

Acesso Labiomandibular com Glossotomia Mediana para Patologias Craniocervicais e Reconstrução em Múltiplos Níveis: opções

Acceso Labiandandibular con Glosotomía Mediana para Patologías Craneocervicales y Reconstrucción de la Columna Vertebral Multinivel: Opciones

Median Labiomandibular Glossotomy Approach for Craniocervical Pathologies and Multilevel Spine Reconstruction: options

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## SUMMARY

**Introdução:** A exposição do clivus e da coluna cervical alta deve, idealmente, fornecer um campo cirúrgico adequado no qual o cirurgião possa, com segurança, descomprimir e estabilizar a junção craniovertebral (JCV). **Apresentamos aqui uma série de 4 casos com revisão narrativa da literatura nos quais a abordagem Labiomandibular com Glossotomia Mediana foi utilizada para o tratamento de patologias da JCV com o objetivo de expor a importância e indicações de tal acesso.** **Material e Métodos:** Foi realizada uma análise retrospectiva dos pacientes submetidos a abordagem Labiomandibular para diversas patologias. O grupo foi composto de 4 pacientes (2 homens e 2 mulheres). 5 abordagens realizadas (1 cirurgia de revisão). **Resultados:** A abordagem foi adequada para todos os casos. O clivus foi atingido quando necessário. Distalmente C4 foi exposto obtendo-se uma osteossíntese satisfatória. Lateralmente, tivemos uma boa visão das margens tumorais e controle vascular da artéria vertebral. Como complicações tivemos infecção superficial, facilmente tratada; deiscência tardia da parede posterior da faringe e pseudoartrose, ocorridos no mesmo paciente. **Discussão:** Existem 3 principais técnicas anteriores para o tratamento de lesões do clivus, forame magno ou vértebras cervicais superiores. Escolhemos a abordagem Labiomandibular como opção primária, o que nos proporcionou uma visão direta do clivus, C3 - C4 caudalmente e um amplo campo cirúrgico. As principais vantagens desta abordagem incluem acesso direto à patologia espinhal, plano avascular através da rafe mediana e um campo cirúrgico ampliado nas dimensões transversa e sagital. **Conclusão:** Esta abordagem proporciona excelente exposição da junção craniocervical e da coluna cervical alta.

**Introducción:** La exposición del clivus y columna cervical superior debe proporcionar un campo quirúrgico adecuado en que el cirujano pueda descomprimir y estabilizar de manera segura la unión craneovertebral (JCV). **Presentamos serie de 4 casos con revisión narrativa de la literatura en que se utilizó el abordaje labiomandibular con glosotomía media para el tratamiento de patologías de JCV y exponer la importancia y las indicaciones de dicho acceso.** **Material y Métodos:** Se realizó un análisis retrospectivo de los pacientes sometidos al abordaje Labiomandibular para diversas patologías. El grupo fue compuesto por 4 pacientes (2 hombres y 2 mujeres). Realizaron 5 abordajes (1 cirugía de revisión). **Resultados:** El acceso fue adecuado para todos los casos. El clivus

se alcanzó cuando necesario. Distalmente C4 fue expuesto obteniéndose una osteosíntesis satisfactoria. Lateralmente, tuvimos una buena visión de los márgenes tumorales y de la arteria vertebral. Como complicaciones, se presento una infección superficial, dehiscencia tardía de la pared posterior de la faringe y pseudoartrosis, en mismo paciente.

**Discusión:** Existen 3 principales técnicas anteriores para el tratamiento de lesiones del clivus, foramen magno o vértebras cervicales superiores. Elegimos el abordaje Labiomadibular como una opción primaria, que nos proporcione una visión directa del clivus, C3 - C4 y un campo quirúrgico amplio. Las principales ventajas de este acceso incluyen vista directa de la patología espinal, plano avascular a través de la rama mediana y un campo quirúrgico largo transversa y sagitalmente. **Conclusión:** Abordaje proporciona una excelente exposición de la unión craniocervical y de la columna cervical alta

**Introduction:** Exposure of the clivus and upper cervical spine should ideally provide an adequate surgical field in which the surgeon can safely decompress and stabilize the craniovertebral junction (CVJ). **We present a series of 4 cases with narrative review of the literature in which the Labiomandibular approach with Medium Glossotomy was used for the treatment of pathologies of CVJ in order to expose the importance and indications of such access.** **Material and Methods:** We performed a retrospective analysis of patients underwent to a MLMG for several pathologies. The group comprised 4 patients (2 men, 2 women). 5 approaches were accomplished (1 revision surgery). **Results:** The approach was suitable for all cases, clivus was achieved when necessary. Distally C4 was exposed obtained a satisfactory osteosynthesis. Laterally we had a good view of the tumor and vertebral artery control. As complications we had superficial wound infection, easily healed, a tardy pharyngeal wound dehiscence and pseudoartrosis, all in the same patient. **Discussion:** There are 3 main anterior surgical techniques for managing lesions of the clivus, foramen magnum or upper cervical vertebrae. We choose the Median Labiomadibular Glossotomy (MLMG) as a primary option which provided us direct view of the clivus, C3 – C4 caudally and a wider surgical field. The main advantages of the MLMG technique include direct access to spinal pathology, avascular plane through the median pharyngeal raphe and a wider surgical field in both transverse and sagittal dimensions. **Conclusion:** This approach provides excellent exposure of the craniocervical junction and upper cervical spine.

**Key-words:** median labiomandibular approach, transoral approach, upper cervical spine, tumors, spine cord compression.

## INTRODUCTION

The exposure of the clivus and upper cervical spine should ideally provide optimal operative field in which the surgeon can decompress and stabilize craniovertebral junction (CVJ). Various surgical techniques have been described using a transcervical retropharyngeal approach<sup>1,2</sup>, retrocarotid lateral<sup>3</sup>, as well as, pharyngeal approach<sup>4</sup>. These techniques are poorly suited to access the upper cervical vertebrae, clivus and CVJ due to limited ventral exposure and risk to critical neurovascular structures<sup>5</sup>.

Roux (1839)<sup>6</sup> described a splitting the lower lip and mandible in the midline for tumors of the anterior tongue. A mandible and tongue splitting procedure was described by Kocher (1911)<sup>7</sup> and Trotter (1929)<sup>8</sup> for exposure the base of the tongue, epiglottis and posterior oropharyngeal wall. Hall et al (1977)<sup>4</sup> reported the use of the median labiomandibular glossotomy to gain the surgical access to the upper cervical spine for treat one case of cervical kyphosis causing mielopathy. Wood et all<sup>9</sup> used the same approach to access the clivus and operate two patients, one with chordoma other with basilar impression.

The most direct approach to the clivus and the upper cervical spine is through transoral and transpalatopharyngeal wall. It has been used to fuse or decompress the upper spinal canal in many cases, such as, chordoma, metastatic cancer, odontoid fracture, basilar invagination and hypertrophic rheumatic connective tissue<sup>10-14</sup>. The primary indication for MLMG approach is to provide access to the CVJ when that cannot be obtained with a standard transoral transpharyngeal technique, particularly in pediatric patients whose anatomy limits exposure, also in cases when is needed to reach the subaxial spine below C2 – C3 disc or in cases which inter-incisor distance is less than 3cm.

The MLMG approach is most often used for extradural pathologies<sup>15</sup>, such, congenital anomalies, benign or malignant neoplasms, inflammatory diseases and traumatic injury<sup>16</sup>. Congenital pathology may includes cervical kyphosis and odontoid malformations<sup>17</sup>.

Between the main primary tumors on clivus and upper cervical spine are chordomas, chondromas and chodrosarcorma. Osteogenic sarcoma and osteoblastoma may represent rare indications<sup>18</sup>. Rheumatoid arthritis is a relatively indication for anterior approach to access CVJ<sup>19</sup>. Traumatic indications for high cervical spine are rare, with the odontoid

fracture being the most common<sup>20</sup>, but the majority can be treated with traction and immobilization<sup>16</sup>.

Here we present four cases in which Median Labiomadibular Glossotomy (MLMG) approach was used to treat CVJ and upper cervical spine pathologies, as well as, review of the relevant literature, in order to expose the importance and indications of this approach.

## MATERIAL AND METHODS

We performed a retrospective analysis of patients underwent to a MLMG for several pathologies. The group comprised for 4 patients (2 men, 2 women). 5 approaches were accomplished (1 revision surgery). The cases were three tumors (aneurysmal bone cyst, chordoma and giant cell tumor) and one congenital kyphosis. Each patient was subjected to tracheostomy, as a first step. Palatotomy was not necessary just in case 1. Reconstructions were made with harms cage filled with bone graft in 3 cases. Case 4 reconstruction was performed with fibular bone allograft. In patients with tumor (cases 2,3,and 4) cerebral angiogram with balloon occlusion of vertebral artery as a test was performed, to determine the feasibility of vessel sacrifice. The main feeding of the tumor was embolized and occluded with coil in these cases. Broad spectrum antibiotic, including anaerobe, was administrated to all patients.

### Operative Technique

Firstly, all patients are underwent a posterior occipitocervical fusion. The patient is placed in the supine position. A tracheostomy tube was placed initially to provide a good view of the posterior oropharynx and to provide a secure airway postoperatively, avoiding complications secondary to a significant lingual and oropharyngeal oedema. Perioral region, jaw, neck, mouth and oropharynx are prepared and draped in a sterile fashion.

A midline incision is made from the lower lip and sublabial crease, curving around the chin pad, back to the midline on submental neck, extends inferiorly to the level of the hyoid bone. (fig 1. A)



Fig. 1: A - *Planning incision.* B - *Planing reconstruction.* C - *Mandibular osteotomy.*

Anterior neck soft tissue is incised in the midline between the hyoid and mandible with anterior exposure of the mandible at the planned osteotomy site. Mini plates and screws are positioned and molded before, for later replacement. (Fig. 1B) The osteotomy is made with an oscillating saw following a median line and preserving the central incisors roots. (fig 1C) This step preserve occlusion relationships postoperatively.

Following a mandibular osteotomy, the soft tissue dissection within the floor of the mouth is continued in the midline between the submandibular ducts and carried into the intrinsic tongue musculature. Retention sutures are then placed on either side of the tongue to retract it. (Fig 2A). An electrocautery incision is made posteriorly along the median raphe to expose the lingual surface of the epiglottis to the level of the hyoid, exposing posterior oropharyngeal wall. The mandibular lingual halves are spread laterally and held in place by retractors. (Fig 2B) . A midline split of the soft plate can be performed.

The posterior pharyngeal wall is infiltrated and then incised along the median raphe, then pharyngeal flaps are swept away to either side to expose clivus and upper cervical spine, (Fig 2C) or incised according Harms-schmelzle<sup>21</sup> technique with an open-door flap which provide a better access to the lateral aspects of C1 – C2 joints and it has additional advantage in cover a metallic prosthesis, preserving arterial blood supply.

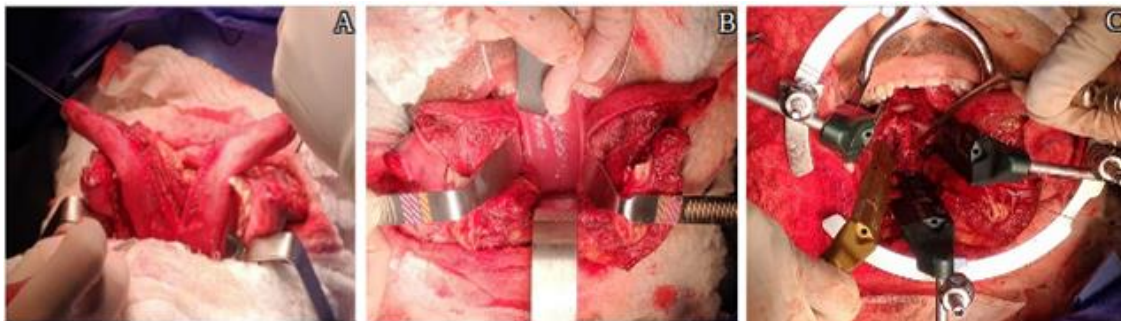


Fig. 2: A - *Tongue split.* B - *Pharyngeal wall.* C - *Clivus and upper cervical spine exposition.*

Retractors provide the exposure and allows for surgical decompression or resection of the lesion. Anterior internal fixation can be made using a titanium implant or a bone graft. Meticulous closure is performed using the longus colli muscle and prevertebral fascia. The pharyngeal wall are closed in two layers: pharyngeal musculature and mucosa. Reconstruction of the tongue is started from posterior to anterior using absorbable suture. The intrinsic lingual musculature is brought together. The ventral surface of the tongue and the floor of the mouth are closed in this order.

Mandibular osteotomy is re-positioned using the prefashioned rigid fixation plate and screws for osteosynthesis. When closing the floor of the mouth, care must be taken to cover the osteotomy site intraorally. The soft tissue of the lip, chin and mentul region are closed in layers with careful reapproximation of the vermilion – cutaneous junction. A nasogastric feeding tube is placed beyond the posterior pharyngeal incision under direct visualization.

## RESULTS

The approach was suitable for all cases, clivus was achieved when necessary (cases 3 and 4). Distally C4 was exposed obtained a satisfactory osteosynthesis. Laterally we had a good view of the tumor and vascular control of vertebral artery.

As complications we had superficial wound infection, easily healed, a tardy pharyngeal wound dehiscence and pseudoartrosis, in the same patient. (case 1). Two cases of velopharyngeal insufficiency. No problem with mandible osteosynthesis or tracheotomy was related. In two cases (3 and 4) anterolateral-retropharyngeal decompression was necessary for recidivate tumors. These two patients were underwent a radiotherapy. One death occurred (case 4) four months after index surgery for recidivate tumor and skull base invasion.

### Illustrative Case

A 20-year-old woman presented with diffuse cervical pain, paresthesia in her left side and Hoffman signal, presumably a Brown-sèquard syndrome cause by a severe ventral cervico-medullary compression secondary to a congenital cervical kyphosis and CVJ instability (Fig 3).

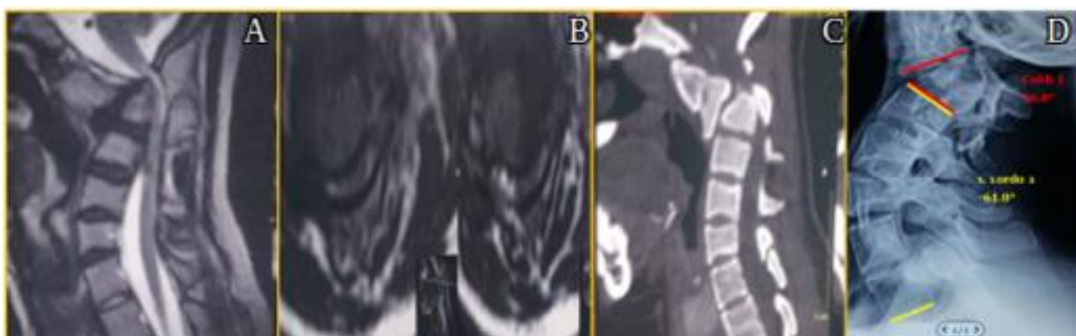


Fig 3. A and B – MRI sagittal and axial views showing severe medullar compression. C and D - CT scan and Xray showing congenital kyphosis.



She was undergone to a occipito-cervico-thoracic fixation and C1, C2 laminectomy on the first step of the procedure followed by a MLMG approach with resection of the lower part of C2 and C3 vertebral body. Good decompression and cervical sagittal balance was achieved. Anterior fixation was secured by titanium cage mesh filled with autograft bone from C2 to C4. (Fig . 4)

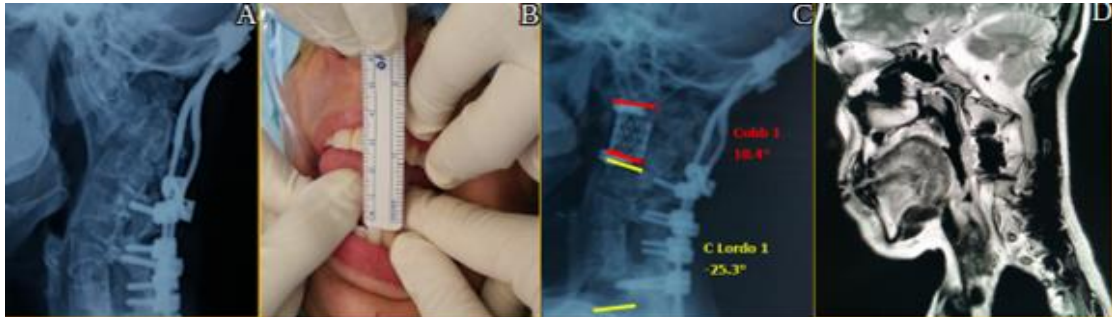


Fig 4: A – Occipitocervical fixation. B – Mouth opening < 3cm. C – Postoperative of anterior approach. D – MRI with medular decompression.

Postoperatively, superficial infection on subcutaneous tissue of the lower lip was treated and she presented asymptomatic pseudoarthrosis in the long-term follow-up.

Almost two years after first surgery, she complains again of numbness and difficult for walking. She presented hemiparesis on her right side and a new symptom: difficult and pain to swallow. She performed new exams that showed pseudoarthrosis progression and posterior pharyngeal wound dehiscence. The patient was undergone to a revision surgery through the same approach (MLMG). Cage was taken out, decompression of the spine cord was achieved. In order to provide fusion we used iliac crest graft. Three weeks later she developed a brown-sequard syndrome on her right side by an epidural abscess which was drained and put a para-pharyngeal drain. In consequence of her paresis thromboprophylaxis was administered, that resulted in a haematoma on pharyngeal wall. We performed angiography of cervical vasculature and no abnormalities were found. Gastrostomy was performed in order to feed. Now she is recovering with improvement of her strength, capacity of swallowing and good pharyngeal wall healing. (Fig. 5)

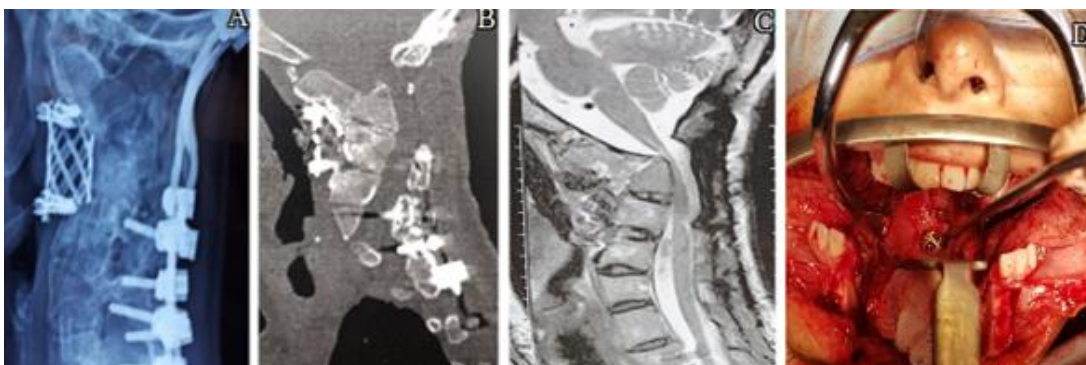


Fig 5: A – X ray showing displaced cage. B – Bone growth behind the cage. C – MRI showing medullar compression. D – Dehiscence of pharyngeal wall.

Our cases are summarized on table 1 followed by images of the cases.

Table 1 – Operative technique and complications

	Diagnosis	Approach	Anterior Reconstruction	Posterior Reconstruction	Complications
1	Congenital Kyphosis with mielopathy	MLMG	Harms Cage with bone graft	OCF	Pharyngeal dehiscence + epidural abscess +haematoma
2	Aneurismal Bone Cyst	MLMG	Harms Cage with bone graft	OCF	-
3	Chordoma	MLMG + Anterolateral retropharyngeal*	Harms Cage with bone graft	Extension of posterior cervical fusion	velopharyngeal insufficiency
4	Giant Cell Tumor	MLMG + anterolateral retropharyngeal*	Fibular bone allograft	Extension of posterior cervical fusion**	velopharyngeal insufficiency

MLMG: median labiomandibular glossotomy, OCF: occipito-cervical-fusion

\*second surgery due recurrence of the tumor. \*\*Extension of previous posterior cervical fusion



Fig 6 (case 2): A – Cervical Kyphosis. B – Litic lesion C1-C3. C – T2 sagittal MRI showing tumor and medullar compression. D – C1-C4 posterior fixation. E – Anterior reconstruction C1-C4. F – Intraoperative view.

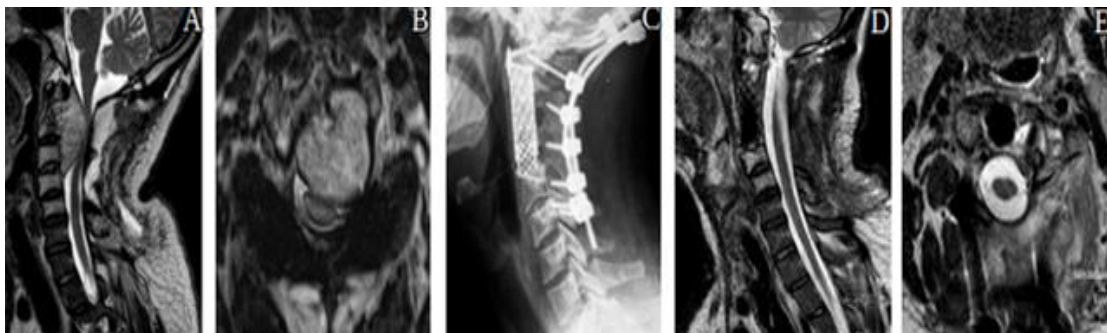


Fig. 7 (case 3): A and B – T2 MRI showing a huge recurrent extradural lesion from C1-C4. C – Anterior reconstruction after tumoral removal. D and E – T2 MRI showing no residual tumor and medullar decompression.

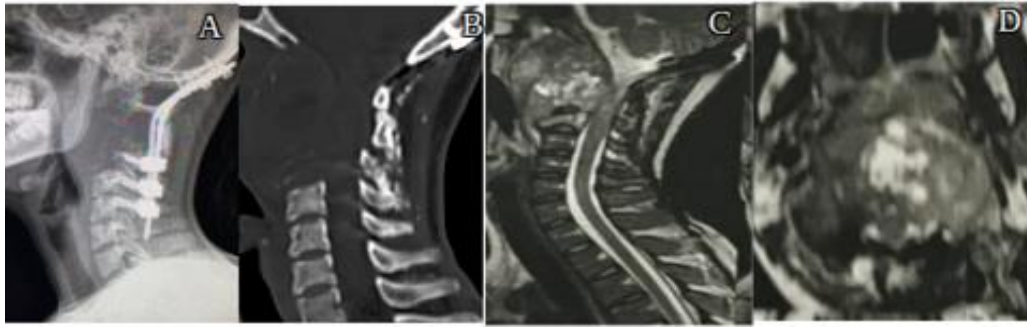


Fig. 8 (case 4): A and B – X ray and CT scan showing lytic lesion from C0-C4. C and D – T2 MRI sagittal and axial views showing heterogeneous lesion in CVJ.

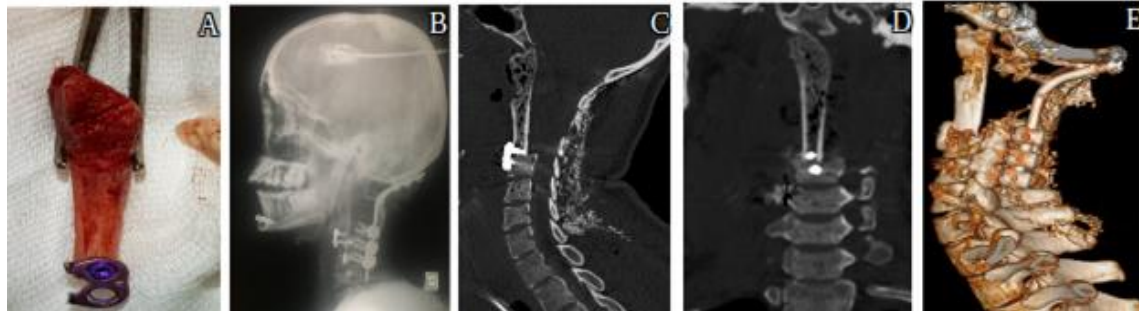


Fig. 9 (case 4): A – Fibular allograft. B, C and D – X ray and CT scan showing anterior CVJ reconstruction. E – 3D CT scan.

## DISCUSSION

A combined transoral – transpharyngeal approach with a median mandibulotomy (median labiomandibular approach) allows for increase caudal exposure to the C3 – C4 interspace and maintains the superior exposure of the lower third of the clivus. Dividing the tongue in the midline further increases the caudal exposure to the C4 – C5 interspace (MLMG approach)<sup>22,23,24,25,26</sup>. This approach has been used in the treatment of a variety of pathological process, with the final goal of decompression, fusion, or both, as we can see in our paper.

There are 3 main anterior surgical techniques for managing lesions of the clivus, foramen magnum or upper cervical vertebrae<sup>27,28,29,30,31</sup>. The transoral standard approach that allow the surgeon to operate directly any intra or extradural lesion located between the clivus and third cervical vertebrae<sup>32</sup>. Disadvantage this approach is limited working space and high risk of surgical field contamination. If patient has limited mouth opening and restricted neck extension, the transoral approach does not provide adequate exposure for surgery, we had a patient with this condition in our serie. The second anterior technique is the transmaxillary approach that allow to access intra or extradural lesions located between the clivus and C2<sup>27,33</sup>, being inappropriate to our patients, because the tumor had invaded the body of C2, C3 extending until C4. The third surgical option for

accessing upper cervical region is high cervical retropharyngeal approach, which was used in cases 3 and 4 due tumor recurrence. The disadvantages of this approach, as first option, are awkward trajectory, restricted depth of exposure and lack of the midline access, that was necessary for complete tumor resection. How our patient had undergone previous neck fusion, neck extension and rotation were no possible and none of these 3 main techniques were suitable<sup>32,34</sup>.

We choose the extended transoral transmandibular approach as a primary option which provided us direct view of the clivus and C3 – C4 caudally<sup>35</sup> and a wider surgical field. The midline approach allowed the surgeon greater exposure without major muscle damage, important blood vessels or nerves lesion. Indications to use a MLMG approach to augment exposure of the CVJ and the upper cervical vertebrae include an inter-incisor opening distance less than 2,5 – 3cm and when access to C4 – C5 is required as we can see in cases 3 and 4<sup>36</sup>. Glossotomy is necessary when the lesion extends down to C2 or below<sup>37</sup>.

The main advantages of the MLMG technique include direct access to ventral spinal pathology, extended head position and an avascular plane through the median pharyngeal raphe. This approach provide a wider surgical field in both transverse and sagittal dimensions. As described by Arbit and Patterson the cosmetic deformity and functional loss are minimal despite the seemingly radical incision<sup>22</sup>. Disadvantages include facial scarring, oral and velopalatine incompetence, dysphagia, malocclusion, limited tongue mobility and sensation, and complications of tracheostomy<sup>23</sup>. We had two cases of phonation disorder and no complications with tracheostomy.

Oral contamination of the wound theoretically carries an increased risk of infection. In study of 72 patients undergoing to a transoral transpalatopharyngeal procedure, only one patient developed an infectious complication, which resolved without sequelae after drainage<sup>38</sup>. The majority of infectious complications are localized to the pharyngeal wall and do not progress to meningitis<sup>39,40</sup>. One of our four patients (case 1) had superficial infection on the subcutaneous tissue of the lower lip and developed an epidural abscess after MLMG revision procedure. Some authors recommend empiric prophylactic antibiotics<sup>40,41,42</sup>. We performed this recommendation for all patients. Other authors indicate that preoperative throat cultures should be used to aid on the choice of prophylactic antibiotics<sup>22</sup>.

We had no case of cerebro spinal fluid (CSF) leakage or meningitis as reported by Menezes<sup>36</sup> in his series, 280 children younger than 16 years submitted to a transoral approach to the pharyngeal wall. We had two cases of dural lesion with CSF fistula intraoperatively treated with primary repair.

A pharyngeal wound dehiscence is a rare complication related in 0,7%<sup>43</sup>. We had this later complication in case 1 and the patient was re-operated with a satisfactory postoperative result.

Velopharyngeal insufficiency (VPI) typically occurs 4 – 6 months following the operative procedure<sup>36</sup>. Authors have implicated soft palate division as a significant risk for VPI, supporting the need for meticulous closure of the palatal defect. We had two temporary VPI episodes in cases 3 and 4, who needed the division of the soft palate in order to obtain better exposure of the superior limit of the tumor.

The limitations encountered for cervical spine chordomas result from involvement of the dura, nerve roots and vertebral arteries<sup>44,45</sup>. The duramater was infiltrated by the tumor in chordoma case and closure with fascia lata and artificial duramater was needed. In the last case the lesion's (GCT) behavior was aggressive, posterior wall of the pharynx was infiltrated and both carotid arteries were encased, this became en bloc resection impossible. Furthermore, some authors have recommended cerebral angiogram and performing temporary balloon occlusion test as part of the preoperative workup in patients with vertebral artery involvement to determine the feasibility of vessel sacrifice to achieve en bloc resection<sup>46</sup> or perform intralésional resection, as we did in our tumor cases.

Instability after resection of C1, C2 and C3 should be corrected. Reconstructions of the anterior column is mandatory. Anterior cage mesh filled with autologous bone graft was used in cases 1, 2 and 3. On the last case we used autologous fibula. The occipitocervical fusion is morbid because of the functional limitations that it places by greatly decreasing the spinal mobility<sup>47</sup>. We decided to perform occipitocervical fusion in all cases to provide sufficient bio-mechanical support.

## **CONCLUSION**

The Median Labiomandibular Glosotomy approach is useful to treat many different CVJ pathologies, including tumors and congenital abnormalities. This approach provides excellent and safe exposure of the craniocervical junction and upper cervical spine for decompression and reconstruction procedures with acceptable few complications.

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