

INTRADURAL JUGULAR FORAMEN TUMORS

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ABSTRACT - Eleven patients with jugular foramen lesions with or without extradural extension were operated at University Hospital of Campinas (UNICAMP), in Campinas, Brazil, between 1998 and 2001. Neck dissection, mastoidectomy without transposition of the facial nerve and myofascial flap reconstruction of the cranial base with an especially developed technique were carried out in 7 patients. Four patients were operated using retrosigmoid craniectomy. Total excision was accomplished in 9 cases. All patients did not show evidence of disease progression at least after 2 years follow-up. There was no mortality. New lower cranial nerve deficits occurred in 5 patients. Nine maintain or improved their preoperative status based on Karnofsky and Glasgow Outcome Scale. A complex anatomy of this region demand wide exposures for treat those tumors. For this reason, an adequate approach for curative resection of most lesions and an efficient skull base reconstruction decreasing postoperative morbidity are essential.

KEY WORDS: jugular foramen, posterior fossa tumor, lower cranial nerves, skull base reconstruction, glomus tumor.

Tumores intradurais do forame jugular

RESUMO - Onze pacientes com lesões expansivas do forame jugular associadas ou não a componente extradural foram submetidos a ressecção cirúrgica no Hospital das Clínicas da Universidade Estadual de Campinas (UNICAMP) entre 1998 e 2001. Foi utilizada cirurgia combinada com dissecação do pescoço, mastoidectomia sem transposição do nervo facial e técnica de reconstrução miofascial da base do crânio desenvolvida por um dos autores. Quatro pacientes foram operados via craniectomia retrosigmoidea. Ressecção total foi feita em 9 pacientes, subtotal em um e parcial em outro. Nenhum dos 11 pacientes teve progressão da doença em 2 anos de acompanhamento. Não houve mortalidade. Cinco pacientes tiveram déficits adicionais de nervos cranianos baixos. Nove pacientes mantiveram ou melhoraram suas pontuações de acordo com a escala de Karnofsky. Para adequada abordagem do paciente com tumor de forame jugular, são necessários bom conhecimento anatômico da região, exposição cirúrgica ampla, e técnica de reconstrução eficiente para obter cura com baixas taxas de complicações pós-operatórias.

PALAVRAS-CHAVE: forame jugular, tumor de fossa posterior, nervos cranianos baixos, reconstrução de base de crânio, tumor de glomus jugular.

Jugular foramen (JF) lesions are rare, being paragangliomas and schwannomas the most common tumors^{1,2}. Complete resection of these benign lesions is the treatment of choice. The surgical management of JF lesions remains difficult because their deep location, locally aggressive behavior, involvement of vessels and nerves, infiltration of skull base bone and structures of the high cervical region and the complex anatomy of the region. In the last decades, development of new skull base approaches allowed total resection of large number of skull base tumors. However, the more aggressive the surgical approach becomes, more complications related to it may occur. Reconstruction of the skull base

after large bone resection is very important to avoid complications. For this reason, the surgical access and the reconstruction techniques are decisive factors concerning the outcome.

We report 11 patients with JF tumors operated at the Skull Base Center of the UNICAMP, between 1998 and 2001. The clinical, radiological, surgical technique and the results are discussed.

METHOD

Patient population – Eleven patients with intradural tumors involving the JF were operated. Seven patients had tumors with additional extradural extension. There were six women and five men. The mean age was 45

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Table 1. Initial presentation of 11 patients with jugular foramen tumors.

	Age/Sex	Initial symptom	VII(#)	VIII	IX	X	XI	XII
1.	59, F	Hoarseness	1	+	+	+	+	+
2.	51, F	Headache	2	+	-	-	-	-
3.	55, F	Hearing loss	1	+	-	-	-	-
4.	63, F	Headache	1	+	-	-	-	-
5.	50, M	Hearing loss	2	+	-	-	-	-
6.	62, M	Hearing loss	1	+	-	-	-	-
7.	47, M	Tinnitus	2	+	-	-	-	-
8.	19, M	Hearing loss	2	+	-	-	-	-
9.	39, F	Hoarseness	1	+	+	+	+	+
10.	38, M	Hearing loss	1	+	+	+	+	+
11.	13, F	Hearing loss	3	+	-	-	-	-

House-Brackmann grade for facial function; VII, VIII, IX, X, XI and XII = cranial nerves; (+) presence of nerve deficit; (-) absence of nerve deficit

Table 2. Curitiba 1988 classification.

E	Tumor located in the ear (E)
EN	Tumor located in the ear (E) and neck (N)
ENI	Tumor in the ear (E), neck (N) and intradural (I) compartment
MIXED	Combination among the anterior types
N	Tumor restricted to the neck (N)
I	Tumor restricted to intradural (I) compartment

years (range 13 to 63 years). The initial symptoms were hearing loss (6 cases), hoarseness (2 cases), headache (2 cases) and tinnitus (1 patient). The mean time between initial symptoms and the search for treatment was 3.3 years. At the time of diagnosis all patients had some degree of hearing loss, five showed facial nerve palsy and three had lower cranial nerves dysfunction (Table 1). The postoperative follow-up period ranged from 2 to 4.2 years. There were 6 paragangliomas, 2 schwannomas, 1 meningioma, 1 choroid plexus papiloma and 1 endolymphatic sac tumor.

Radiological evaluation – All patients were evaluated pre and postoperatively with computerized tomography (CT) and magnetic resonance imaging (MRI). The lesions were classified using “Curitiba 1988” classification (Table 2) according to involvement of ear, neck and intracranial compartment in types E, EN and ENI³. Four vessel angiography and preoperative embolization with Ivalon® particles were carried out (6 cases) 48 to 72 hours before surgery when the radiological findings revealed a high vascular tumor as usually found in paragangliomas.

Surgical approaches – A multidisciplinary team composed by neurosurgeons and otologists performed the surgical procedure. The decision concerning the surgical approach was based on radiological aspects. Intracranial tumors that showed no evidence of ear, temporal bone or neck involvement were approached by retrosigmoid

craniectomy (4 cases). In the remained 7 cases, retrosigmoid craniectomy associated to neck dissection and mastoidectomy was performed to remove both intradural and extradural tumor in one surgical procedure.

The patient is placed supine with underneath shoulder roll and the head turned approximately 45 degrees to the opposite side and fixed in a Mayfield® three pin head - holder.

A C shaped incision from the temporal fossa, passing behind the ear, to the anterior border of sternomastoid muscle in the high cervical region is performed for the combined approach (Fig 1). A linear retro auricular incision 4 cm behind the external auditory canal, 4 cm above the helix and 4 cm under the lobule is used for retromastoid craniectomy. The skin flap is rotated anteriorly exposing the posterior temporal, parietoccipital and high cervical areas.

Miofascial flap for skull base reconstruction – The temporal fascia is incised in the middle portion of the tempo-



Fig 1. Patient positioned in the surgical table with C-shaped line showing skin incision, dotted line showing posterior fossa craniectomy and dense mark as transverse and sigmoid sinus.

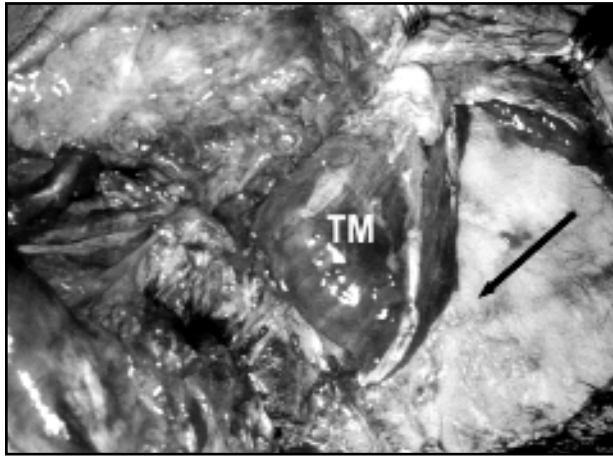


Fig 2. Posterior third of the left temporalis muscle (TM) dissected from its anterior portion for inferior and posterior rotation (black arrow) covering the mastoid bone defect.

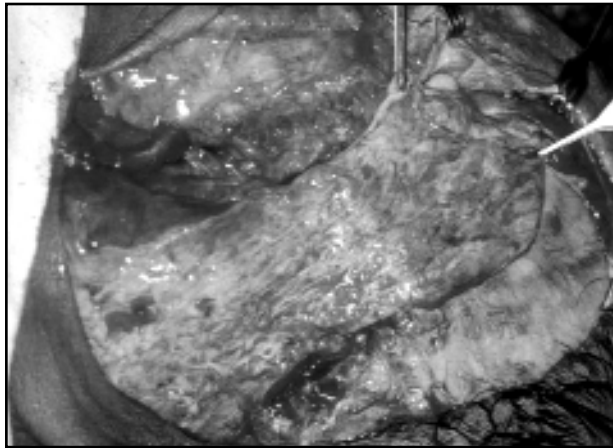


Fig 3. A third myofascial layer, the sternocleidomastoid muscle, covering the entire bone defect, preserving the neck contour.

ral region and dissected to the temporal line. The cervical fascia is cut posterior to the external auditory canal, mastoid tip and over the sternocleidomastoid muscle (SCM). The insertion of SCM is removed from the mastoid and the vascularized myofascial flap is turned posterior and inferiorly. In patients with preoperative deafness, the external auditory canal is transected and closed with separate sutures covered by a posterior auricular muscle flap.

Neck dissection – Neck dissection with identification of the Xth, XIth, and XIIth cranial nerves, common carotid, external carotid and internal carotid arteries, and the jugular vein is performed. The vertebral artery is always identified at the craniocervical junction. The facial nerve is identified superiorly to the digastric muscle using the pointer and the mastoid process as parameters.

Mastoidectomy – A radical mastoidectomy is performed. Removal of the semicircular canal is carried out if

the lesion infiltrates this structure. The middle fossa and presigmoid dura mater are exposed. The facial nerve remains in its bone canal if the lesion does not infiltrate it. The retrofacial mastoid cells are removed.

Retrosigmoid craniectomy – A key-hole on asterion followed by a 3 cm posterior fossa craniectomy is performed exposing the transverse sinus superiorly and unroofing completely the sigmoid sinus. The JF is widely opened.

Ligature of sigmoid sinus and jugular vein – A small dura incision is made on each side of sigmoid sinus (SS) inferior to the superior petrosal sinus. The SS is ligated with two sutures. The internal jugular vein (IJV) is ligated and cut in the high cervical region.

Tumor removal – After opening the posterior wall of SS, the extracranial portion of tumor is removed from the JF with the IJV. The dura mater is incised in the medial wall of the SS and the intracranial portion of JF and its contents are exposed. The lesion is resected using microsurgical techniques. Infiltrated dura mater is completely resected. Dura mater infiltration is more frequently found in meningiomas and paragangliomas.

Skull base reconstruction (myofascial flap) – If primary watertight dural closure is not possible, a temporal fascia flap is used to cover the defect. Fibrin glue is always used. A second skull base reconstruction layer is formed by the inferior rotation of posterior portion of temporal muscle (Fig 2).

The myofascial flap is rotated anteriorly, the temporal fascia is approximated and sutured and the cervical fascia is sutured in the parotid fascia and anterior neck fascia. This three layer closure covers the entire bone defect due to mastoidectomy and posterior fossa craniectomy (Fig 3). The SCM muscle shape is maintained. The scalp flap is closed in a single layer with separated non absorbable sutures. No drain is used and prophylactic. Lumbar drainage was not necessary with this technique.

RESULTS

Total tumor resection could be achieved in 9 patients confirmed by postoperative radiological evaluation that failed to show residual expansive lesions with contrast enhancement. Five patients presented with postoperative IXth and Xth nerve deficits, 2 showed XIth and XIIth deficits and 2 worsening their facial weakness. The new cranial nerve deficits were not associated with tumor size or histopathologic findings but with tumor infiltration of the nerves and dura. Incomplete resection was performed in 2 cases with giant tumors presenting with involvement of the ear, neck and intradural compartments. Eight patients who achieved complete tumor resection maintained or improved their pre-

Table 3. Tumoral aspects and results.

Case	ENI*	Size (cm)	Approach	Resection	New deficit	Complication	Karnofsky	GOS	Tumor
1.	EN	2 x 2	R + M	Total	None	None	90 / 100	4	Paraganglioma
2.	ENI	2 x 2	R + M	Total	VII,IX,X,XI	None	100 / 80	4	Paraganglioma
3.	NI	4 x 3	R + M	Total	IX,X,XI,XII	None	90 / 100	4	Paraganglioma
4.	I	4 x 3	R	Total	IX,X	None	80 / 90	4	Meningioma
5.	I	2 x 2	R	Total	IX,X	None	100 / 100	4	Papiloma
6.	I	3 x 3	R	Total	IX,X	CSF leak	70 / 90	4	Schwannoma
7.	I	4 x 2	R	Total	VII	None	90 / 90	4	Schwannoma
8.	?	3 x 2	R + M	Total	None	None	90 / 90	5	Endolymphatic tumor
9.	ENI	6 x 4	R + M	Partial(<90%)	None	None	80 / 70	4	Paraganglioma
10.	ENI	4 x 4	R + M	Total	None	None	90 / 90	4	Paraganglioma
11.	ENI	4 x 3	R + M	Subtotal	None	None	90 / 90	4	Paraganglioma

R, retrosigmoid craniectomy; R + M, retrosigmoid craniectomy + cervical and mastoidectomy combined approach; * Curitiba 1988 classification (Table 2), GOS, Glasgow Outcome Scale.

operative status confirmed by Karnofsky Performance Scale of 90 or 100 and Glasgow Outcome Scale (GOS) 4 or 5. One patient in the group of complete resection worse his clinical status from Karnofsky 100 to 80. The remaining 2 patients had an incomplete resection. One had partial (< 90%) and the other subtotal (> 90%) tumoral removal accordingly to contrast enhanced residual lesion in the postoperative MRI. One patient remains with his preoperative Karnofsky status and the other one decreased it from 80 to 70. Both patients showed no evidence of progressive disease after follow up examinations and radiological evaluation 28 months after the surgical procedure. One patient developed cerebrospinal fluid (CSF) leak and meningitis treated with antibiotics. There was no mortality and no local infection (Table 3).

DISCUSSION

JF lesions are uncommon^{4,5}. Paraganglioma was the most common tumor in our series followed by schwannoma. This finding is in accordance with the literature^{1,2,5}. Paragangliomas are rare benign tumors that arise from paraganglionic cells along the body. In the skull base, the most frequent site for paragangliomas is in the jugular and tympanic paraganglia also called glomus jugulare or glomus tympanicum tumors, respectively^{5,6}. Schwannomas are the second most common tumor at this location, arising from IXth, Xth or XIth cranial nerves. Meningiomas, metastases, giant cell tumors, chondrosarcomas, plasmocytomas, epidermoid cysts, amyloidomas, endolymphatic sac tumors are described in this location as primary or extended lesions^{2,7-12}.

The growth pattern of tumor seems to be deci-

sive for clinical presentation of this group of pathology. Predominantly intracranial expansion is related to symptoms of posterior fossa compression and deafness. Tumors with primary bone or extracranial involvement usually present with lower cranial nerve dysfunctions¹³⁻¹⁵. Hearing loss is the most frequent symptom of JF tumors^{13,14,16-20}. In our series, all tumors had intracranial component and auditory dysfunction was present in all cases. The jugular foramen syndrome is not always present¹⁶; however, normal cranial nerve function before surgery does not exclude cranial nerve tumor infiltration²¹. Glomus jugulare tumors are more common in female patients^{5,14,19,21,22}. Multiple paragangliomas are found in approximately 10% of cases^{5,16,21,23}. The presence of bilateral glomus jugulare or contralateral glomus vagale tumor may result in postoperative bilateral lower cranial nerve palsies¹⁶. The mean time between the initial symptoms and the diagnosis demonstrate the slow growth pattern of paragangliomas^{5,22} and schwannomas^{13,14}, as found in our series. The mean age at presentation is around the fifth decade^{5,19,21}.

Radiological investigation – Computerized tomography is very useful to show bone structures, the jugular foramen contours and its variations. Bone erosion is usually found in paragangliomas in contrast to schwannomas that frequently present with JF enlargement with smooth indistinct sclerotic margins^{13,24}. Angiography is useful to differentiate between schwannoma and paraganglioma. Schwannomas are usually less vascularized than paragangliomas¹³. Ascending pharyngeal arteries and meningeal branches of occipital artery supply most of tu-

mors located at this region. In our series, all paragangliomas were submitted to embolization. The ascending pharyngeal artery supply was found in every case (6 cases). It is also important to delineate the patency of jugular bulb before surgery as progressive obstruction is often seen in tumors at this location and permits the development of collateral circulation; however, dominant or non communicating sigmoid sinus can lead to intracranial hypertension if ligation is achieved during surgical procedure²⁵. After the introduction of the MRI, the anatomic details, tumor margins, relationship of major vessels, vascular supply and even the patency of jugular bulb can be observed¹⁴. Meningiomas, choroid plexus papillomas and paragangliomas shows high contrast enhancement.

Jugular foramen anatomy – The jugular foramen is an aperture in the posterior half of the skull base behind the carotid canal. Its anterolateral border is formed by petrous temporal bone and the posteromedial wall by the occipital bone^{14,24,26}. The classic division of JF in three parts is described. The anterior, containing the inferior petrosal sinus as it enters the jugular vein; the middle part, the IXth, Xth and XIth cranial nerves and the posterior, the jugular bulb and meningeal branches of occipital and ascending pharyngeal arteries²⁴. Hovelake divided the JF in two compartments²⁷. The larger posterolateral also called pars venosa containing jugular bulb, Xth and XIth cranial nerves and a smaller compartment (pars nervosa) containing IXth cranial nerve. The perforation can be separated by fibrous septum in 74% to 80%^{26,28} or by bone septum in the remained cases. Ayeni et al. described that both the division in pars venosa and nervosa or three compartments should not be used as the IXth, Xth and XIth cranial nerves are entirely anteromedial to jugular bulb^{28,29} in spite of the presence of anatomic separation of JF by fibrous or bone bridge. The right JF is usually larger than the left in 68% to 70%^{26,28} and equal in 20%²⁸. The cranial nerves (CN) lay in a connective tissue sheath with IXth CN being the most anterior, completely separated from the others and the XIth CN the most posterior^{20,28}. The IXth nerve is usually individual inside the JF, the XIth CN is frequently divided in two parts (spinal and cranial) and the Xth nerve with multiple fascicles^{20,26,28}. The superior jugular bulb is posterolateral to lower cranial nerves and shows multiple configurations concerning the inferior petrosal sinus (IPS) entrance.

The IPS can drain into the jugular bulb, internal jugular vein or both by single or multiple channels anterior to IXth nerve and posterior to XIth nerve^{20,26,28}.

Treatment modalities – Complete surgical removal is the treatment of choice for jugular foramen paragangliomas^{5,29,30}. Comparative studies between radiotherapy and surgery lack of consistency¹⁹. The follow up periods are short considering the slow growth of this tumor and its natural history. Some reports describe long time survival and even involution of the tumor without interventional procedures^{19,22,30}. Radiotherapy is not curative and has no activity in the tumoral cells but in the vascular supply limiting the tumor vascularity leading to fibrosis⁵. This kind of treatment may be indicated in patients with poor surgical condition and residual or recurrent tumors^{5,21,22,30}. The age may be a limiting factor but the therapeutic decision must be individually considered.

Tumors at JF may arise from extradural compartment, intradural or both. Intradural involvement means potential complications such as CSF fistula, meningitis, brain structures adherence and their consequences. In our series, all patients had intradural tumors. Different surgical techniques are used for those tumors. In neurosurgical series, essentially JF intradural tumors as meningiomas, schwannomas, epidermoid cysts and some metastatic lesions are approached by retrosigmoid route in most cases¹³⁻¹⁵. Many techniques are described for large lesions with transcranial component. Transcanal and posteroauricular approaches are not discussed because their use is restricted to small tumors at promontory, middle ear and hypo tympanum. We use an infralabyrinthine or translabyrinthine approach depending on the auditory function. Different skin incisions are described for large tumors. Glasscock et al.²² use modified infratemporal fossa approach with C shaped incision above and just behind the auricle but without extension to temporal or posterior fossa region. Samii et al.¹⁴ use a retro auricular incision extending to the SCM muscle border making possible a combined posterior fossa cervical approach for schwannomas that have extracranial component. Reconstruction of the cranial base is of vital importance in those cases. The surgical approach should be planned to permit adequate skull base reconstruction. Identification of the important structures before tumor removal is essential and is achieved with wide

exposures. Some authors transpose the facial nerve during tumor removal^{5,16,21,22,29}, especially in those tumors with anterior extension, with the objective of increase the surgical exposure with less mobilization of lower cranial nerves. This maneuver leads to transitory facial nerve dysfunction in most patients and definitive deficit in some. Some authors do not transpose the facial nerve^{14,31}. In our surgical technique the facial nerve is skeletonized and left intact in fallopian canal except if there is tumoral infiltration. In our cases, the facial nerve function was maintained in nine patients with this technique. Two patients showed postoperative facial nerve impairment. The first one had infiltrative paraganglioma. The second patient had an intradural tumor and the facial palsy was due intracranial dissection of the tumor capsule from the VIIth nerve. The extracranial portion of tumor is first removed followed by dural opening and intradural tumoral microsurgical resection. The dura is resected when there is any degree of infiltration and reconstruction of the skull base in three layers is done as previously described in this text.

Complications – The lower cranial nerve deficits leading to dysphagia, breathing difficulty, hoarseness and pneumonia due to aspiration are potential dangerous problems. According to literature many prophylactic or therapeutic procedures are performed such as tracheostomy, gastrostomy, vocal cord manipulations and cricopharyngeal myotomy^{5,14,18,21,22,31}. Samii et al.¹⁴ advocate temporary tracheostomy if the lower cranial nerves are sacrificed during the surgical procedure and swallowing function is injured in the postoperative period. Persistent hoarseness is treated with Teflon injection into paralyzed vocal cord after 6 months from surgical procedure according to George⁵ or earlier in postoperative period by Fenton et al.¹⁸ resulting in a better voice and cough reflex. Gastrostomy is employed if persistent aspiration occurs¹⁴. In our series, temporary dysphagia and hoarseness were common. All patients are maintained with enteral feeding until the swallowing function permit the oral intake. Nasogastric tube was kept in place for as long as 14 days but no permanent procedure as tracheostomy or gastrostomy was necessary. No patient had aspirative pneumonia. No patient underwent vocal cord augmentation.

CSF fistula is another type of life threatening complication. Meningitis can occur increasing the morbidity and mortality. The use of fibrin glue

(Beriplas[®]) after the dura closure followed by miofascial flap rotation has been found very efficient in avoiding this complication.

CONCLUSION

Surgical removal of JF benign lesion may be curative. Adequate exposure and skull base reconstruction are very important to achieve complete resection and to avoid postoperative complications.

An experienced multidisciplinary team, an adequate individually pre and postoperative evaluation of the patients are the requisites to achieve radical tumor removal with low morbidity rates and good overall functional outcome.

The fact that some patients had short follow-up period and that most JF tumors has slow growth pattern must be considered. However, the good outcome of the patients demonstrated by Karnofsky scale in the postoperative period added to absence of residual tumor or tumoral progression in all patients shows that the goal of treatment was accomplished.

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