Review Article
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SURGICAL MANAGEMENT OF ANTERIORLY LOCATED TUMORS AT THE CRANIOVERTEBRAL JUNCTION: ADVANCES AND CHALLENGES

TRATAMENTO CIRÚRGICO DE TUMORES LOCALIZADOS ANTERIORMENTE NA JUNÇÃO CRANIOVERTEBRAL: AVANÇOS E DESAFIOS

MANEJO QUIRÚRGICO DE TUMORES DE LOCALIZACIÓN ANTERIOR EN LA UNIÓN CRANEOVERTEBRAL: AVANCES Y DESAFÍOS

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ABSTRACT

Primary tumors affecting the craniovertebral junction (CVJ) range from benign to malignant lesions. The CVJ tumors are challenging due to their critical anatomical location adjacent to neural and vascular structures. Therefore, more detailed planning of the surgical approach is required to achieve complete tumor resection, reduce risks and offer better clinical outcomes. Several surgical approaches have been described for removal of CVJ tumors. Additionally, the removal of anteriorly located tumors at the CVJ is particularly complex and requires precise and demanding surgical strategies. The transoral, anterolateral, labiomandibular, and circumglossal approaches are usually indicated to access these tumors. However, these surgical approaches are still poorly reported in the literature. In this article, we describe the main approaches to access the anterior tumors at the CVJ, the surgical steps, and their main challenges. ***Level of Evidence II; Review Article***.

Keywords: Cervical spine tumors; Craniovertebral junction; Transoral approach; Anterolateral approach; Labiomandibular approach; Circumglossal approach.

RESUMO

Os tumores primários que afetam a junção craniovertebral (JCV) geralmente incluem lesões benignas e malignas. Os tumores da JCV são desafiadores devido à sua localização anatômica crítica ao redor de estruturas neurais e vasculares. Portanto, é necessário um planejamento mais detalhado da abordagem cirúrgica a fim de obter uma ressecção completa do tumor, reduzir riscos e oferecer melhores desfechos clínicos. Diversas abordagens cirúrgicas têm sido descritas para a remoção desses tumores. Além disso, a remoção de tumores localizados anteriormente na JCV é particularmente complexa e requer estratégias cirúrgicas adequadas e desafiadoras. As abordagens transoral, anterolateral, labiomandibular e circunglossal são geralmente indicadas para acessar esses tumores. No entanto, essas abordagens cirúrgicas ainda são pouco relatadas na literatura. Neste artigo, descrevemos as principais abordagens para acesso aos tumores anteriores da JCV, as etapas cirúrgicas e seus principais desafios. ***Nível de Evidência II; Artigo de Revisão***.

Descritores: *Tumores da coluna cervical; Junção craniovertebral; Abordagem transoral; Abordagem anterolateral; Abordagem labiomandibular; Abordagem circunglossal.*

RESUMEN

Los tumores primarios que afectan la unión craneovertebral (UCV) generalmente incluyen lesiones benignas a malignas. Los tumores de la UCV representan un desafío debido a su ubicación anatómica crítica, que rodea estructuras neurales y vasculares. Por lo tanto, se requiere una planificación más detallada del abordaje quirúrgico para lograr una resección completa del tumor, reducir los riesgos y ofrecer mejores resultados clínicos. Se han descrito diversos abordajes quirúrgicos para la extirpación de tumores de la UCV. Además, la extirpación de tumores de localización anterior en la UCV es particularmente compleja y requiere estrategias quirúrgicas adecuadas y complejas. Los abordajes transoral, anterolateral, labiomandibular y circungloso suelen estar indicados para acceder a estos tumores. Sin embargo, estos abordajes quirúrgicos aún están poco documentados en la literatura. En este artículo describimos los principales abordajes para acceder a los tumores anteriores a nivel de la UCV, los pasos quirúrgicos y sus principales desafíos. ***Nivel de Evidencia II; Artículo de Revisión***.

Descriptores: *Tumores de la columna cervical; Unión craneovertebral; Abordaje transoral; Abordaje anterolateral; Abordaje labiomandibular; Abordaje circungloso.*

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INTRODUCTION

The cervical spine represents the most common site for primary spinal tumors. Based on their anatomical location, these tumors can be divided into the following three groups: 1) intramedullary tumors, usually benign, such as gliomas and ependymomas; 2) intradural extramedullary tumors, commonly benign, including meningiomas, schwannomas, and neurofibromas; and 3) extradural tumors, commonly represented by malignant tumors, such as chordomas, chondrosarcomas, and osteosarcomas.[1-3] These tumors account for less than 10% of all primary spinal tumors, resulting in local compression, with pain being the most frequently reported symptom.

The craniovertebral junction (CVJ) is the anatomical region that comprises the occipital bone and the two cervical vertebrae (C1 or atlas and C2 or axis).[4] The differential diagnosis for CVJ tumors is based on computed tomography (CT) scans and magnetic resonance imaging (MRI), which are essential for surgical planning. Sometimes a biopsy is required for a definitive histopathological diagnosis.[1,5] Despite being rare and mostly benign, the CVJ tumors are challenging due to their critical anatomical location adjacent to neural and vascular structures, which may result in neurological deficits and progressive functional decline.[6,7]

Several surgical approaches are described to access these tumors. However, the technical complexity of achieving tumor-free resection margins with minimal morbidity represents a major challenge to be considered, particularly for anteriorly located lesions at the CVJ. This review study, approved by the institutional board (CAAE: 56429322.7.0000.5273), aims to describe the most suitable approach to access anteriorly located tumors at the CVJ, as well as the key technical aspects of each approach and challenges in surgical management.

Surgical approaches at the CVJ

Resection with tumor-free margins has been associated with prolonged disease-free survival compared with intralesional resection and radiotherapy.[6-10] However, this surgical management is complex and challenging, requiring a detailed analysis of vertebral artery involvement and tumor extension for decompression and stabilization of the CVJ.[11-13] The posterior surgical approach is the most commonly described for the CVJ.[6,14,15] However, the anterior surgical approaches are the most indicated to access tumors resulting in ventral compression.[16]

Transoral approach

The transoral approach was first described in 1947 by Thomson and Nagus to treat retropharyngeal infections.[17] In 1951, Scoville and Sherman employed it for the treatment of basilar impression.[18] The use of this approach for tumor resection was only reported in 1957 by Sothwick and Robinson, who described the excision of a C2 osteoma.[14,19]

The transoral approach is commonly used to access the upper cervical spine and is preferentially used to treat extradural lesions located ventral to the CVJ.[20-25] This approach provides the most direct access to lesions involving the upper cervical spine and encompasses the region from the lower third of the clivus to the C2-C3 interdiscal space.[21,26] One major advantage is its relatively avascular access in the midline, offering direct access and a shorter learning curve, even with the raphe of several layers of mucosa and fascia.[16,27-29] Since most chordomas are midline tumors at the CVJ, the transoral approach is often used.[20]

However, this approach has some disadvantages including the possible need for tracheotomy prior to surgery, risk of infections, dehiscence of the pharyngeal suture, edema and/or necrosis of the tongue, vertebral artery injury, and cerebrospinal fluid fistula.[20,29-31] Another disadvantage involves accessing intraspinal tumors due to limited surgical exposure of the C1–C2 joints.[13,16] Also, the transoral route allows an anterior approach to the upper cervical spine through a retropharyngeal or retrovascular access.[15,32-36] Furthermore, the limited operative view of the spinal cord during tumor removal increases the risk of local injury.[16] The endoscopic technique can be combined with the transoral approach to increase anatomical exposure with less local aggression, in addition to providing increased rostral exposure and avoid complications related to sectioning the soft palate.[24,37]

Surgical details of the transoral approach

The surgical steps for the transoral approach will be briefly described as follows (Figure 1). The patient is positioned in the prone position and the skull is fixed with the Mayfield in slight extension. Afterwards, a slight inclination in the Trendelenburg position allows for the most rostral exposure. Transoral retractors are applied to retract the mandible, exposing the oropharynx. Oral antiseptic preparation is performed with chlorhexidine and antibiotic prophylaxis. Subsequently, a midline incision is made using the atlas tubercle as a guide. In case of a more cephalic approach, velotomy can be performed for a wider exposure. The incision continues 3 cm distal to the C2-C3 disc space. The longus colli and capitis muscles are laterally retracted to expose the anterior longitudinal ligament. As an option, the pharyngeal layer could be addressed by an *open-door* technique allowing access more laterally to C1-C2 joints and tumor resection, with the advantage of performing anterior instrumentation and reduce tension in the soft tissues. At this stage, the surgeon is able to remove the ventral tumors guided by imaging, and a single layer or double closure is performed.

Anterolateral retropharyngeal approach

The anterolateral retropharyngeal approach offers an extramucosal access route from the skull base to the C2-C3 intervertebral disc, occurring above the hyoid bone and medial to the neurovascular bundle of the neck in the parapharyngeal space.[20,32,33,35,38-40] The classic anterolateral approach was first described by Smith

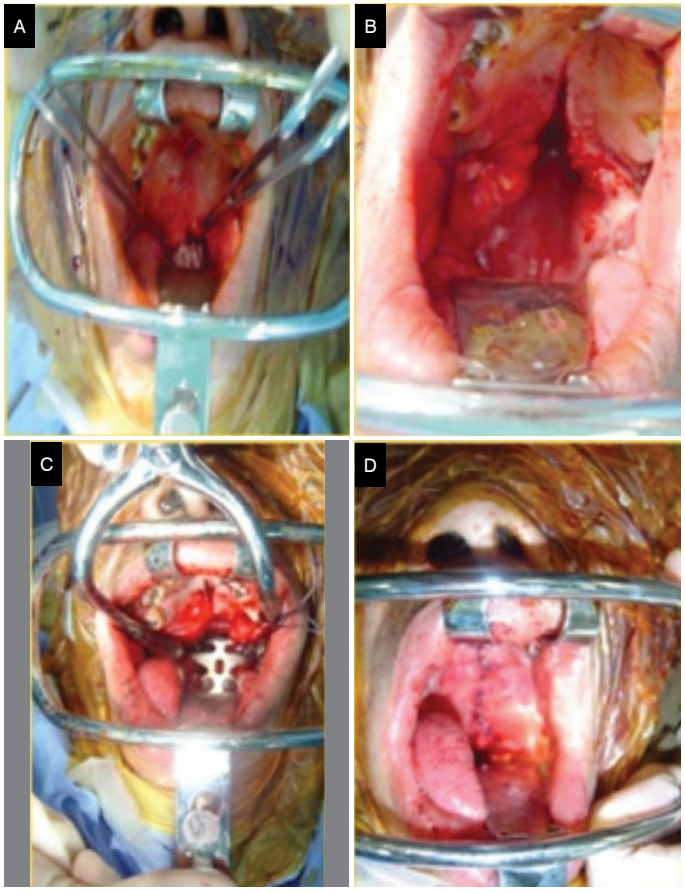

Figure 1. Illustrative case showing the transoral approach. A) Transoral approach. B) Velotomy. C) Instrumentation with Harms Transoral Plate. D) Double layer closure.

and Robinson.[41] Modifications have been proposed by McAfee et al. (1959) to access the upper cervical spine in patients with osteochondroma.[33] Whitesides et al. described, in 1966, the approach proposed by Arnold K. Henry, being a longitudinal access along the anterior border of the sternocleidomastoid muscle (SCM) to the base of the mastoid bone. This approach allows *en bloc* resections of malignant tumors at the CVJ, exposing the transverse processes of all vertebrae.[42] In 1982, Böhler described the anterolateral approach for patients with odontoid pseudoarthrodesis.[43] In 1987, Lesoin et al. proposed a bilateral anterolateral approach to access tumors that advanced to both lateral ends of the cervical spine.[44] More recently, Visocchi et al. (2024) simplified the technique with three surgical steps and avoided the removal of critical anatomical structures, reducing surgical time while minimizing risk of complications.[32]

As advantages, this access allows decompression and anterior fusion to occur at the same surgical time. Also, the anterolateral approach preserves the mucosal layers and also provides satisfactory access to the anterior and lateral sides of the CVJ, as well as the anterior fixation can be performed at the same surgical time.[32,45] However, the approach requires meticulous dissection around critical anatomical structures, increasing the risk of damaging the hypoglossal and superior laryngeal nerves and the marginal mandibular branch of the facial nerve.[32,46]

This approach provides an approximate 25° craniocaudal and 42° laterolateral working angle, allowing exposure from the anterior border of the foramen magnum to the inferior endplate of C2.[32] Laterolateral exposure is partial, which exposes the entire contralateral C1 and C2 joint and most of the ipsilateral joint (approximately 70%).

Surgical procedure of the anterolateral approach

To perform this approach, the surgical procedure involves positioning the patient in a supine position with a Mayfield support, with slight extension of the cervical spine. Subsequently, the incision is made on the appropriate side, approximately 2 cm below the lower border of the mandible, extending anteriorly for 3 cm in the direction of the skin tension line to avoid injuring the inferior marginal mandibular branch of the facial nerve. Afterwards, a lower curvilinear incision is made, respecting the anterior edge of the SCM, with a boomerang-shaped incision.

The outer layer of the platysma is exposed, the fibers are opened and the myocutaneous flap is reflected anteriorly. The external jugular vein, located immediately below the platysma, is ligated. The submandibular gland is visible inferior to the vein, with its cranial part covered by the mandibular arch. The vascular supply from the facial artery is then ligated. Wharton's duct is ligated and the hypoglossal nerve is dissected. Finally, the submandibular gland can be removed *en bloc*.

Next, the intermediate tendon of the digastric muscle is sectioned and mobilized, and then the stylohyoid muscle is detached from the hyoid bone. This resulting space is delimited by the lateral border, which contains the main vessels of the neck. The superior border is composed by the hypoglossal nerve and the inferior border by the facial artery. The surgical procedures for the anterolateral approach are shown in Figure 2.

Labiomandibular approach

Roux described, in 1839, an approach with central division of the lower lip and mandibular osteotomy to access tumors of the tongue. The transmandibular approach combined with glossotomy was first proposed by Trotter in 1920.[47,48] Hall et al. used a mandible and tongue-splitting approach to reconstruct the spine with cervical kyphosis after tumor resection.[49,50] Wood was a pioneer in using this access for tumor resection.[51]

The labiomandibular approach with glossotomy is a surgical option that allows extended access beyond the CVJ and distally to the C2-C3 disc space, being especially useful in patients with restricted mandibular opening (<3 cm).[32,49,52] Furthermore, it allows lesions to be reached up to the C5 disc space. Advantages include a direct and relatively avascular route to the spinal cord. Also, this approach provides adequate anatomical exposure with a larger surgical field of view than the transoral approach.[49,50,53] However, surgical site infection secondary to wound dehiscence remains one of the major reported complications. Other disadvantages include oral and velopalatine incompetence, dysphagia, malocclusion, and tracheostomy-related complications. Despite being a technically demanding procedure, the resulting deformities and functional impairments are minimal, and this is the preferred technique in our center.[32,52]

Surgical procedure of the labiomandibular approach

First, the patient is placed in the supine position followed by a tracheostomy. Then, a midline incision is made from the lower lip to the hyoid bone in a curvilinear fashion. The incision becomes deeper until exposure of the jawbone. Therefore, a mandibular osteotomy is performed in stages to facilitate subsequent reconstruction. Soft tissue dissection follows the midline to the base of the tongue. At this stage, the tongue is divided at its central raphe and the oropharyngeal cavity is exposed, in which the separation of the structures ensures adequate visibility of the surgical field.

The incision of the pharyngeal wall occurs in an *open-door* or Harms-Schmelzle technique, in which an open-door flap is made. This procedure improves access to the lateral aspects of the C1–C2 joints and helps preserve the arterial blood supply as it can be covered by a metal prosthesis.[49,54] Next, the clivus and upper cervical spine are exposed. Closure is performed in order to reconstruct the previous anatomy. An orogastric tube is placed prior to jaw fixation. These surgical steps of the labiomandibular approach are shown in Figure 3.

The jaw is reduced and fixed with a pre-molded plate and screw. After fixation, the postoperative bite force is tested, and the process is completed by suturing the subcutaneous tissue and skin. (Figure 4)

Circumglossal approach

A wide approach may be necessary for tumors located more inferiorly, allowing for their complete removal.[55] The circumglossal approach was first described by Biller et al. (1981) and later refined by Ammirati et al. (1993).[53,56,57] The main indication for this approach is to access tumors affecting the medial region with expansion to

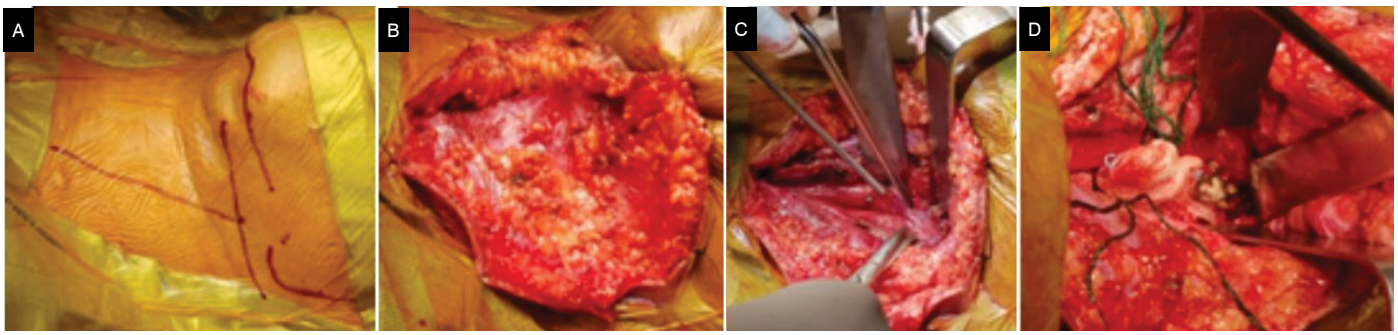

Figure 2. Illustrative case showing the anterolateral approach. A) Skin plannig approach with inferior incision anterior to SCM to allow tumor resection from C1 to C4 vertebrae. B) Careful dissection to avoid marginal mandibular branch of facial nerve. C) Identification of hipoglossal nerve. D) Image showing revision of recurrence cordoma case.

the lateral compartment.[55,58] This approach should be considered to access tumors that compromise both the anterior and lateral compartments, as well as for tumors extending to the subaxial cervical spine.[55] This approach offers visualization from the skull base to C7 vertebra, in which both *en bloc* resection of the tumor and reconstruction of the cervical spine can be performed, achieving the widest anatomical exposure of the CVJ.[55,56,58] However, this approach should be used with caution due to complications related to greater anatomical exposure, such as reduced tongue motricity and swallowing deficits.

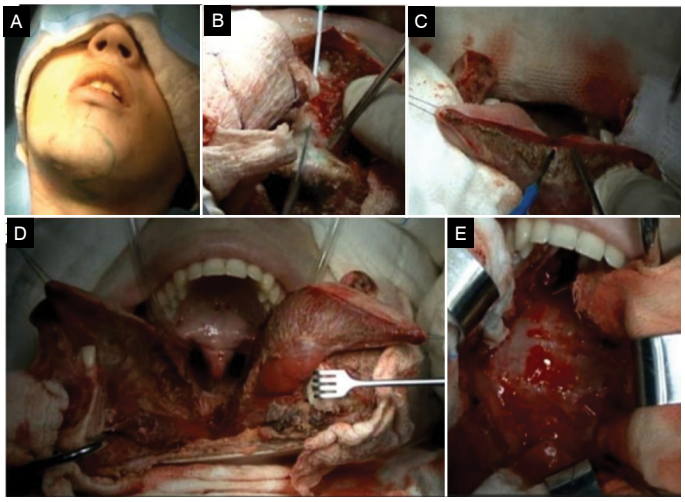

Figure 3. Illustrative case showing the labiomandibular approach. A) Image showing the marking of the skin incision line. B) Mandibular osteotomy. C) Central division of the tongue. D) Mandibular and tongue separation. E) Surgical field.

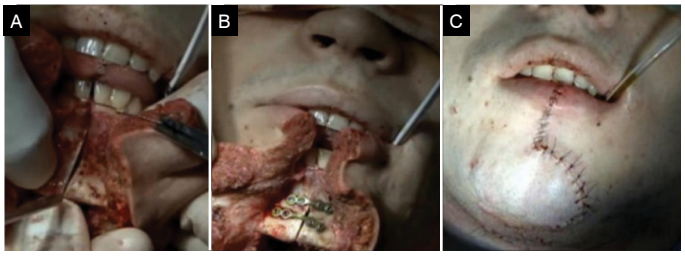

Figure 4. Illustrative case showing the final steps of the labiomandibular approach. A) Anatomic reduction of the mandible. B) Fixation with plate and screw. C) Final patient's postoperative appearance.

Surgical procedure of the circumglossal approach

To perform this procedure, the patient is tracheostomized and the skull is fixed in the Mayfield. Then, a curvilinear incision is made from the mastoid apex to the center of the lower lip, medially and inferiorly along a skin crease to the mentum and through the lip. The subplatysmal flaps are elevated exposing the submandibular gland and adjacent tissues. A dissection performed above the hyoid facilitates identifying the internal carotid artery, hypoglossal and lingual nerves. The flap dissection is continued superiorly until the mandible is exposed, and an osteotomy is performed in the center of the mentum. The dissection advances to the floor of the mouth and the caudal part of the incision extends into the hypopharynx, passing laterally to the orifice of the eustachian tube. The tensor muscle of the auditory tube is sectioned and, by retropharyngeal dissection, the longus capitis is identified and detached to expose the prevertebral fascia. After maximum tumor resection, reconstruction is initiated with closure of the tongue base and mandibular fixation. Finally, the platysma is approximated and a drain can be placed in the neck, where the procedure is completed with the hermetic closure of the subcutaneous tissue and skin. These surgical steps of the circumglossal approach are shown in Figure 5.

The Table 1 summarizes the main advantages and disadvantages of each surgical approach described in this review, as well as the indication of the most appropriate technique according to the spine level of surgical access.

DISCUSSION

Achieving a free margin in tumor resection may be the only curative treatment for tumors located at the CVJ.[6,8,9] Regarding disease-free survival, more promising results are found compared to intralesional resection and radiotherapy.[6,8,10] Nonetheless, performing an *en bloc* resection is a complex task. A critical assessment of vertebral artery involvement and tumor extent is essential for this purpose.[11,12] Silva et al. reported that CVJ exposure should provide an optimal view of the tumor lesions and, through the same access route, should facilitate decompression, tumor resection, and spinal stabilization.[49]

In this review, we described the four main surgical approaches to access anteriorly located CVJ tumors. No single approach is

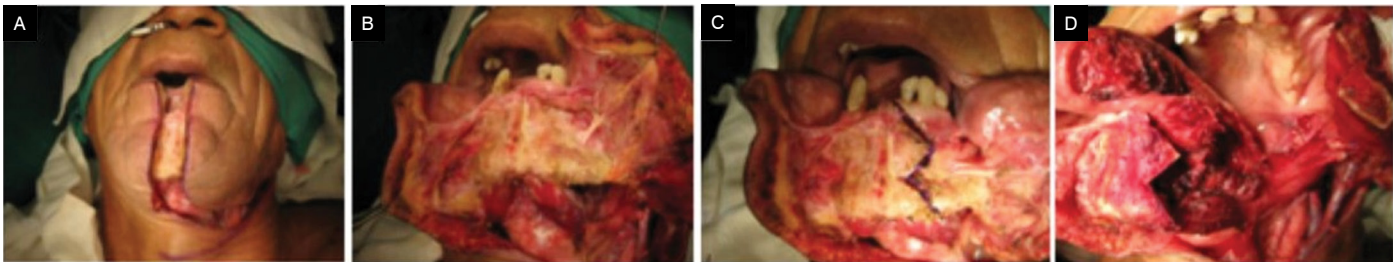

Figure 5. Illustrative case showing the circumglossal approach. A) Skin and subcutaneous incision. B) Subperiosteal detachment of the mandible. C) Mandibular osteotomy. D) Anterolateral view of the CVJ.

Table 1. Summary of the surgical approaches for removal of anteriorly located tumors at the CVJ, their advantages and limitations.

Parameters	Transoral	Anterolateral	Labiomandibular	Circumglossal
Spine level of surgical access	Clivus to C2-C3 disc space	Skull base - upper edge of hyoid bone	Clivus to C5	Skull base - C7
Advantages	Direct and avascular access	Access that does not injure the mucosal layers	Avascular access without the need for excessive removal of important anatomical structures	Access with greater anatomical exposure of the CVJ; extensive anterior and lateral approach.
Disadvantages	Infections, post-operative and nutritional care, wound healing, cerebrospinal fluid fistula	Risk of injury to the hypoglossal nerve, superior laryngeal nerve, marginal mandibular branch of the facial nerve	Infections, suture dehiscence, tracheostomy need	Meningitis, hearing loss, serous otitis media, and swallowing deficit.

applicable for tumors located in this anatomical region; therefore, the extent and anatomical location of the tumor should guide the choice of access route.[20] Midline ventral tumors with a craniocaudal extension of less than 5 cm and a lateral extension of 3 cm may be better approached via the transoral access. Furthermore, the transoral approach allows reconstruction and fixation of the upper cervical spine through the same access and in the same surgical time.[20]

For CVJ tumors affecting children, patients with inadequate oral cavity opening or tumors extending beyond the C2-C3 disc, the transmandibular approach is the most appropriate indication. Despite its radical intraoperative appearance, this approach offers excellent exposure of medial tumors that extend caudally beyond the limits of the CVJ. Arbit and Patterson described that this approach offers a larger surgical field in both sagittal and coronal plane than the transoral access.[52] Additionally, the incisions heal with minimal functional and aesthetic deficits, being well tolerated by patients.

Visocchi et al. reported higher complication rates associated with transmucosal approaches (transoral and transmandibular).[32] The authors also described that an anterolateral transmucosal approach is indicated when the tumor mass occurs laterally to the CVJ. The anterolateral approach allows decompression, tumor removal, and reconstruction in a single stage. Furthermore, this approach exposes the entire anatomical area of the upper cervical spine in the craniocaudal direction, the entire contralateral facet joint, and more than half of the ipsilateral joint, making it suitable for midline tumors. For tumors extending to one side of the upper cervical spine, a contralateral approach is indicated.

The circumglossal approach offers the widest anatomical exposure among the methods described; therefore, this technique should be used with caution due to the substantial related morbidity, including hearing loss and serous otitis media.[55] Therefore, this approach should be reserved for select cases requiring extensive midline exposure, along with exposure of the lateral compartment of the vertebral skull.[58]

CONCLUSIONS

The upper cervical spine is an anatomically complex area and the CVJ anatomy is difficult to expose, regardless of the access route. The removal of anteriorly located tumors at the CVJ requires appropriate and challenging surgical strategies. The tumor location and spine level of surgical access should be considered to determine the most suitable surgical approach.

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