Original Article

Surgical management of embolized jugular foramen paragangliomas without facial nerve transposition: Experience of a public tertiary hospital in Brazil

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ABSTRACT

Background: Jugular foramen paragangliomas (JFP) treatment represents a challenge for surgeons due to its close relationship with facial nerve (FN), lower cranial nerves (LCN), and internal carotid artery. Due to its hypervascularization, preoperative tumor embolization has been indicated.

Methods: Retrospective analysis of the clinical evolution of 26 patients with JFP class C/D previously embolized treated through infratemporal/cervical access without FN transposition.

Results: Total and subtotal resections were 50% each, regrowth/recurrence were 25%, and 23%, respectively, and mortality was 3.9%. Postoperatively, 68.4% of patients had FN House and Brackmann (HB) Grades I/II. New FN deficits were 15.4% post embolization and 30.7% postoperatively. Previous FN deficits worsened in 46.1%. Tumor involved the FN in 30.8% and in 62.5% of them these nerves were resected and grafted (60% of them had HB III). Lateral fall, ear murmur, and vertigo improved in all patients. Tinnitus improved in 77.8% and one patient developed tinnitus after surgery. Hearing loss did not improve, eight partial hearing loss remained unchanged and four worsened. New postoperative LCN deficits were 64.3%. Postoperative KPS between 80 and 100 dropped 8.3%. Two patients with secretory paragangliomas with arterial hypertension difficult to control had better postoperative blood pressure control.

Conclusion: Although still with significant morbidity due to FN and LCN injuries, the treatment of patients with JFP Fisch C/D has good long-term results. Surgical techniques without FN transposition have less intraoperative nerve damage, lower rates of total resection, and higher recurrence. Preoperative embolization of JFP reduces the intraoperative blood loss but can cause FN deficit.

Keywords: Clinical outcome, Jugular foramen paraganglioma, Preoperative embolization, Surgical treatment without facial nerve transposition

INTRODUCTION

Jugular foramen paragangliomas (JFP) constitute 22% of head and neck paragangliomas, 8.6% of temporal bone tumors and 80% of jugular foramen (JF) tumors.[11,12,24,33] They originate
from chromaffin cells of the neural crest and are called chemodectomas or paragangliomas.[11] Early surgical treatment of JFP dates back to 1952;[6] its deep location, complex regional anatomy, and hypervascularization became challenging this treatment. However, diagnosis improvements, the use of microsurgical techniques and better understanding of the regional anatomy, have made JFP resection possible.[1,4,16]

Technical details such as the anterior transposition of the facial nerve (FN) have been recommended,[5,10,11,32] allowing for wide exposure and control of the internal carotid artery (ICA); however, transposition can cause temporary/permanent facial paralysis which occurs in most cases.[9,28,32] Recent studies have suggested that even large tumors can be totally resected without FN transposition.[2,30,33] Preoperative JFP embolization has been advocated to reduce bleeding during tumor resection.[7,10,17,18,33,34,39] However, embolization can occlude the blood supply to low cranial nerves (LCN), making them more susceptible to injury during dissection. Although the surgical resection appears to be the most effective treatment for these tumors, technical details as previous embolization, the best approach or transposition or not of the FN are controversial.

We retrospectively analyzed the clinical evolution of a series of 26 patients with Fisch class C/D JFP previously embolized and surgically treated without transposition of the FN.

**MATERIALS AND METHODS**

**Patient population**

This study is a retrospective analysis of 26 patients with diagnosis of JFP surgically treated at the Division of Neurosurgery, Hospital das Clínicas, Ribeirão Preto Medical School, University of São Paulo by a multidisciplinary team, from February 2006 to January 2020. The study was approved by the Research Ethics Committee of our institution (No. 736,988). The diagnosis was made using non enhanced and enhanced brain and temporal bone computed tomography (CT) and magnetic resonance imaging (MRI) and confirmed by histopathological examination. The epidemiological characteristics and clinical manifestations are shown in [Table 1].

All patients were submitted to urine metanephrine dosages and more recently also to dosage of catecholamines and vanillylmandelic acid. Two patients who had arterial hypertension difficult to control had increased metanephrine dosage requiring preoperative beta-blockers administration.

**Surgical treatment**

The patients underwent microsurgery, using four variations of the intrabulbar infratemporal approach (IFTA)-A[1,4,11,13] individualized for each patient based on their symptoms and on the tumor extension.[4] Type A. Retrofacial and

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**Table 1:** Demographic characteristics, preoperative clinical signs, and symptoms and urine metanephrines measurement in 26 patients with glomus jugulare paragangliomas.

| Age | Range: 14–65 years (mean 44.6 ± 13.3 years, median 46.5 years) |
|-----|---------------------------------------------------------------|
| Follow-Up | Range: 1–142 months (mean 55.6±40.6 months, median 44 month) |

≥25 meses: 20 (76.9%) patients

| Sex | Nº (%) |
|-----|--------|
| Female | 20 (76.9) |
| Male | 6 (23.1) |

**Clinical signs**

| Hearing loss | 18 (69.2) |
| Facial palsy | 14 (53.9) |
| Dysphagia | 13 (50.0) |
| Hoarseness/Dysphonia | 11 (42.3) |
| Drop shoulder | 09 (36.4) |
| Tinnitus | 07 (26.9) |
| Tongue hemiatrophy | 07 (26.9%) |
| Tongue deviation | 05 (19.2) |
| Vertigo | 04 (15.4) |
| Parotid gland bulging, otorrhea, facial pain | 03 (11.5 each) |
| ECM palsy, tearing | 02 (07.7 each) |
| Cervical pain, headache, hemifacial | 02 (03.9 each) |
| Hyperesthesia, lateral fall, otalgia, otorrhagia, mandibular deviation, murmur in the ear, nausea and vomiting, snoring, apnea breathing difficulty, tongue fasciculation, and dysarthria |  |

**Cranial nerve palsy**

| V | 03 (11.5) |
| VI | 01(3.9) |
| VII | 14 (53.5) |
| VIII | 18 (69.2) |
| IX/X | 16 (61.5) |
| XI | 09 (34.6) |
| XII | 10 (38.5) |

**Otoscopic examination**

| Normal | 19 (73.1) |
| Tumor in the external acoustic meatus/middle ear | 07 (26.9) |

**Tonal audiometry**

| Normal | 12 (46.1) |
| Mild hypoacusis | 01 (03.9) |
| No useful hearing (>60 db) | 13 (50.0) |

**Systemic arterial hypertension**

| Controlled | 03 (11.5) |
| Uncontrolled | 02 (07.7) |

**Measurement of metanephrines in urine**

| Normal | 24 (92.3) |
| Increased | 02 (07.7) |
IFTA-A, with the internal auditory canal (IAC) and the middle ear (ME) structures kept intact. This was indicated for tumors with/without neck or intracranial extension and without ICA irrigation or an upper extension Type B. Pre-/retrofacial IFTA-B, without IAC occlusion and with removal of the ME structures. The tumor removal is performed anterior and posterior to the FN maintained in the bone canal (fallopian bridge). In patients with facial paralysis, the canal is opened for decompression or grafting of the nerve. This approach is indicated for tumors with/without neck or intracranial extension and with ICA irrigation Type C. Pre-/retrofacial IFTA-C with IAC occlusion and resection of the ME structures. This approach allows tumors with anterior and superior extensions and those with extensions around the ICA to be reached. The FN is maintained in its bone canal Type D. IFTA with transposition of the FN (IFTA-D). This approach is used for tumors that have destroyed the FN canal and allows the most extensive exposure of the temporal bone and JF. Twenty-five (96.1%) patients underwent surgery for the 1st time, and one (3.9%) patient underwent surgery 10 years ago elsewhere.

Surgical technique

The technique used was reported early,[4] with changes in opening and closing in the last 20 patients [Figure 1]. The incision was made in two planes, one involving the scalp and the galea, and the other involving the temporal and sternocleidomastoid muscles and the pericranium, exposing the temporal bone and the mastoid process [Figure 1c]. The anterior cutaneous-muscular flap is folded anteriorly to expose the external acoustic canal (EAC), the posterior root of the zygomatic arch and the mastoid tip [Figure 1d]. Mastoidectomy starts with the removal of the cortical bone [Figure 1e and f], followed by the usual approach. The cervical exposure and tumor resection are similar to those previously described[4] [Figure 1g and h]. The dura, if opened, is closed with a fascia lata or a pericranium graft. The mastoid cavity is filled with fragments of abdominal fat to eliminate dead space and it is held in place with commercial fibrin glue. The external carotid artery (ECA) branches supplying the tumor are not systematically ligated during cervical dissection because their usual embolization. The cortical bone of the mastoid is replaced and fixed with miniplates and screws [Figure 1i]. The sternocleidomastoid muscle attached to the temporal fascia is replaced in its original position and sutured to the anterior part of the fascia/temporal muscle [Figure 1j], followed by suturing of the scalp flap.

Facial and LCN monitoring was used routinely since 2015 (last 12 cases) and auditory evoked potential is used for patients that have useful hearing. No preoperative traqueostomy or gastrostomy was preoperative used and no prophylactic lumbar external drainage was routinely used even in cases of intradural extension of the tumor.

Figure 1: Steps for surgical approach of an infratemporal (IFTA-B) and cervical approach of a paraganglioma of the jugular foramen (JF) C3. (a and b) Preoperative axial T1-weighted contrast-enhanced MR images showing a large mass (Tu) located in the JF with extension to the cervical region and posterior fossa, pushing the dura-mater. (c) Skin incision. (d) Exposure of the temporal bone and mastoid process after anterior displacement of the skin and of the fascio/muscular (temporal and sternocleidomastoid muscles) flaps. (e) Cortical bone of the mastoid (c) separated and keep in place. (f) Cortical bone (c) of the mastoid displaced. (g) Mastoidectomy with a fallopian bridge (FB) and cervical exposure. (h) Magnification of the fallopian bridge (FP) after bone removal. (i) Cortical of the mastoid replaced and fixed with mini-plates and screws. (j) Suture of the fascio-muscular plane.
Extension of resection
The extension of resection was estimated macroscopically during surgery and by a neuroradiologist (pre- and postoperative CT and/or MRI comparisons). Recurrence/progression was assessed radiologically in the first 3 and 6 postoperative months and then annually/biannually.

Clinical outcome
The evolution of cranial nerve (CN) deficits and the Karnofsky Performance Scale (KPS) index were evaluated. FN lesions were classified according to the HB scale. KPS indices were obtained before, 3 months after surgery and at the last follow-up. Global survival (GS) and recurrence-free survival (RFS) curves were also assessed.

Statistical analysis
Chi-square and Fisher tests were used to compare proportions and the log-rank test (Mantel-Cox) was used to compare curves and survival estimates. Error probabilities not greater than 5% were considered significant for two-tailed probability tests. The tests were performed using the GraphPad PRISM program (version 9.0.0; GraphPad Software, Inc., San Diego, CA, USA).

RESULTS
[Table 2] shows the preoperative CT/MRI findings. The tumor reached the ICA in the petrous bone in 19 (73.1%) patients. All patients underwent preoperative embolization by catheterization of the ECA branches or by direct intratumoral injection of particles of polyvinyl acetate (PVA) or onyx and occlusion of the ECA branches with coils or histoacryl. Time elapsed from embolization to surgery was 24 h in 11 (42.3%), 48 h in 5 (19.2%), 72 h in 2 (7.7%), 96 h in 6 (23.1%), and more than 96 h in 2 (7.7%).

Based on the CT/MRI images, the JFP was classified according to Fisch[15] into class C (carotid foramen involvement): C1 in 5 (19.2%), C2 in 3 (11.5%), C3 in one (3.9%), and in class D (presence of intracranial tumor): De1 in 9 (34.6%), De2 in one (3.9%), Di1 in 4 (15.4%), and Di2 in 3 (11.3%).

Surgical treatment
The results of the surgical treatment are summarized in [Tables 3 and 4]. Intraoperative (26 patients) and radiological (24 patients) assessments showed total resection in 69.2% and 45.8%, respectively, \( P = 0.1513 \), Fisher exact test. Total resection in patients with radiological involvement of the ICA was 31.6%. There was no difference between the GS and RFS curves according to the type of approach (\( P = 0.95 \) and \( P = 0.66 \), respectively), or the extension of resection (\( P = 0.38 \) and \( P = 0.95 \), respectively). The relationship of extension of embolization and extension of resection is shown in [Table 3]. There was no significant difference between the numbers of total resection for patients submitted to total versus subtotal and total versus partial embolization (\( P = 0.14 \) and \( P = 1.00 \)).

| CT/MRI** | CT | MRI |
|----------|----|-----|
| Tumors location | Bone erosion (N° of Patients) | Tumor (N° of Patients) |
| Jugular foramen | 26 | 26 |
| Middle ear | 07 | 20 |
| Hypotympanum | 03 | 16 |
| Carotid foramen | 19 | 19 |
| Internal carotid artery contact | 19 |
| Vertical portion | 16 |
| Horizontal portion | 03 |
| Posterior fossa | 16 |
| Extradural | 11 |
| Intradural | 05 |
| External acoustic meatus | 02 | 09 |
| Internal acoustic meatus | 01 | 04 |
| Glenoid cavity | 02 | 02 |
| Cavernous sinus | 02 |
| Mastoid cells | 01 | 01 |
| Middle Fossa | 02 | 01 |
| Cochlea | 01 |
| Semicircular canals | 01 |
| Petrous apex | 01 |
| Middle ear bones | 01 |

*Calcifications (N° of Patients)

| Angiography | N° (%) |
|--------------|--------|
| Tumor supply |        |
| ECA* | 26 (100.0) |
| ICA§§ | 09 (34.6) |
| VA§§§ | 7 (26.9) |
| Other findings |        |
| ICA |        |
| Normal | 24 (92.3) |
| Stenotic | 02 (07.7) |
| Sigmoid Sinus |        |
| Previous | 02 (07.7) |
| Occluded | 10 (38.5) |
| Ectatic | 02 (07.7) |
| Baloon occlusion test (15 patients) |  |
| Collateral circulation | 10 (38.4) |
| No collateral circulation | 05 (19.2) |

*CT: Computed tomography, MRI: Magnetic resonance imaging, §ECA: External carotid artery, §§ICA: Internal carotid artery, §§§VA: Vertebral artery
Table 3: Treatment of 26 patients with glomus jugulare paragangliomas according Borba et al., 2010[4] approaches classification.

| Tumor classification (Fish[15]) | Evaluation of Extent of Tumor Resection | Radiological (24 Patients) |
|-------------------------------|----------------------------------------|---------------------------|
|                               | Intraoperative (26 Patients)           |                           |
|                               | N° (%) | N° (%) | N° (%) | N° (%) |
| Total                         | 4 (66.7) | 2 (23.3) | 1 (20.0) | 4 (80.0) |
| Type A                        | 10 (76.9) | 3 (23.3) | 7 (58.3) | 4 (41.7) |
| Type B                        | 4 (80.0) | 1 (20.0) | 2 (40.0) | 3 (60.0) |
| Type C                        | 0 (0.0) | 2 (100.0) | 1 (50.0) | 1 (50.0) |
| Total                         | 18 (69.2) | 8 (30.8) | 11 (45.8) | 13 (54.2)† |

**Extent of embolization**

| Radiological extent of resection (24 Patients) |
|-----------------------------------------------|
| N° (%) | N° (%) | N° (%) |
| Total  | 1 (9.1) | 5 (38.5) | 6 (25.0) |
| Subtotal | 9 (38.5) | 5 (19.2) | 14 (58.3) |
| Partial | 1 (9.1) | 3 (23.1) | 4 (16.7) |
| Total  | 11 (45.8) | 13 (54.2) | 24 |

Blood loss: (21 (80.8%) Patients mean=1152.1 cc±828.8 median=850 cc

Radiotherapy

| Modality                              | N° (%) | Results                                      |
|---------------------------------------|--------|----------------------------------------------|
| Adjuvant IMRT                         | 1 (3.9) | Tumor reduction (61 months)                  |
| FRT** for tumor progression           | 1 (3.9) | Tumor reduction (70 months)                  |
| FRT for tumor progression             | 1 (3.9) | Death (1 month)                              |
| Primary FRT (Surgery postponed due to stenosis of the contralateral IJV/SS§) | 1 (3.9) | Tumor progressions (20 months)               |

Outcome (25 patients with >3 months follow-up)

| Survival rates | Total resection | Subtotal resection | Total |
|----------------|----------------|--------------------|-------|
| 1 year         | 12 (100%)      | 12 (92.3%)         | 24 (96.0%) |
| 5 years        | 7 (66.7%)      | 5 (38.5%)          | 13 (52.0%) |
| 10 years       | 3 (25.0%)      | 1 (7.7%)           | 4 (20.0%) |
| Recurrence free estimates              |               |                    |       |
| 1 year         | 12 (100.0%)    | 12 (92.3%)         | 24 (92.3%) |
| 5 years        | 7 (58.3%)      | 5 (38.4%)          | 12 (48.0%) |
| 10 years       | 1 (8.3%)       | 2 (15.4%)          | 2 (8.0%) |
| Recurrence rate (All follow-up)        | 6 (24%)        |                    |       |

Mortality

| Global (Along the follow-up) | 3 (11.5%) |
| 1 month (Operative mortality) | 1 (3.9%) |

Non neurological complications

| Complications                     | N (%) | Treatment                                      |
|-----------------------------------|-------|-----------------------------------------------|
| ICA^ Laceration (tumor invasion)  | 2 (7.7) | ICA Compression/ligation                      |
| Cerebrospinal Fluid Fistula       | 1 (3.9) | Reoperation+dural closure                     |
| Bulging                           | 1 (3.9) | External lumbar drainage                      |
| Wound infection                   | 2 (7.7) | Surgical debridement                          |
| Scar pain                         | 1 (3.9) | Medical Treatment                              |
| Glottis edema                     | 1 (3.9) | Transient reintubation                         |
| Scalp necrosis/Titanium mesh exposure | 1 (3.9) | Surgical scalp flap rotation                   |
| Sigmoid sinus laceration          | 1 (3.9) | Sinus ligation                                 |

^No significant difference between intraoperative and radiological estimations (P=0.5663, Fisher’s exact test), †No significant difference between numbers of resection for total versus subtotal embolization (P=0.1329, Qui-square test). IMRT: Intensity-modulated radiation therapy, **FRT: Fractionated radiation therapy.
| Initials | Sex | Age | Preop CN | Preop ANGIO | Embolization Arteries/Agent | Embolization OcclusionPost-Emb. CN | Tu Clasification (Fish) | IOBL Postop CN (ml) | Non-Neurological Complication | Extent of Resection | Radiotherapy | Recurrence | KPS Index Preop | KPS Index Postop | KPS Index Final |
|----------|-----|-----|----------|-------------|-----------------------------|-----------------------------------|---------------------------|----------------------|--------------------------|------------------|--------------|------------|--------------|----------------|----------------|
| OVM      | M   | 63  | VII, ECA | IX, X, (APA) XI, XII | APA/PVA Total | A-C1/IFTA-A | NF-HB V | Subtotal | Yes | No | 100 | 80 | 90 |
| ZSSF     | F   | 46  | II, V, ECA VI, (APA, VIII, AO), IX, X, XII | APA, AO./ Total PVA | A-C1-Di1/IFTA | IX, X | Titanium mesh exposure, Scalp necrosis-Reoperation | Subtotal | Yes | Yes | 80 | 80 | 80 |
| IMP      | F   | 56  | NF- ECA HB II, (APA, VIII, IMA, IX, ICA, VA, X, XII (ACAD) | Direct puncture/ EVOH | Total | A-C1-Di1/491 NF-HB II IFTA-A | YNA | 80 | Death |
| TMFB     | F   | 36  | NF- ECA HB II, (APA, VIII, OA) IX, X, XII | APA, AO/ Total PVA/Direct puncture/ NBCA | A-C1-Di1/587 V, VI, NF-HB IV, VIII, IX, X | SS Laceration | Subtotal | Yes | Yes | 90 | 90 | 90 |
| RAM      | M   | 51  | NF- ECA HB II (APA, AO) PVA | APA, AO/ Subtotal | A-C2-De1/1302 IFTA-A | NF-HB V, IX, X (dysphagia, improvement), XII ICA | Total | No | Yes | 100 | 100 | 100 |
| MRS      | F   | 25  | VIII, ECA IX, X, (APA, PVA/ XI, IMA), XII ICA, (Oph A) VA | APA, IMA/, Subtotal | A-C2-De1/IFTA V, VIII, IX (worsened), X ICA XI, (Transient) Laceration | Subtotal | Yes | No | 90 | 70 | 80 | Death |
| EWDV     | F   | 32  | VIII, ECA (APA, OA, PAA/PVA | APA, OA, Subtotal | A-C2-De1/ IFTA-A 850 NF-HB I (transient), VIII, IX, XI | Subtotal | Yes | IRMT-Adj | Yes | 70 | 70 | 70 |

(Contd.)
### Table 4: (Continued)

| Initials | Sex/Age | Sex/Age | Preop CN | Preop Angiography | Arteries/Agent | Embolization | Tu Classification (Fish) | IOBL Postop CN (ml) | Postop CN | Preop CN | Non-Neurological Complication | Extent of Resection | Radiotherapy | Recurrence | KPS Index |
|----------|---------|---------|----------|-------------------|----------------|--------------|------------------------|------------------|------------|----------|----------------------------|------------------|--------------|-----------|----------|
| MRFC     | fF/51   | VIII, ECA (APA, PAA) | APA, PAA/PVA | Subtotal | B-C1/IFTA-B | 1250 NF-HB I (transient) | Total | Yes | No | 100 | 100 | 100 |
| LSC      | M/49    | VIII, ECA (APA, OA), VA | APA, OA/ Gelatine | Total | B-C1/IFTA-B | VIII, IX, X, XII | Subtotal | Yes | No | 100 | 100 | 100 |
| SPSR     | F/47    | VIII, ECA (APA, OA), VA | APA, OA/ PVA | Total | B-C1-Di1/ IFTA-B | 1250 V, VI, VII HB V, VIII, IX, X, XII | Total | No | FRT Progression | Yes | 70 | 70 | 70 |
| MABO     | F/59    | NF-HB V, III, VIII, IX, X, XI, XII, ECA (APA), Direct puncture, APA, PAA< MMA/PVA, Gelatine, NBCA | Direct Subtotal | Total | B-C2/IFTA-B | 528 NF-HB III, VIII IX, X | Total | No | Yes | 80 | 80 | 80 |
| NCS      | F/43    | NF-HB (APA), PAA | Subtotal | B-C2/IFTA-B | 505 NF-HB V, VIII | Total | No | 100 | 100 | 80 |
| CSP      | F/43    | VIII, ECA (APA, OA, PAA) | Subtotal | B-C2-De1/ IFTA-B | 2219 NF-HB I (transient), XII | Total | No | 100 | 100 | 100 |
| LHAR     | M/14    | VIII, ECA (APA), ICA | APA/PVA | Subtotal | B-C2-De1/ IFTA-B | 2852 NF-HB I (transient), VIII | Total | No | 100 | 80 |
| ASHF     | F/25    | NF-HB (APA, OA, PAA, IMA, AO, VIII, PAA) | APA, IMA, Subtotal | B-C2-De1/ IFTA-B | 1595 NF-HB III | Total | Yes | 80 | 80 | 80 | 80 |

(Contd...)
### Table 4: (Continued)

| Initials | Sex/Age | Preop Arteries/Agent | Preop Embolization | Tu Classification (Fish) Surg. Approach/Date of Surgery | IOBL| Postop CN | Non-Neurological Complication | Extent of Resection | Radiotherapy Recurrence | KPS Index |
|-----------|---------|----------------------|--------------------|--------------------------------------------------------|-----|-----------|--------------------------------|---------------------|------------------------|-----------|
| AMGR      | Chronic Pain | NT, (APA, OA), VA | Partial NT  | B-C2-D1/ IFTA-B, APA, STA, ECA (APA)/VA ICA/EVOH, | 505 | B-C2-D1/ | Subtotal | Total | NA | 100 |
| IGD       | F/39    | NT, (APA, OA), ICN | Partial NT | B-C2-D1/ IFTA-B, APA, STA, ECA (APA)/VA ICA/EVOH, | 2219 | B-C2-D1/ | Subtotal | Total | NA | 100 |
| AMM       | F/39    | NT, (APA, OA), ICN | Partial NT | B-C2-D1/ IFTA-B, APA, STA, ECA (APA)/VA ICA/EVOH, | 2852 | B-C2-D1/ | Subtotal | Total | NA | 100 |
| TAS       | F/31    | NT, (APA, OA), ICN | Partial NT | B-C2-D1/ IFTA-B, APA, STA, ECA (APA)/VA ICA/EVOH, | 980 | B-C2-D1/ | Subtotal | Total | NA | 100 |
| MHO       | F/62    | NT, (APA, OA), ICN | Partial NT | B-C2-D1/ IFTA-B, APA, STA, ECA (APA)/VA ICA/EVOH, | 791 | B-C2-D1/ | Subtotal | Total | NA | 100 |
| RHDM      | F/42    | NT, (APA, OA), ICN | Partial NT | B-C2-D1/ IFTA-B, APA, STA, ECA (APA)/VA ICA/EVOH, | 2000 | B-C2-D1/ | Subtotal | Total | NA | 100 |
| MLF       | F/32    | NT, (APA, OA), ICN | Partial NT | B-C2-D1/ IFTA-B, APA, STA, ECA (APA)/VA ICA/EVOH, | 3352 | B-C2-D1/ | Subtotal | Total | NA | 100 |
| Initials | Sex/Age | PreopCN | PreopAN | Angiography | Embolization | Tu Classification (Fish) | IOBL, PostopCN (ml) | Non-Neurological Complication | Extent of Resection | Radiotherapy Recurrence | KPS Index |
|----------|---------|---------|---------|-------------|--------------|--------------------------|---------------------|-----------------------------|-----------------------|--------------------------|-----------|
| SCC      | F/38    | NF-HB I | ECA     | APA, AO, ICA | Subtotal     | C-C1/IFTA-C (transient), VII, IX, X | 262 NF-HB V (transient), VIII, IX, X | Cervical pain Total Yes FTR-1° Therapy | No         | 90 90 90              |
| TAS      | M/36    | IX, X, ECA XI (APA) | APA, AO, ICA | Subtotal     | C-C1 /De2/IFTA-C/SNG | 1250 NF-HB V, IX, X, XI | Subtotal Yes No         | 90 90 80              |
| MGPG     | F/65    | VIII    | ECA (APA), ICA | APA/PVA, Onix* | Subtotal     | C-C2/IFTA-C | 750 VIII | Total No No | 90 70 90              |
| LRPO     | F/48    | NF-HB II,(APA, OA, PAA/PVA, VIII, PAA), XI ICA, VA | APA, OA, ICA, ECA | Subtotal     | C-C2-Di1, IFTA-C-AMNG | III, V, NF-HB III, VIII, XII | Total Yes FTR-Progression Yes | 80 80 80              |

Adj: Adjuvant, AMNG: Auricular magnus graft, APA: Ascending pharyngeal artery, AICA: Anterior inferior cerebellar artery, AMI: Acute myocardial infarct, CN: Cranial nerves, CSE: Cerebrospinal fluid, ECA: External carotid artery, ELD: External lumbar drainage, Emboliz: Embolization, EVOH: Etilen-Vinil-Alcohol, Decr: Decrease, FRT: Fractionated stereotactic radiotherapy, HB: House and Brackmann, ICA: Internal carotid artery, IFTA-A: Infra-temporal retrofacial, IFTA-B: Infra-temporal pre- and retrofacial, IFTA-C: Infra-temporal with IAC occlusion and resection of ME structures, IMA: Internal maxillary artery, IMRT: Intensity-modulated radiation therapy, IO: Intraoperative, IOBL: Intraoperative blood loss, KPS: Karnofsky performance scale, MMA: Middle meningeal artery, NA: Not available, NBCA: N-butyl-2 cyanoacrylate, NET: Nasoenteric tube, OA: Occipital artery, OnyxTM-Medtronic, OphA: Ophthalmic artery, PAA: Posterior auricular artery, PICA: Posterior inferior cerebellar artery, PCA: Posterior cerebral artery, Postop: Postoperative, Preop: Preoperative, PVA: Polyvinyl alcohol, RL: Residual lesion, SNG: Sural nerve graft, SS: Sigmoid sinus, STA: Superficial temporal artery, Tu: Tumor, VA: Vertebral artery
The overall recurrence rate was 25%, and the progression rate was 23.1%. Operative and overall mortality were 3.9% and 11.5%, respectively.

Non-neurological complications occurred in 9 (34.6%) patients [Tables 3 and 4]. Bleeding control due to ICA laceration during tumor dissection was done with oxidized cellulose in one patient and with ICA trapping in another. In both cases, the balloon occlusion test showed adequate collateral circulation. One patient with late postoperative infection (at 4 months) died. Other complications did not interfere with the patient's final outcome. One patient operated before the modifications introduced on the surgical technique (opening and closure of the surgical wound) developed a cerebrospinal fluid (CSF) leak through the nose and through the EAC requiring surgery for closure of a small dural tear; another patient developed a wound bulging (operated after the modifications) requiring external lumbar drainage. Both patients had no intracranial invasion.

CN outcome

The CN deficits in 25 patients with follow-up ≥3 months are shown in [Tables 4 and 5]. Hemifacial pain and V CN motor deficits improved, but hemifacial hypoesthesia/dysesthesia remained unchanged after surgery. The only VI CN lesion improved postoperatively.

Thirteen (50%) patients had preoperative FN deficits. Good FN function was observed in 68.4% (10 HB 1 and 3 HB II), 5 had HB III, 2 HB IV, and 7 HB V. New postoperative FN deficits occurred in 30.7%, worsened of the previous deficits occurred in 46.1% and improvement occurred in 15.4%. Tumoral involvement of the FN occurred 28.6%; 62.5% of them was grafted (two patients with sural and three with auricular nerves) and 60% reach HB III and 40% HB V. Two patients (15.4%), one sub totally and the other partially embolized, had FN lesions after embolization, one of which was transient.

New postoperative VIII CN deficits occurred in 8%. Lateral fall, ear murmur and vertigo improved in all patients. Tinnitus improved in 77.8% and postoperatively it occurred in one patient. Hearing loss improved in 4.6%, remained unchanged in 77.3% and worsened in 13.6%.

Nine patients (64.3%) had new postoperative LCN deficits, one from IX/X, 2 from XI, and 6 from XII CN [Table 4]. All patients with pre- or post-operative dysphagia showed some functional improvement; however, the neurological deficits (deviation of the palate, and abolition of the vomiting reflex)

### Table 5: Treatment and clinical outcome of 26 patients with embolized jugular foramen paraganglioma underwent surgical treatment.

| Cranial preoperative nerves | Preoperative Normal | Deficits | Post embolization | Postoperative Normal | Postoperatively improved | Unchanged | Worsened | New |
|----------------------------|---------------------|----------|-------------------|----------------------|-------------------------|----------|----------|------|
| II                         | 24                  | 1 (4.0%) |                   | 24                   |                         | 1 (4.0%) |          | 1 (3.9%) |
|   III                      |                     |          |                   |                      |                         |          |          | (Transient) |
| V                          | 22                  | 3 (12.0%)|                   | 22                   | 1 (4.0%)                |          | 2 (8.0%) |      |
| VI                         | 24                  | 1 (4.0%) |                   | 24                   | 1 (4.0%)                |          |          |      |
| VII                        | 13                  | 13 (50.0%)|(15.3%)(1 Transient)| 10 HB I*| 2 (8.0%)|          | 7 (28.0%) | 6 (46.1%) | 4 (30.7%) |
|                           |                     |          |                   | 3 HB II              |                         |          |          |      |
| VIII                       | 3                   | 22 (88.0%)|                   | 1                    | 1 (4.0%)                |          | 17 (68.0%) | 3 (12.0%) | 2 (8.0%) |
| IX/X                      | 11                  | 14 (60.0%)|                   | 14                   | 14 (56.0%)              |          |          | 1 (4.0%) |
| XI                         | 16                  | 9 (36.0%)|                   | 14                   | 1 (4.0%)                |          | 8 (32.0%) | 2 (8.9%) |
|                           |                     |          |                   |                      |                         |          |          | (Trapezius paresis) |
| XII                        | 15                  | 10 (40.0%)|                   | 9                    | 1 (4.0%)                |          | 9 (36.0%) | 6 (24.0%) |
|                           |                     |          |                   |                      |                         |          |          | (Tongue fasciculation) |

| KPS**                      | Score               | >70 | 70 | 80–100 | Mean±SD |
|----------------------------|---------------------|-----|----|--------|---------|
| Preoperative               | 1 (3.9%)            | 2 (7.7%) | 25 (92.3%) | 85.8±8.3 |
| Postoperative              | 2 (7.7%)            | 24 (83.5%) |            | 83.3±7.3 |
| KPS Changes               | 10 Points ≥ 20 Points | Total (26) |  |  |  |
| Improved                   | 2 (7.7%)            | 0    |  | 2 (7.7%) |
| Unchanged                  | 14 (53.8%)          | 0    |  | 14 (53.8%) |
| Worsened                   | 8 (30.8%)           | 2 (7.7%) | 10 (38.5%) |  |
| Total                      | 24 (92.3%)          | 2 (7.7%) | 26 |  |  |

*HB: House and Brackmann, **KPS: Karnofsky performance scale
persistent, as did patients with atrophy of the trapezius muscle and of the tongue. One patient with deltoid paresis and another with tongue fasciculation improved after surgery. Patients with vocal cord paralysis and dysphonia had partial functional recovery.

The evolution of the KPS indices is shown in [Table 5]. There was an 8.3% decrease (one death and one worsening) and an 8.3% improvement in the postoperative number of patients with KPS between 80 and 100. 45.8% remained unchanged, 8 (33.3%) worsened by 10 points, and 8.3% worsened by 20 points or more.

Both patients with secretory paragangiomas with arterial hypertension have better postoperative blood pressure control.

Radiotherapy

Three (11.5%) patients underwent fractionated stereotactic radiotherapy, two as rescue treatment and one as the first treatment (surgery was postponed due to stenosis of the contralateral sigmoid sinus); it was performed after post radiation tumor progression and verification of collateral circulation through the paravertebral plexus. A fourth patient underwent adjuvant intensity-modulated radiation therapy [Table 3]. Two patients had reduction occurred in two (one adjuvant and another rescue therapy), one patient had tumor progression (first treatment) and another died (rescue therapy).

DISCUSSION

JFP is an uncommon tumor, usually benign, that can cause important neurological deficits due to its location.\textsuperscript{[1,13]} Eventually, they can secrete norepinephrine that can cause peaks of refractory systemic arterial hypertension and arrhythmias, even intraoperatively.\textsuperscript{[1,11,12]} Therefore, secreting tumors should be preoperatively identified and they require preoperative beta-blockers.\textsuperscript{[24]} The treatment of these tumors has been controversial over the years. The involvement of the CN, blood vessels, temporal bone, and posterior fossa are difficulties imposed to its surgical treatment, which can result in neurological deficits and major complications, such as FN and LCN lesions, CSF fistulas, and infections.

More effective and safer resections have been achieved with the use of microsurgical techniques, intraoperative electrophysiological monitoring and better understanding of the temporal bone anatomy. The surgical technique used in this series was IFTA-D,\textsuperscript{[4]} with modifications. The opening was performed in two planes, the scalp and galea and the muscle/fascial, aiming to reduce time and facilitates closure. The mastoid cortical bone is removed and replaced during closure, becoming the reconstruction more anatomical, avoiding depression and pain in the mastoid region, and allowing the containment of the fat placed in the mastoid to eliminate dead space. Furthermore, the need for reconstruction with non-biological material is avoided. After these modifications we did not observe postoperative CSF leak, except for a bulging of the wound requiring lumbar external drainage. As the patient that had a CSF leak due to a dural tear and this patient that had a bulging of the wound had no intracranial invasion, care should be taken in all patients during the dissection of tumor adherent to the dura mater to avoid dural tear.

Fisch C/D class JFP resection is performed classically by infratemporal lateral approaches involving mastoidectomy and cervical exposure and eventually the posterior fossa. The approach to tumors that invade the tympanic cavity (ME and hypotympanum) through the lateral route has the FN and the ICA as obstacles. The transposition of the FN from its bone canal to the parotid region\textsuperscript{[13]} allows for ample access to the tympanic cavity and the petrous ICA, but it is followed by FN paralysis in most patients. Alternatively, the posterior and anterior approach to the FN (falloppian bridge) allows total resection of most of these tumors, preserving the FN in its canal and reducing postoperative deficits. Using this approach, we were able to perform total/subtotal resection even of large tumors invading the ME, hypotympanum, cervical region and eventually the posterior fossa, without deliberate NF transposition in any patient. When the FN was completely invaded by the tumor, it was sectioned, allowing ample access to the tympanic cavity. There was no difference between the extension of the resection and the access performed for each patient suggesting the adequacy of the indication. Therefore, we did not routinely indicate FN transposition for resection of Fish C/D class JFP, independent of its size, and extent. Therefore, we do not indicate routine FN transposition to Fisher C/D class JFP, regardless of its size and extension.

Preoperative angiography is essential to check tumor involvement and supply by ICA involvement and to check for the presence of collateral circulation. As these are hypervascularized lesions, preoperative intravascular\textsuperscript{[1,17,18,20,34]} or direct percutaneous\textsuperscript{[17]} embolization has been used. Intraoperative blood loss ranges from 0 to 8,000 ml\textsuperscript{[1,2,21,26,27]} for embolized and nonembolized tumors. Intraoperative blood loss was reported from 300 to 8000 ml (mean = 3057.8 ± 1934.6 ml) for all patients and 3922 ml versus 2716 ml for patients undergoing intraoperative ligation of ECA branches and undergoing preoperative embolization respectively (non-significant difference).\textsuperscript{[5]} Preoperative embolization of JFP reduces the intraoperative blood loss (mean = 1152.1 ± 828.8 ml in our patients), facilitating their surgical resection. On the other hand, embolization can compromise the blood supply of CNs irrigated by the same vessels that nourish the
tumor. The tympanic and mastoid portions of the FN are nourished by the stylomastoid artery (EA) and by middle meningeal artery (MMA) petrous branch. The EA originates from the occipital artery branch in 60% of cases and from the posterior auricular artery (PAA) in 40% of cases. However, 10% of patients have tympanic and mastoid portions of the FN nourished only through the EA. Thus, patients with single FN irrigation and patients undergoing multiple vessel embolization are more likely to suffer FN injury after embolization. Based on NF nourishment, the probability of post embolization injury was estimated at 6% and in the literature, it ranges from 0% to 14.2% (mean 5%). The NF lesions occurred in two (18.2%) of our patients occurred after partial and subtotal embolization, which suggests that post embolization NF lesions are more related to their vascularization than to the extension of embolization. After our present results, we indicate preoperative embolization only in large and highly vascularized tumors, especially if the patient already has FN deficit.

LCN irrigation comes from the neuromeningeal branch of the ascending posterior artery (APA) through its jugular branches in the stylomastoid foramen (glossopharyngeal, vagus, and accessory nerves branches) and hypoglossal canal (hypoglossal nerve branch). We did not observe post embolization LCN lesions.

The extension of resection and recurrence of JFP depend on the aggressiveness of the surgical technique and of the follow-up duration. In the first decade of this century, total resection varied from 51% to 92% and recurrence varied from 0% to 27.8%. In the last decade, they ranged from 61% to 100% (mean = 80.4 ± 11.7) and 0.6–31.2% (M = 17 ± 11), respectively, and the regrowth rate varied from 6.3% to 69%. Ten-year recurrence ranged from 13.2% to 28.2%, and the 15-year RFS was 79%. Total resection of JFP Fisch C/D using IFTA without transposition of the FN transposition is reported ranging from 63% to 91%. A literature review, showed better performance of IFTA with transposition of the NF versus non transposition in relation to the extension of JFP resection (84 ± 9.0% vs. 76 ± 10.1%), but with a lower FN preservation rate. In our series, total resection was similar to reported in the literature using the IFTA without FN transposition (total radiological resection was 50%, with 25% recurrence, and 23.1% progression during follow-up) (55.6 ± 40.6 months).

We did not observe any difference between GS and RFS in relation to total and subtotal resections. Macroscopic intraoperative assessment of the extension of resection of JFP is not reliable because of bone invasion. The independent radiological evaluation showed a 20% decrease in the total resection rate.

Postoperative evolution of CNs

Signs of CN compression and irritation in patients with JFP Fisch C/D usually improve after surgery, but deficits do not always improve and may worsen. We observed improvement in hemifacial pain and motor deficit of the V CN and paresis of the VI CN after cavernous sinus decompression; nevertheless, hypoesthesia/hemifacial dysesthesia remained unchanged.

Tinnitus and hearing loss in patients with Fisch C/D class JFP, improved postoperatively in 37.3% and 6.6–29.4%, respectively, and worsening occurs in 2% and 0–50%, respectively. In our patients, improvement in tinnitus occurred in 88% and worsening/new hearing loss occurred in 20% of patients. No patient with hearing loss improved.

New postoperative FN deficits occur in 5.8-55.4% for JFP and preoperative deficits improve by 9.8%. Patients undergoing IFTA with a retrofacial approach or with a fallopian bridge have better preservation of the FN (HB I/II) than patients undergoing short and long transposition of the FN (mean of 90, 92.2, and 66 months, respectively). We observed similar results (76.9% of patients with FN HB I/II function compared to the preoperative period). Patients undergoing FN grafts show recovery of HB III in 42–100%, and the remainder have HB IV and V. This was also observed among our patients. Intraoperative monitoring of the FN is an important help in nerve function preservation during preparation of the fallopian bridge and in the dissection of tumor involving or invading the nerve.

LCN injuries occur in 0–67% of patients with JFP treated surgically. Although all of our patients with LCN injuries showed a satisfactory degree of functional improvement, all persisted with the related neurological signs, indicating a compensation for function by the opposite side nerve. Adaptation was faster and better in patients with preoperative evolutionary injuries than in patients with acute postoperative injury. We do not use routinely preoperative tracheostomy or gastrostomy; our current policy is to perform swallowing tests before starting any diet and to perform an early tracheostomy whenever there is breathing difficulty and a risk of aspiration. Deficits due to XI and XII CN injuries also did not change in the postoperative neurological examination. Management of patients with lesions of LCN by a multidisciplinary team can avoid severe complications and improve the results of surgical treatment of JFP.

The operative mortality of patients with JFP operated is relatively low (0–2.6%). Our operative mortality was slightly higher (3.8%).
Radiotherapy

Recent systematic reviews of the literature show good tumor control in the primary treatment of jugulo-tympanic paragangliomas with stereotactic radiosurgery (STR) and Gamma-knife and good effectiveness and fewer complications when compared to adjunctive surgery or isolated microsurgical treatment. However, several factors, such as inclusion of most retrospective series, lack of randomized studies, and absence of comparisons with the natural evolution of the disease, do not consider the tumor size and the relatively short follow-ups need to be highlighted in these studies, making it difficult to generalize results that are considered to be overestimated. More recently, we have STR and IMRT available at our hospital, and we recommend STR only as a rescue treatment for patients with no indication for new surgery.

This study exhibits and presents the limitations that are inherent of a retrospective study, which that must be considered in its interpretation of the results. In addition, despite the evidence in the literature about the better results of postoperative FN function without transposition, we have no own series of patient with FN transposition to compare.

CONCLUSION

The treatment of patients with JFP Fisch C/D, although still with morbidity, has good long-term results. Surgical techniques without the transposition of the FN cause fewer intraoperative lesions; however, it also causes lower rates of total resection and higher recurrence. Preoperative embolization of JFP reduces the intraoperative blood loss but can cause FN deficit. Our results indicate that JFP can be treated with good results in general hospitals using parsimonious preoperative embolization and the posterior and anterior approach to the FN (fallopian bridge) approach, if a multidisciplinary team is available. Rescue SRS in cases of recurrence should be considered in each case, depending on the characteristics of patients and tumors.

Declaration of patient consent

Institutional Review Board (IRB) permission obtained for the study.

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Conflicts of interest

There are no conflicts of interest.

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