arch and odontoid removal. The vertebral canal was anteriorly decompressed.

For reconstruction, an 18 mm length screw was inserted close to the middle at the lower part of the clivus. The screw trajectory was placed in the cranial direction, perpendicular to the extracranial clivus. A titanium mesh cage filled with semi-solid bone cement with a diameter of 25 mm and appropriated length was set in such a manner that the superior end could be attached at the clival margin with the screw inside its center. After the bone cement solidified, the screw became an anchoring site for the cylinder stability (Figure 5(A)). We also chose bone cement instead of bone graft because expected postoperative radiotherapy could increase the possibility of occurrence of osteonecrosis. The caudal end was wedged into position over the superior epiphysis of C3 using a bone mallet to position the mesh. Parallel to the C3 endplate, another screw was inserted in order to prevent future migration of the cylinder through the endplate (Figure 5(B)).

Histopathological examination showed rhabdoid cells, vimentin diffusely positive and loss of immunohistochemical staining for INI1l (SMARCB1). The patient had an uneventful recovery, except for transitory dysphagia (for 2 weeks) related to pharyngeal traction . The KPS score was of 60, strength on a 4/5 scale in upper limbs and 3/5 in lower limbs. She was then transferred to the oncology department of another hospital, where she received radiotherapy treatment. There were no complications related to the craniovertebral fusion. The patient died after 12

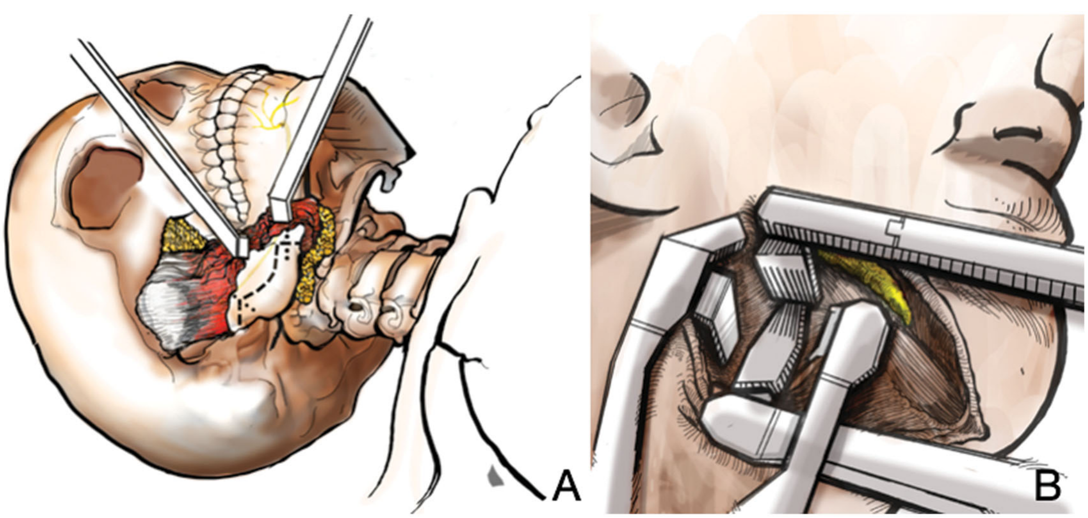

Figure 4. (A) Osteotomies with conic drill were performed. As a reference to the osteotomy and to avoid damage to neurovascular bundle, the *antilingula* was identified. It was located approximately 2 cm from the posterior margin of the mandibular ramus and 1.5 cm from its inferior margin; (B) The fascia was incised and the carotid sheath was retracted laterally. The pharyngeal constrictor muscles were retracted medially, and the prevertebral fascia was incised what exposed the retropharyngeal space.

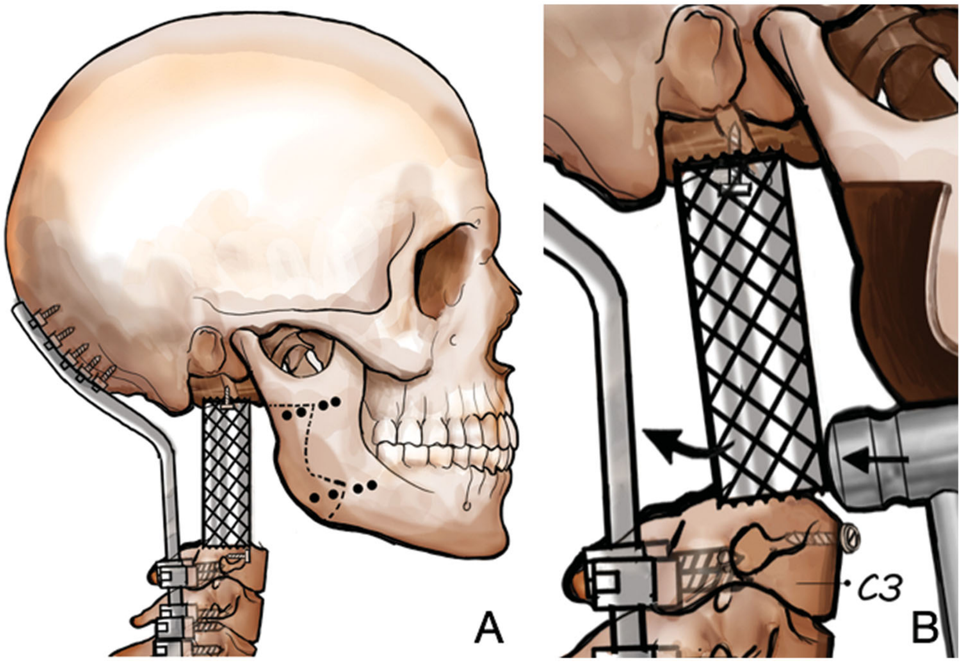

Figure 5. (A) For reconstruction, a screw was inserted close to the middle at the lower part of the clivus. A titanium mesh cage filled with semi-solid bone cement was set in such a manner that the superior end could be attached at the clival margin with the screw inside its center. After the bone cement solidified, the screw became an anchoring site for the cylinder stability; (B) The caudal end was wedged into position over the superior epiphysis of C3 using a bone mallet to position the mesh.

months due to clinical complications, secondary to local progression of the disease in soft tissues of the cervical region, despite of oncological and clinical management.

Discussion

A variety of mass lesions occur in the OCJ and can arise from bony and ligamentous or neural and vascular structures. Lesions, such as tumors, arthritis, infection, trauma and congenital abnormalities are observed in this region. Concerning primary osseous tumors occurring in occipitocervical region, the most common ones are chordomas, chondrosarcomas, fibrous dysplasia, eosinophilic granulomas, plasmocytomas, osteoblastomas and giant cell tumors.[1]

Atypical teratoid rhabdoid tumors (ATRTs) are extremely uncommon in adults and are rarely encountered in the spine; only nine such cases have been reported in medical literature.[2,3] These tumors typically occur in pediatric patients younger than 3 years old and are found intracranially but should also be considered in the differential diagnosis of children presenting with a new spinal mass.[4] Different multimodal strategies combining surgery, radiotherapy, and chemotherapy (with or without stem cell support) have been used. Chemotherapy regimen has used multiple agents in combination with radiotherapy and definition of some predictive biomarkers might personalize therapy. Nevertheless, ATRTs are consistently marked by multiple relapses and very poor prognosis.[3,4]

All these lesions carry the risk of high-level cervical spinal cord lesion and instability. When lesion resection and decompressions are required, reconstruction is exceedingly complicated due to vital adjacent structures and its complex anatomy.[5]

Surgery to expose the upper anterior cervical spine is taxing, and ideal approaches are debatable. Several anterior approaches have been proposed for the OCJ. Transoral approaches provide direct access. The extended transoral approaches may be classified as with upper extension by maxillotomy and lower extension by transmandibular approach. The extended maxillotomy provides access of the clivus to lower border of C2 vertebra. The transmandibular approach provides access down to C4 vertebral bodies but more inferior access requires also a transglottic approach.[1] Currently, the endoscopic approach to the clivus has become well-established alternative to traditional approaches. However, risk of infection is concerning, as these techniques are performed through the mouth and dissection of oropharyngeal mucosa and the exposure can be insufficient for arthrodesis.

Additionally, the retropharyngeal approach, preserving the mucosa of the oropharynx, could be used for the anterior craniocervical reconstruction.[6] This high anterior cervical retropharyngeal approach allows for good, safe access in the area of the superior cervical spine, and anterior stabilization, including C1-3 It can be performed using this method without severe morbidity.[7] It was first described by J.R. de Andrade and Ian Macnab. These authors reported an extension of former approaches in order to access the basiocciput and the ventral aspect of all the cervical vertebrae, by performing an anterior occipitocervical fusion.[8]

The recommended patient's position is with the head extended and rotated contralateral to the side of approach. The surgeon perspective is a rostrally oblique direction, and unwavering attention to this is necessary to preserve appropriate orientation. On the reported case, there was already a posterior occipitocervical fixation, what precluded ideal head positioning (Figure 6). The mandibular angle surgical access and osteotomy were planned because it can be performed easily during the usual approach and increases the surgeon's angle of vision (Figures 7 and 8).

One must be aware of the safe transversal distance between the clival screws and look for a 'safety zone'. The lateral borders are limited between the hypoglossal nerves, the abducent nerves,

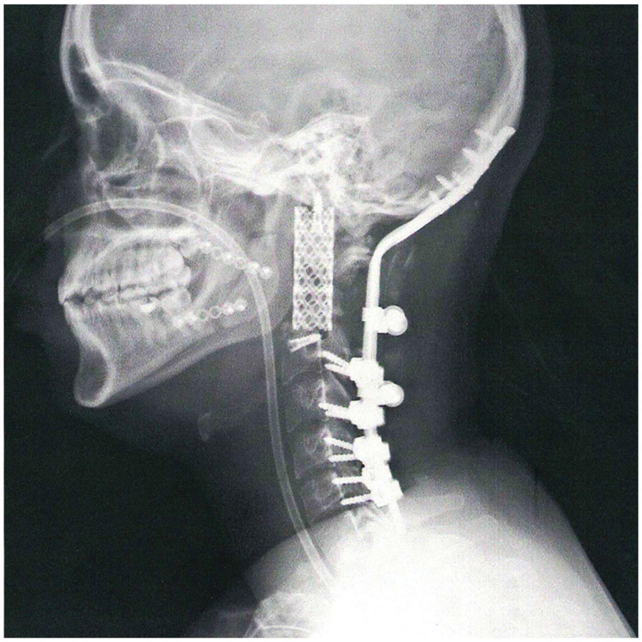

Figure 6. Postoperative lateral view radiograph demonstrating anterior and posterior occipitocervical fixation with osteotomy of the angle of the mandible.

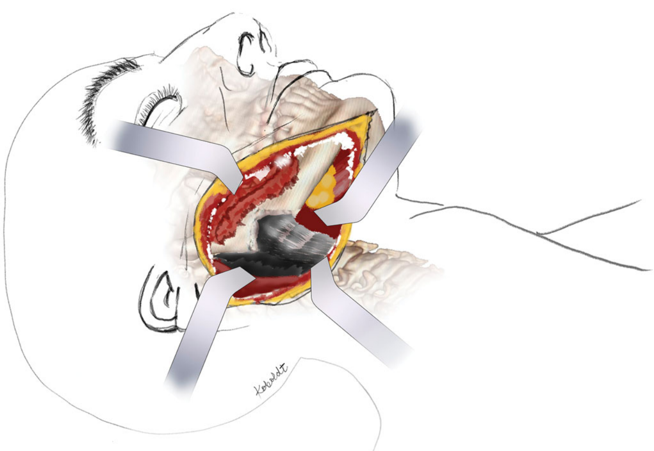

Figure 7. The mandibular angle surgical access and osteotomy were performed to improve the surgeon's viewing angle.

the cavernous and petrous segments of the internal carotid artery, and the optic canals. This limits the opening of the inferior clivus to 34 mm regarding the distance between the two hypoglossal canals. This opening becomes narrow as one moves cranially on the clivus with the distance between the abducent nerves at 20 mm.[9] The vertebral arteries path 14 to 15 mm on each side lateral to the anterior atlantal tubercle level.[1]

Anatomical landmarks of the midline skull base and upper cervical vertebrae include the anterior atlantal tubercle and the pharyngeal tubercle. Another key technical point includes an appropriate anatomical dissection of each fascial plane in the submandibular triangle cranial to the hyoid bone and lateral to the superior pharyngeal constrictor muscle, eventually accessing the space between the vertebral bodies and the prevertebral fascia. The superior limit of this exposure is the attachment of the superior pharyngeal constrictor muscle to the pharyngeal tubercle and, at this point, the key to avoid disorientation is attention to anatomical landmarks. Neuronavigation systems are important technological tools to orientate the surgeon, which is more relevant when anatomical landmarks are not evident. Many imaging modalities such as CT, MRI and intraoperatively acquired ultrasound are being employed as a real time image guide.[9]

The upper ventral cervical spine has typically been reconstructed with common mesh cage. Not long ago, a clival screw and plate fitted to the osseous anatomy of the craniovertebral junction was developed for this pathologic status.[5,10] The difficulty in using such device in this case is related to the fact that it had been planned for C1 mass and for low clivus fixation, and in the present case there was no integrity of these structures.

All of the approaches to the clivus and upper cervical spine carry a risk of a high rate of complications, such as bleeding, infection, dysphagia, plate dehiscence, cerebrospinal fluid leakage and meningitis.[11] The problems that are most found with this approach are dysphagia and dysphonia.[8,12] The pharyngeal and laryngeal branches of the vagus and glossopharyngeal nerves originate from the parent trunk just below the jugular foramen and are carried forward with the pharynx. The traction stretches these nerves, and probably is a factor in temporary hoarseness and dysphagia.[8] Retraction of the superior pharyngeal constrictor muscle can also lead to protracted dysphasia. The hypoglossal nerve is especially vulnerable to traction injury.

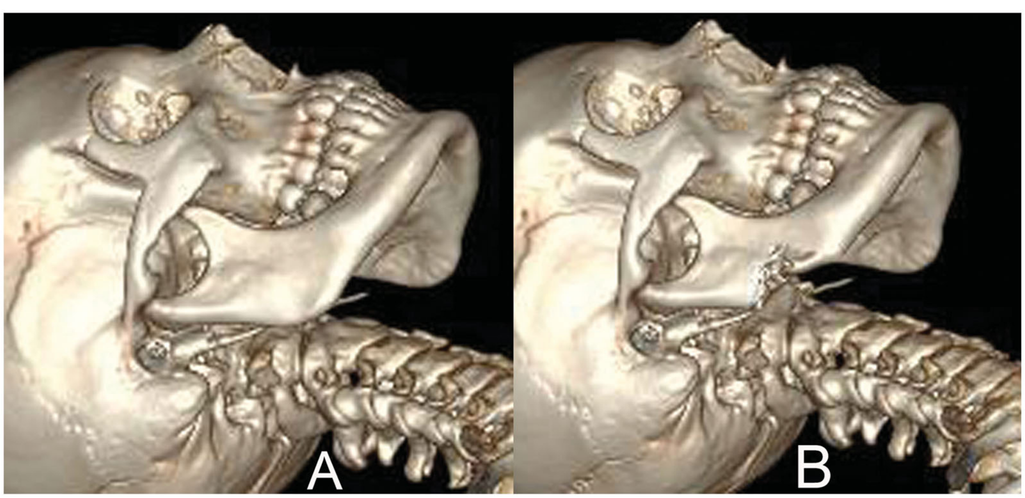

Figure 8. Cranial and spine *Volume Rendering* tridimensional reconstructed image demonstrating a simulated surgeon's view of the craniovertebral junction with (A) and without (B) the angle of the mandible. When the carotid sheath is retracted laterally, the retropharyngeal space is opened; after the pharyngeal constrictor muscles are retracted medially and the prevertebral fascia opened.

We decided the strategy of a 360-degree excision of the tumor and fusion of the OCJ and the higher cervical spine based on some topics. The lesion was encasing the vertebral arteries and the spinal cord causing rapid neurological deterioration. We believe that a first posterior approach would fasten and make easier to stabilize the OCJ and decompress the spinal cord on its posterior and lateral aspects, associated to a better proximal and distal control of vertebral arteries. In the decision-making process for a second stage we did not find relative contraindications to aggressive resection such as poorly controlled systemic diseases, extensive intradural involvement, high radiosensitive tumors or anticipated inability to achieve radical extirpation.[13] The decision of whether to use the transoral or the retropharyngeal approach was based on our opinion the retropharyngeal route provides the most direct access to extradural pathology with a more suitable angle to insert the clival screw and to insert the mesh cage between C3 and clivus as it was described. Moreover, we think there would be less potential contamination from pharyngeal cavity and the extensive involvement of soft tissues and longus colli muscles could compromise the closure of the posterior pharyngeal wall.

Finally, we considered some drawbacks of transoral approach. A tracheostomy must be performed prior to lower extended transoral surgeries. Reconstruction after these approaches can cause a sort of specific complications such as oronasal fistulae, temporomandibular dislocation, velopharyngeal dysfunction.[13,14] These routes require specialized instruments other than the usual devices for anterior cervical spine access. Dermal fat grafts, fascia lata grafts and mucosal flaps may be needed. The floor of the mouth healing can be complicated by hematoma formation, scarring and swallowing difficulties. In general, the spine surgeons are not used to deal with this kind of complications.

The primary objective remains the resection of the lesion, and this should not be compromised by inappropriate exposure or technique. The surgeon's experience with performing one or another technique must be considered in the final choice of which approach to use.[15]

Once the patient was suffering of intractable pain and presenting fast progressive loss of strength, we decided to operate on within 48h after admission in order to spinal cord decompression and stabilization. We considered it was an emergency situation and we could not have the imunohystochemical analysis result in such a short time. The posterior approach was sufficient to achieve pain relief and neurologic stabilization. Then we had enough time to plan the more complex anterior approach and to obtain the biopsy results.

Conclusion

The high anterior cervical retropharyngeal approach to the upper cervical spine is an advantageous method that affords direct and large exposure for fusion and anterior decompression of the upper cervical spine. This approach is another option to the transoral approach to the ventral occipitocervical union. A modified approach with mandibular angle osteotomy can be considered when the patient's head cannot be extended and rotated in order to increase the angle of sight to the prevertebral area. The occipitocervical fixation through a bone cement filled mesh cage at the clival edge fixed by a clival screw was successful for anterior reconstruction in the present case.

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Disclosure statement

No potential conflict of interest was reported by the author(s).

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