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OTOLOGY

Preservation of the facial and lower cranial nerves in glomus jugulare tumor surgery: modifying our surgical technique for improved outcomes

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Abstract

Purpose To describe the neurological results obtained in six patients with large Fisch C glomus jugulare tumors (GJT) in which a less aggressive, nerve-preserving surgical strategy was used to reduce surgical morbidity.

Methods Prospective study of six patients with Fisch C GJT who underwent surgery in a tertiary care referral center from February 2015 to August 2017 with an average follow-up of 18 months. The intervention is the surgical technique used and the main outcome measures are recurrence and the functional preservation of the facial and lower cranial nerves.

Results Gross total removal was obtained in the six patients with preservation of the medial wall of the jugular bulb protecting the lower cranial nerves. After follow-up, we obtained a House–Brackmann (H–B) grade II in three patients who were managed with an inferior facial nerve transposition. One patient managed with a facial bridge technique preserved a normal facial function and two patients who presented a H–B III before surgery went to H–B V after surgery and recovered to a H–B III after 4 months. Four patients were presented with dysphagia after surgery and required nasogastric tube placement. The average time for removal with return to normal oral feeding was 4.3 weeks. Three patients with preoperative Xth nerve dysfunction showed an adequate compensation of the opposite vocal fold in the postoperative period without dysphonia or aspiration.

Conclusion The surgical techniques used in these patients provided good functional preservation without recurrence after an 8–30-month follow-up.

Keywords Glomus jugulare tumor · Skull base tumor · Paraganglioma · Infratemporal approach

Introduction

The treatment of cranial base tumors has changed a lot during the last two decades, especially for the case of benign tumors. Together with the different surgical approaches and radiotherapy, wait and scan is now considered a good alternative in many cases. In the particular case of glomus jugulare tumors (GJT), the effectiveness of surgery compared with radiotherapy as the primary treatment remains controversial. For managing such tumors, the infratemporal fossa type A (IFA) approach, with complete facial nerve transposition and a full jugular vein resection as described by Fisch in 1978, is considered as the standard [1]. In our center, the classic IFA was the surgical procedure performed in most of our patients during the last 20 years. Much of the criticism against the surgical management of these tumors is based on the morbidity rates associated with cranial nerve loss. Considering that GJT are benign, many surgeons have adopted the concept of maximal tumor removal without risking cranial nerve function, even if this means leaving some macroscopically identifiable disease in areas where removal could impair nervous preservation or increase surgical morbidity. Small residual lesions can later be irradiated if growth is observed during the follow-up period.

The preservation of the facial nerve function during GJT surgery is still a great challenge for surgeons, especially for large tumors with important involvement of the internal

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carotid artery, which require a large surgical exposition. Facial nerve damage is very traumatic both for the patients and their surgeons. In current days, intraoperative monitoring is considered mandatory for facial nerve preservation and the most important debate when considering GJT surgery is if a partial or inferior transposition of the nerve, or some times none at all, is sufficient to warrant good visualization of large tumors or if a complete transposition should always be performed as described by Fisch for the classic IFA approach [1–3].

The preservation of the lower cranial nerves (LCN) is also of great importance. Pharyngolaryngeal paralysis from Xth or Xth lower cranial nerve damage, producing either aspiration or dysphonia, is especially troublesome during the postsurgical recovery period and it may disturb the patient's quality of life in a serious manner [4]. Monitoring the LCN and having a good visualization and control of each one in the neck is paramount to obtain good results for functional preservation. In 2002, Al-Mefty and Teixeria introduced the intrabulbar removal technique for GJT, in which the anterior wall of the jugular bulb and internal jugular vein are kept in place. This technique aids in preserving the function of the LCN but may relate with higher recurrence of the disease [5, 6].

In this prospective study, we describe the neurological results obtained in six patients with large Fisch C GJT managed from 2015 to 2017 and with a follow-up of 8–30 months, in which a modified infratemporal type A approach and a nerve-preserving strategy were used. This nerve-preserving surgical technique was used to substitute the classic IFA approach performed in our center for the last two decades.

Materials and methods

The Research Ethics Committee and Institutional Review Board of the University of São Paulo Hospital das Clinicas approved this prospective study. Six patients with Fisch C GJT were operated in a tertiary referral center since 2015 and followed after surgery for an average of 18 months. We included every patient diagnosed with a GJT at Hospital das Clinicas during 2015 (14 patients) and excluded patients who had smaller GJT (Fisch A and B), patients who refused surgery or patients who were previously treated with radiation. The patients in this series became candidates for surgery when they developed symptoms of cranial nerve dysfunction, either facial paralysis or alterations of the LCN. A thorough neurological examination, nasopharyngolaryngoscopy, high-resolution CT scans of the temporal bone and multiplanar MR images, both with and without contrast, were obtained in every patient. All had a unilateral disease and underwent angiography and embolization 72 h before the surgical procedure (Fig. 1).

The nerve-preserving surgical technique for GJT removal employed in these cases used most of the main steps of the IFA approach as described by Fisch, but it was modified with the total removal of the tympanic bone to gain an anterior expansion which allows for complete control of the intratemporal carotid artery and a total exposition of the components of the jugular foramen. The facial nerve is either partially/inferiorly mobilized by completely removing the bone around it and slightly displacing it anteriorly from the second genu (together with some parotid tissue to preserve irrigation from the stylomastoid artery) or kept in place using a facial bridge technique (Fig. 2) [1]. The decision to move the nerve or to keep it in place depends on the amount of tumor included in the tympanic cavity, over the promontory and carotid, and under the facial nerve. With smaller involvement of these areas the tumor may be maneuvered under the facial without a partial/inferior transposition. The sigmoid sinus is opened and packed with absorbable hemostat (Surgicel®, Ethicon US, LLC). From the cervical approach, the Xth–Xth cranial nerves are identified and followed as close to the jugular foramen as possible (Fig. 3) and the jugular vein is sectioned after ligation at the level of the transverse process of C2.

The complete occlusion of the internal jugular vein at this stage usually causes some tumor swelling and increased bleeding. Tumor removal begins with the opening of the sigmoid sinus towards the bulb and progressive exposure of the jugular bulb, which is filled with tumor. Once complete visualization of the tumor is obtained, removal continues by dissecting its anterior part overlying the carotid artery and pushing it downwards and under the facial nerve towards the jugular bulb. A minimal anterior displacement of the facial nerve is sufficient, allowing for proper dissection with little risk of damaging the nerve in most cases. When the tumor is properly separated from the carotid, the outer (lateral) wall of the lower part of the sigmoid sinus is incised continuing along the jugular bulb and jugular vein. The tumor is then removed from inside the jugular bulb and sigmoid sinus. The medial venous wall of the jugular bulb, separating the tumor from the nerves is left intact to minimize manipulation of the LCN (Fig. 4). LCN monitoring aids when the surgeon is dissecting the tumor in the area of the jugular foramen to avoid unnecessary manipulation. The tumor is removed by a combination of suction and bipolar coagulation. After complete tumor removal, some bleeding in the remaining wall of the jugular bulb may come from the inferior petrosal sinus. Bleeding at this area or risky areas over the cranial nerves is better controlled with absorbable hemostat than with bipolar cauterization. Extensive coagulation of the anterior wall of the jugular bulb must be avoided.

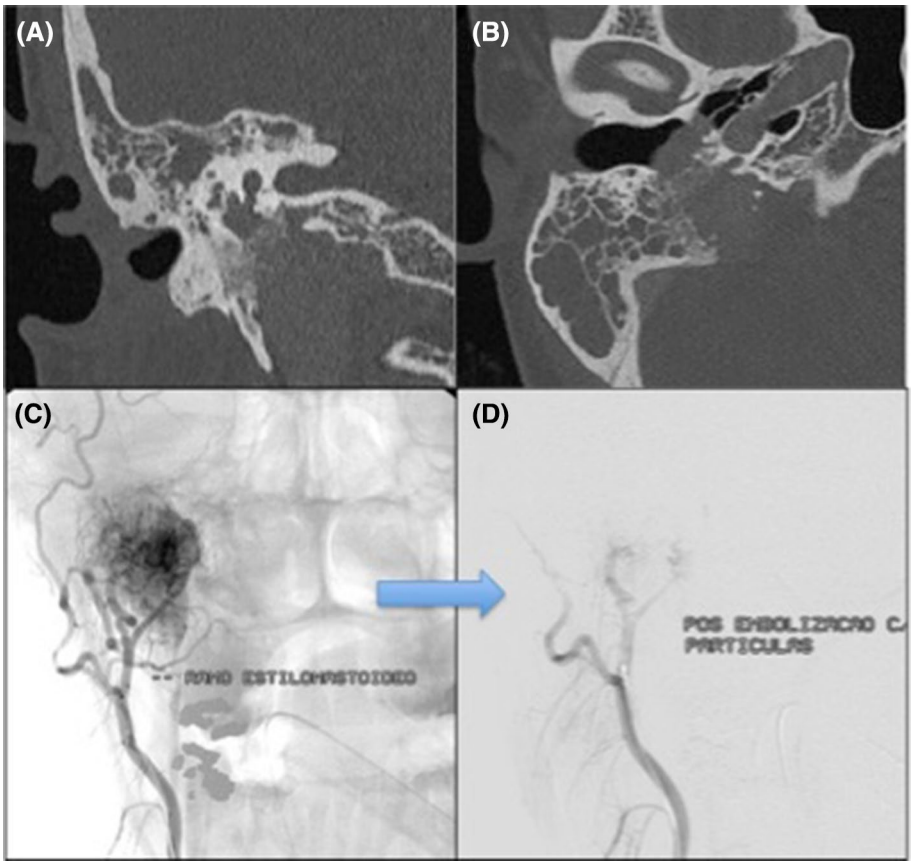

Fig. 1 CT images showing a glomus jugulare tumor without contrast in coronal (**a**) and axial (**b**) sections. Cerebral angiogram demonstrating the tumor before embolization (**c**) and after embolization (**d**)

The mastoid defect is packed with strips of abdominal fat and fibrin glue and the inferior portion of the temporalis muscle is then sutured to the sternocleidomastoid muscle. Blind sac closure of the external auditory canal was performed in all of the patients. The wound is closed in an ordinary fashion and regular dressing is maintained for 48 h.

Results

The six patients included had pulsatile tinnitus and either moderate or severe mixed hearing loss in the affected ear. Auditory symptoms had been present in these patients from 1 to 6 years earlier (3.5 years average). The gender distribution was 5 female patients and 1 male, with an age range from 23 to 58 years old (average 43.6). The average surgical bleeding in the six patients was 725 ml (range from 400 to 1600 ml). Gross total resection (GTR), defined as total removal of the lesion without residual tumor on the MR, was obtained in the six patients and it was determined during surgery and confirmed by postoperative MR images with fat suppression sequences obtained 1 week after surgery and 6 months later. In all our cases, we then perform MR yearly for the following 5 years after surgery and the six patients are currently in this stage.

Hearing decreased in all of the patients, mainly as an increase in air-bone gap related to the obliteration and blind sac closure of the ear canal. Bone hearing thresholds stayed the same in four of the six patients and decreased in patients 3 and 5 who had large Fisch C2 and C3 tumors with cochlear involvement. The course of cranial nerve function and new cranial nerve deficits are listed in Table 1. Patients 1, 4, 5 and 6 had facial paralysis before surgery. In patients 1 and 4, House–Brackmann (H–B) grades III and IV were seen, respectively, before surgery and were maintained immediately after surgery. After 6 months, these patients showed important improvement in facial nerve function and at the end of their follow-up both had a H–B grade II. Patients 5 and 6, who presented a H–B III before surgery, evolved to H–B V immediately after surgery and recovered to a H–B III after 6 months of follow-up. Patient number 2 preserved a normal facial function as observed prior to the surgical procedure and patient 3, who had normal facial function before surgery presented with facial paresis immediately after surgery, involving the inferior levels of the face but preserving complete eye closure. After 6 months, he had a HB grade II that stayed the same during posterior follow-up. In five of our patients a partial, inferior transposition of the mastoid portion of the nerve was performed and only in patient number 2, who had a Fisch C1 tumor and no previous

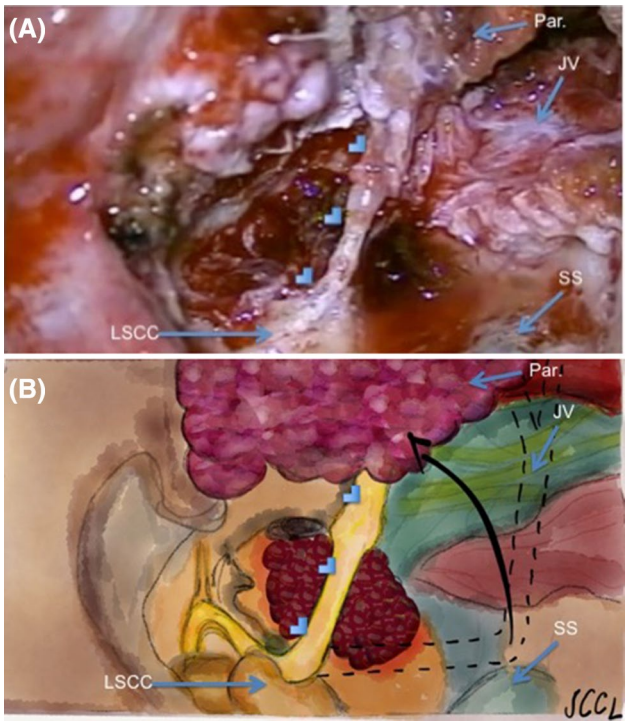

Fig. 2 An inferior transposition of the facial nerve in a right-sided surgery (**a**) and schematic drawing (**b**). The tympanic bone and mastoid tip are removed. Arrowheads show the mastoid portion of the facial nerve as it is anteriorly displaced together with the surrounding tissues of the stylomastoid foramen. The dotted lines in **b** show the normal position of the nerve. *LSCC* lateral semicircular canal, *SS* sigmoid sinus, *JV* jugular vein, *Par* parotid tissue

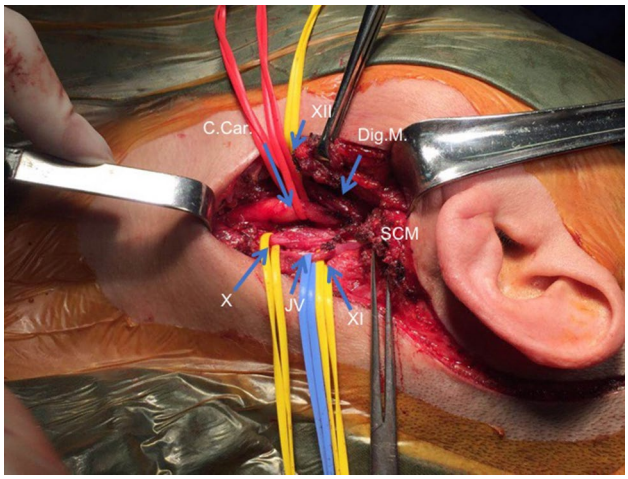

Fig. 3 Cervical dissection for exposure of the lower cranial nerves and vessels. *C.Car* common carotid, *Dig.M* posterior belly of digastric muscle, *JV* jugular vein, *X* vagus nerve, *XI* spinal nerve, *XII* hypoglossal nerve

facial paralysis where we able to maintain the nerve in its bony canal and work around it to remove the tumor.

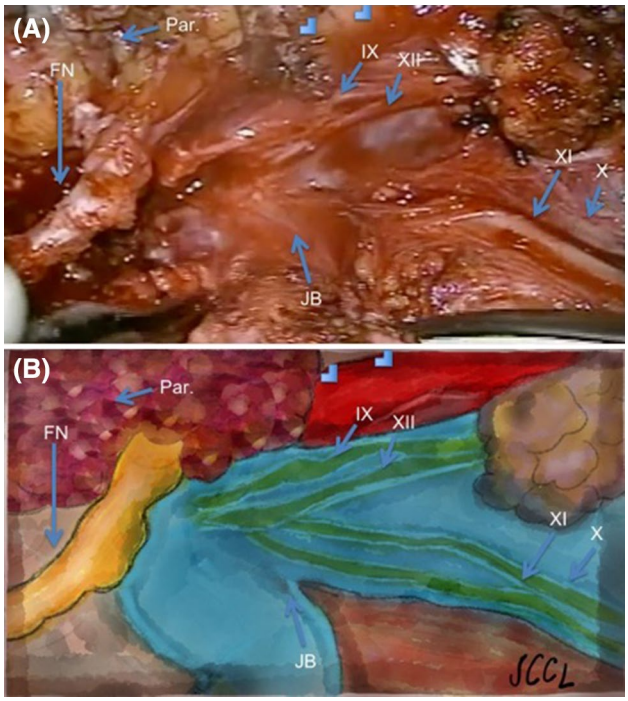

Fig. 4 The medial venous wall of the jugular bulb, separating the tumor from the nerves is left intact to minimize manipulation of the lower cranial nerves as seen in a right-sided surgery (**a**) and schematics (**b**). The facial nerve is partially transposed. The arrowheads show the internal carotid artery after tumor removal. *FN* facial nerve, *Par* parotid tissue, *JB* medial wall of the jugular bulb, *IX* glossopharyngeal nerve, *X* vagus nerve, *XI* spinal nerve, *XII* hypoglossal nerve

Four patients presented with dysphagia and required nasogastric tube placement to avoid aspiration during feeding. In two cases, a vocal fold paralysis was observed before surgery and in another only paresis. Another patient showed diminished pharyngolaryngeal sensibility preoperatively from glossopharyngeal nerve compromise, producing aspiration even with preserved vocal cord movements. The average time for removal of the nasogastric tube with return to normal oral feeding in these four patients was 4.3 weeks. All patients with preoperative Xth nerve damage showed an adequate compensation of the opposite vocal fold in postoperative period, without dysphonia or aspiration and without the need of other surgical procedures. The three patients with the largest tumors presented also with preoperative XIth cranial nerve impairment. In all of these patients, the nerve was identified and preserved. Rehabilitation exercises helped this patients regain function of the affected muscular groups showing a near normal function after 1 year. No patients presented alterations of the hypoglossal nerve function before or after surgery. The average follow-up period of 18.6 months (8–30 months) in this series is, unfortunately, relatively short.

Table 1 Case description and evolution after the surgical procedure

Px	Gender/age	Staging (Fisch[a])	Nerve function										Follow-up (months)/recurrence
			CN VII (H–B)		CN IX		CN X		CN XI		CN XII		
			Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	
1	F/54	C2	3	2	Nml	Nml	Imp	Com	Nml	Nml	Nml	Nml	30/No
2	F/47	C1	1	1	Nml	Nml	Nml	Nml	Nml	Nml	Nml	Nml	24/No
3	F/58	C2	1	2	Imp	Nml	Nml	Nml	Nml	Nml	Nml	Nml	22/No
4	M/34	C3	4	2	Nml	Nml	Imp	Com	Imp	Nml	Nml	Nml	18/No
5	F/46	C3	3	3	Nml	Nml	Nml	Nml	Imp	Nml	Nml	Nml	10/No
6	F/23	C3	3	3	Nml	Nml	Imp	Com	Imp	Nml	Nml	Nml	8/No

Px patient, *F* female, *M* male, *CN* cranial nerve, *H–B* House–Brackmann staging system, *pre* before surgery, *post* after the follow-up period, *Imp* impaired, *Nml* normal, *Com* compensated

[a]Fisch type C1 tumors have limited involvement of the internal carotid, type C2 are tumors invading the vertical portion of the carotid canal and type C3 have invasion of the horizontal portion of the carotid canal

Discussion

In 1977, Fisch described the infratemporal approach, a great improvement that allowed the removal of large jugular foramen tumors, especially GJT types C and D from his own classification [1, 7]. One of the main surgical steps described by Fisch is a complete anterior transposition of the facial nerve, from the geniculate ganglion to the extratemporal, intraparotid portions of the nerve. This allows for an excellent visualization of the infratemporal fossa with better tumor exposition and improved control of the internal carotid artery and LCN. On the other hand, this anterior facial nerve transposition increases the duration of the surgery, requires great surgical skills, and considering that the facial nerve might loose most of its extrinsic vascularity (from the stylomastoid and deep petrous arteries), depending only on its intrinsic vascular plexus after transposition, a certain degree of facial paralysis is almost guaranteed [8].

In 1987, Brackmann modified the technique proposed by Fisch, suggesting the mobilization of the nerve together with all of the surrounding tissues of the stylomastoid foramen avoiding damage to the stylomastoid artery. He practiced this technique in 32 patients with GJT obtaining H–B grades II and I in 86% [7]. With this technique other authors as Green et al. and Fayad et al. reported a complete removal of the tumor in 85 and 81% of their patients, respectively, with H–B I and II in 95% of the cases [9, 10]. In our institution, this same technique has been used for the last two decades obtaining comparable results, especially in smaller, Fisch B and C1 tumors. Operating smaller tumors, which demonstrate fast growth through serial imaging studies is a good option, since the degree of neural preservation in this cases is higher and it is easier to obtain a complete removal of the tumor without major risk of damaging important vascular structures. In our experience, morbidity for both the facial and LCN increases in an important manner when removing larger tumors (Fisch grades C2, C3 and D).

A useful surgical strategy to preserve facial nerve function with less mobilization of the facial nerve is the partial or inferior transposition of the facial nerve, which implies only mobilizing the mastoid segment of the facial nerve anteriorly. Shapiro, Neues and Capps described this technique and then it was made popular by Glasscock [11–13]. These authors suggest this technique for tumors with limited involvement of the internal carotid and with no intracranial extension (Fisch C1 and C2 grades at the most). Manolidis et al. in 1997 reported their surgical results with this technique in 58 patients obtaining H–B grades II and I in 92% with total tumor removal in 81% of the cases [14].

In 1996, Selesnick et al. presented a systematic review of the postoperative facial function comparing surgeries that included a complete anterior transposition with those where partial transposition was performed. They noted H–B grades I, II in 91% of the patients in which a partial transposition had been performed, compared to 74% of those with complete anterior transposition. It was not considered in this article that partial transposition was performed mostly for smaller tumor sizes, whereas most cases where complete transposition was performed had much larger tumors, which may have intimate contact with the mastoid portion of the nerve [15–17]. Five of our patients, which presented with large Fisch C2 and C3 tumors, were managed with the partial transposition technique. As noted by other authors, we believe that it carries a lower risk for postoperative facial paralysis than the long anterior rerouting allowing an adequate tumor visualization and complete removal.

Similar to what was reported by Selesnick et al., in 2001 Tran Ba Huy et al. compared the results of nine patients with GJT which needed a complete anterior transposition, nine with a partial or inferior transposition and 24 patients which did not need any nerve transposition to remove the tumor.

They observed H–B grades II or I in 97% of the 24 patients that did not need transposition and in 67% of the patients that needed some type of transposition [18].

The surgical technique to remove GJT maintaining the facial nerve in situ has been described by various authors. Al-Mefty et al. refined the infratemporal fossa approach in 1987 and he subdivided the approach in four types (A–D). In infratemporal approache types A–C, the nerve is preserved in its original position and preferably inside the Fallopian canal; Type D represents the classic complete anterior transposition described by Fisch [5, 19].

In 2010, Borba et al. reported their results in 34 patients with large GJT (Fisch C and D), operated with the techniques of no mobilization described by Al-Mefty. An infralabyrinthine, retrofacial approach (Type A) was used in 32.5% of the patients. An infralabyrinthine pre and retrofacial approach without occlusion of the external acoustic meatus (Type B) was performed in 20.5%. An infralabyrinthine pre and retrofacial approach with occlusion of the external acoustic meatus (Type C) was done in 41% and the Type D approach in only 6%. A complete resection was obtained in 91% of the patients obtaining a H–B I in 94.7% of the patients in which the nerve was maintained in the canal (Types A–C). Six patients (17.6%) developed lower cranial nerve deficit after surgery. In this series, it was shown that even when tumors are large, a very small amount (6%) might require a complete transposition [20]. Only one of our patients, who presented a C1 tumor, was managed keeping the facial nerve in its bony canal and performing a Type C surgery. This patient presented no alterations of facial nerve function before or after surgery but we must consider that this tumor was smaller than those of the other five patients.

In 1997, Pensak and Jackler promoted the fallopian bridge technique to manage GJT. Similar to what had been described by Al-Mefty, in this approach, tumor resection is obtained by drilling through the retrofacial air cells communicating the spaces anterior and posterior to the facial nerve. These authors reported complete gross tumor removal in 71% of the patients, preserving a H–B grade I facial function in 92% [21]. Different from Al-Mefty and Borba, who considered that GJT of any size may be removed leaving the facial nerve in situ, Pensak and Jackler suggested the facial bridge technique only for Fisch class C1 tumors or smaller [5, 16, 20, 21]. It is the authors' opinion that the facial bridge technique may diminish the possibility of complete gross tumor removal in cases with large tumors and that partial transposition improves visibility without a significantly increased risk of facial nerve damage. In one patient, we managed with this technique gross total tumor removal was obtained but it was a smaller tumor than the other five of our series.

As mentioned earlier, the surgical treatment of GJT must follow a series of surgical steps that improve the chances of gross tumor resection and functional preservation, not only for the facial nerve but also for the LCN. The adequate control of the internal carotid artery, the ligation and coagulation of afferent vascular structures before tumor removal, the proximal control of the sigmoid sinus and internal jugular vein and an adequate identification of the LCN are paramount steps in this procedure. Currently, some surgeons suggest a less aggressive surgical strategy with the following recommendations: (1) preservation of the facial nerve inside the Fallopian canal whenever possible; (2) preservation the internal carotid artery adventitia even when infiltrated by the tumor and the epineurium and perineurium of LCN; (3) sparing the infiltrated medial wall of the jugular bulb to preserve LCN as suggested by Al-Mefty and Teixeira; (4) abandoning gross tumor removal if important changes are observed during nerve monitoring [16, 20, 22]. We believe that the modification that impacted the most in the evolution of our patients was the preservation of medial-anterior wall of the jugular bulb since the morbidity related to LCN dysfunction in our patients was minimal.

In 2016, Li et al. presented their results in 51 patients with large GJT, with 37 patients classified as Di1 and Di2, in which they practiced the aforementioned less aggressive surgical strategy. Their follow-up was of up to 15 years with an average of 85.7 months. Total gross tumor resection was obtained in 51% of the cases and subtotal (defined as 95–99% of the quantified lesion volume removed) in 43%. Comparing with the preoperative stage, swallowing and facial function improved or stabilized in 96.1 and 94.1%, respectively. A H–B grade II or I was obtained in 84.3% of the patients. The tumor recurrence/ regrowth (*R*/*R*) rate was 11.8% and it was related mainly to the presence of pathologic mitosis observed during the histopathological analysis. Radiotherapy was recommended in every patient in which subtotal resection was performed and when the histopathological analysis showed pathologic mitosis. Overall, *R*/*R*-free survival and overall survival at 15 years were 78.9 and 80.6%, respectively [22].

In our center, the aim of surgery is the maximal elimination of GJT and the preservation of neurological function. For this reason, we consider it important to obtain an early diagnosis and perform surgery as early as possible. The nerve preserving surgical technique is promising, with good overall results for functional preservation. We obtained total gross tumor removal in all patients, still the absence of recurrence is not guaranteed because of our short follow-up. When subtotal resections are obtained radiotherapy is offered if regrowth is noticed during follow-up. Radiotherapy alone is offered as an alternative for patients with an unresectable GJT or those who are poor candidates for surgery, considering serious comorbidities or contraindications. In elderly patients (> 75 years old) with comorbidities, the tumor is left alone if it shows no growth and radiotherapy is

offered when growth is observed. We consider this to be an adequate treatment algorithm and patients are generally discharged if no growth is observed after a 10-year follow-up.

Conclusions

The nerve preservation surgical techniques used in these patients provide a high percentage of functional preservation with no recurrence observed during follow-up. None long lasting surgical morbidities were observed and patients were able to carry out normal life activities in shorter periods than what we observed with the classic, more aggressive IFA approach. Avoiding the complete anterior transposition of the facial nerve by minimally displacing it anteriorly carries a lower risk for postoperative facial paralysis and allows an adequate tumor visualization and removal. The preservation of the medial wall of the jugular bulb is an excellent strategy to avoid LCN damage.

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Compliance with ethical standards

Conflict of interest The authors declare no conflicts of interest.

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