Effect of Surgical Resection on Jugular Foramen Tumors: A Retrospective Study

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Abstract

The complex anatomical structure of jugular foramen (JF) makes a challenge for the diagnose and treatment of this region's tumors. The purpose of the study was to investigate the effect of surgical resection in patients with JF tumors. A total of 77 patients with JF tumors treated with surgical resection between 2012 and 2021 were retrospectively reviewed. General characteristics, tumor classification, surgical approach and outcomes were retrospectively analyzed. The mean follow-up period was 4.5 years (ranging 0.5-8.5 years). Of the enrolled patients, 63 patients (81.82%) were diagnosed with JF schwannomas. 67 cases (87.01%) of the total patients received gross-total resection. Preoperatively, hoarseness and/or dysarthria served as the most complained symptoms (n=41, 53.25%), followed by swallowing disturbance (n = 37, 48.05%), hearing disturbance and/or tinnitus (n = 32, 41.56%). Postoperatively, a total of 52 patients (67.53%) were without significant symptoms. Worsened hoarseness and/or dysarthria symptoms were found in 14 patients (18.18%). Besides, 6 cases (7.79%) of the enrolled patients showed worsened swallowing disturbance. 8 patients (10.39%) presented with facial palsy after operation and 3 of them got improvement during the follow-up. Nine patients (12.68%) had tumor recurrence and there was one death (1.3%) associated with surgery during the perioperative period in the cohort. Therefore, surgical resection was an effective treatment of JF tumors with improvements of symptoms and good control of tumor. However, operation on this region was at the risks of worsening cranial nerve deficits and death, which needed advance in skull base surgery and rich experience.

Introduction

Jugular foramen (JF), located in the posterior cranial fossa, has a complex anatomy structure. This foramen contains important neurovascular structures (e.g., the jugular bulb, sigmoid sinus and lower cranial nerves (CNs)), which makes a challenge for the diagnose and treatment of the JF tumors. Jugular paragangliomas serve the most frequent lesions involving the JF, followed by jugular foramen schwannomas (JFSs) and meningiomas [9]. Other lesions include chordomas, squamous cell carcinomas and temporal bone malignancies [7]. Arising from CNs IX, X, and XI, JFSs represent about 2.9–4% of all intracranial schwannomas [8, 17]. Patients with JF tumors may complain about hearing loss, pulsatile tinnitus and CNs deficits symptoms (including facial palsy/spasm, hoarseness and/or dysarthria, swallowing disturbance, shoulder weakness and shoulder weakness). However, operation on the JF region makes the CNs and vessels at high risk. It was reported that 5.7% patients (3/53) with JFSs had a recurrence of tumor postoperatively [3]. Besides, previous study reported that 3.9% patients (4/102) died after the surgery of removing JF tumors [9]. To achieve total resection without injuring the CNs and vessels, advances in skull base surgery, neurophysiological monitoring technology and rich experience are warranted.

The present study reports the surgical outcomes of 77 patients with JF tumors in our center. All the patients were operated by the senior authors between May 2012 and June 2021. Baseline characteristics, tumor classification, surgical approach, surgical complications and follow-up findings are presented.

Material And Methods

Patients

Altogether, 77 patients with JF tumors treated with surgical resection between May 2012 and June 2021 were retrospectively enrolled. These patients were operated by the senior authors (NZ and ZH) at the First Affiliated Hospital of Sun Yat-sen University, (Guangzhou, Guangdong, China). The collected data (including baseline characteristics, tumor classification, surgical approach, postoperative complications and follow-up findings) were analyzed. Preoperatively, hearing tests, neurological examinations, laryngoscopy, magnetic resonance imaging (MRI) and computed tomography (CT) were performed. Digital subtraction angiography (DSA) was performed in three patients with glomus jugulare tumor and two of them underwent vascular embolism operation before the tumor removal. Lumbar drainage was performed to the total patients preoperatively. Intraoperative monitoring included motor evoked potentials, somatosensory evoked potentials, CNs monitoring (VII, IX, X, XI and XII nerve) and brainstem auditory evoked potentials. A CT scan was applied on the first postoperative day. Follow-up MRI images were performed at 3 months, one year after operation and annually thereafter. Based on the intraoperative findings, postoperative first-day CT scan and MRI performed 3 months postoperatively, resection degree was determined. Average follow-up period in the

present study was 4.5 years (ranging 0.5-8.5 years) and 5 patients lost follow-up. This study was approved by the Ethics Committee of the First Affiliated Hospital of Sun Yat-Sen University.

Tumor classification and surgical approach

Based on the lesion locations, tumors involving the JF were classified into four types (type A, B, C, and D), as proposed by Samii, et al (Table 1, Fig. 1) [11]. The most frequent type of JF tumor in this study was type D (40 cases, 51.95%), followed by type A (27 cases, 35.06%), type B (7 cases, 9.09%) and type C (3 cases, 3.9%). We used the transjugular approach and endoscope assisted–retrosigmoid approach (EA-RS approach) to remove the JF tumors (Table 1), which were described detailly in previous literatures [2, 5, 10, 16]. In total, sixty-seven patients (87.01%) underwent the transjugular approach for the tumor removal. Ten JF lesions (12.99%) were approached via an EA-RS approach. To reconstruct the cranial base, a technique with three-layers vascularized flaps (first layer: dura mater (or temporalis fascia and fibrin glue); second layer: temporalis fascia; third layer: cervical fascia, sternocleidomastoid muscle and temporalis muscle) indicated by Ramina et al [9] was used.

Table 1
Tumor classification and surgical approach of the patients*

Tumor classification	Tumor location	Surgical approach		Total (%)
		Transjugular approach	EA-RS approach§	
A	Primarily intracranial with minimal enlargement of JF [#]	19	8	27 (35.06)
B	Primarily JF with intracranial extension	7	0	7 (9.09)
C	Primary extracranial with foraminal extension	3	0	3 (3.9)
D	Intra-/extracranial extension (dumbbell shape)	38	2	40 (51.95)
*Proposed by Samii, et al; [#] JF: Jugular Foramen; §EA-RS approach: Endoscope assisted–retrosigmoid approach.				

Results

Baseline characteristics

Altogether seventy-seven patients with JF tumors were included in the present study. Baseline characteristics of these patients are listed in Table 2. Based on the clinical characteristics and histopathology, lesions involving the JF on the enrolled patients were included: schwannomas (63 cases), meningiomas (3 cases), chordomas (3 cases), glomus jugulare tumor (3 cases), low-grade malignant tumor of unknown origin (1 case), endolymphatic sac tumor (1 case), malignant solitary fibrous tumor (1 case), pleomorphic parotid adenoma (1 case) and melanoma (1 case). Of these patients, there were 5 patients (6.49%) who had history of surgical resection due to the JF tumors. Mean age of the enrolled patients was 40 years, ranging from 8-72 years. Of the total patients, female and male patients accounted for 42 cases (54.55%) and 35 cases (45.45%), respectively. The clinical presentations of the patients in the study included symptoms of CNs deficits (including VII, VIII, IX, X, XI and XII nerve), gait disturbance, headache and/or dizziness. Hoarseness and/or dysarthria served as the most frequent symptoms, shown in 41 cases of the total patients (53.25%). The following symptoms were swallowing disturbance (n = 37, 48.05%), hearing disturbance and/or tinnitus (n = 32, 41.56%), headache and/or dizziness (n = 11,14.29%). Furthermore, there were five (6.49%) and four (5.19%) patients presented with gait disturbance and hypoglossal nerve palsy, respectively. Facial palsy/spasm and shoulder weakness were respectively found in 3 cases (3.9%) and 2 cases (2.6%) of total patients.

Table 2
Baseline characteristics of 77 patients with jugular foramen
tumors who received surgical resection

Characteristics	Value
Age (years)	
Mean	40
Range	8-72
Gender	
Female	42 (54.55%)
Male	35 (45.45%)
History of surgical resection	
Yes	5 (6.49%)
No	72 (93.51%)
Preoperative KPS* score	
100	21 (27.27%)
90	43 (55.84%)
80	12 (15.58%)
70	1 (1.3%)
Pathological type	
Schwannomas	63 (81.82%)
Meningiomas	3 (3.9%)
Chordomas	3 (3.9%)
Glomus jugulare tumor	3 (3.9%)
Low-grade malignant tumor of unknown origin	1 (1.3%)
Endolymphatic sac tumor	1 (1.3%)
Malignant solitary fibrous tumor	1 (1.3%)
Pleomorphic parotid adenoma	1 (1.3%)
Melanoma	1 (1.3%)
Clinical symptoms	
Hearing disturbance and/or tinnitus	32 (41.56%)
Facial palsy/spasm	3 (3.9%)
Swallowing disturbance	37 (48.05%)
Hoarseness and/or dysarthria	41 (53.25%)
Shoulder weakness	2 (2.65%)
Hypoglossal nerve palsy	4 (5.19%)
Gait disturbance	5 (6.49%)
* KPS: Karnofsky Performance Status.	

Characteristics	Value
Headache and/or dizziness	11 (14.29%)
* KPS: Karnofsky Performance Status.	

Surgical Outcomes

Postoperatively, 67 cases (87.01%) of the total patients received gross-total resection and 10 patients (12.99%) remained residual (Table 3). The mean follow-up in the present study was 4.5 years (ranging 0.5-8.5 years) and there were 5 patients losing the follow-up. There were no mortalities during the follow-up. In total, 9 patients (12.68%) had tumor recurrence with a mean period of 2.17 years (including 4 cases of schwannomas, 2 cases of chordomas; one case of endolymphatic sac tumor, one malignant solitary fibrous tumor and one meningioma; Table 4). 62 patients (87.32%) were without tumor relapse. 14 of the 71 patients (19.72%) had 100 scores of KPS and 51 patients (71.83%) were with 90 scores. There were five patients (7.04) whose KPS scores were 80. However, one patient complained with a 60 score of KPS. Altogether, there were 52 patients (67.53%) who didn't show significant complains postoperatively. 14 patients (18.18%) presented with new/worsened hoarseness and/or dysarthria symptom. Facial palsy was found in 8 patients (10.39%) after operation and 3 of these 8 patients got improvement during the follow-up. 6 cases (7.79%) of the patients presented with new/worsened swallowing disturbance. Symptoms of preoperative shoulder weakness and hypoglossal nerve palsy didn't get worse after surgery. Cerebrospinal fluid leakage was shown in 9 patients (11.69%) and 4 of them were received further surgical management to treat the leakage. Lumbar drainage was applied to treat the rests 5 patients conservatively. Besides, there were one case (1.3%) of intracranial infection and one case (1.3%) of pneumonia after operation. Nine patients (11.69%) accepted tracheotomy postoperatively and eight of them removed the tracheal cannula successfully, except one patient losing the follow-up. Cerebral infarction occurred in one patient postoperatively due to internal carotid artery thrombosis. One patient (1.3%) with type D JFSs died for the postoperative cerebellar swelling and hemorrhage during the perioperative period.

Table 3
Surgical outcomes in patients with jugular foramen tumors

Outcomes	No. of patients (%)
Postoperative KPS* score	
100	14 (19.72)
90	51 (71.83)
80	5 (7.04)
70	0
60	1(1.41)
Resection degree	
Gross-total resection	67 (87.01)
Residual	10 (12.99)
Tumor recurrence	
Yes	9 (12.68)
No	62 (87.32)
Post-complications	
None	52 (67.53)
New/worsened swallowing disturbance	6 (7.79)
New/worsened hoarseness and/or dysarthria	14 (18.18)
New/worsened shoulder weakness	0
New/worsened hypoglossal nerve palsy	0
Facial palsy	8 (10.39)
Cerebrospinal fluid leakage	9 (11.69)
Intracranial infection	1 (1.3)
Pneumonia	1(1.3)
Tracheotomy	9 (11.69)
Cerebral infarction	1(1.3)
Dead	1(1.3)
* KPS: Karnofsky Performance Status.	

Table 4
Baseline characteristics of 9 patients with tumor recurrence

Patients	Pathology	Side	Classification	Preoperative symptoms	Resection	approach	Follow-up period	Post-complications
1	Recurrent Chordomas	Left	D	Facial palsy; swallowing disturbance; Hoarseness	Residual	Transjugular approach	2 years	Facial palsy; improvement of swallowing disturbance and hoarseness
2	Recurrent schwannomas	Left	D	Hearing disturbance and headache	Residual	EA-RS approach	3 years	Hearing disturbance
3	Endolymphatic sac tumor	Right	B	Hearing disturbance and tinnitus	Residual	Transjugular approach	1 year	Tinnitus; hearing disturbance and new facial palsy
4	Malignant solitary fibrous tumor	Left	D	Hoarseness; swallowing disturbance and hypoglossal nerve palsy	GTR*	Transjugular approach	1 year	Stable hoarseness, swallowing disturbance, hypoglossal nerve palsy and CSF leakage [#]
5	Schwannomas	Left	C	Headache; dizziness and tinnitus	GTR	Transjugular approach	4 years	New hoarseness and swallowing disturbance
6	Schwannomas	Left	B	Headache	GTR	Transjugular approach	3 years	None
7	Schwannomas	Right	D	Hearing disturbance; facial spasm and hoarseness	Residual	Transjugular approach	2 years	Tracheotomy; CSF leakage; facial palsy; worsen hoarseness and swallowing disturbance
8	Chordomas	Left	A	Hearing disturbance and hoarseness	GTR	EA-RS approach	1.5 years	Improvement of hoarseness
9	Meningiomas	Right	D	Headache and dizziness	GTR	Transjugular approach	2 years	CSF leakage; Cerebral infarction; new hoarseness and swallowing disturbance
*GTR: Gross-total resection; [#] CSF leakage: Cerebrospinal fluid leakage.								

Illustrative cases

1. Patient with JFSs

This was a 68-years old women with a type D JFSs (Fig. 2). Chief complains were right tinnitus and dizziness for one year. Preoperative MRI showed a right dumbbell-shaped jugular foramen tumor with a size of 22 mm x 38 mm x 49 mm (Fig. 2A, 2B); Postoperatively, this patient presented pneumonia and newly developed hoarseness. MRI of 3 months after operation demonstrated that the tumor was under gross-total resection (Fig. 2C); MRI images of 1 year after operation showed the tumor was without recurrence (Fig. 2D).

2. Patient with low-grade malignant tumor of unknown origin

This was a 21-years old man with low-grade malignant tumor of unknown origin involving the JF (Fig. 3). Chief complains were right tinnitus for one year, hearing disturbance and pain in the right ear for 3 months. Preoperative MRI showed a right dumbbell-shaped JF tumor with a size of 34 mm x 40 mm x 47 mm (Fig. 3A, 3B); The patient presented CSF leakage, newly developed hoarseness and swallowing disturbance postoperatively. MRI of 3 months after operation showed the tumor had residual (Fig. 3C); MRI of 1 year after operation showed the residue tumor didn't enlarge significantly (Fig. 3D).

3. Patient with malignant solitary fibrous tumor

This was a 57-years old man with JF malignant solitary fibrous tumor, WHO III (Fig. 4). Chief complains were hypoglossal nerve palsy for one year, hoarseness for 20 days and swallowing disturbance for 10 days. Preoperative MRI showed a right dumbbell-shaped JF tumor with a size of 35 mm x 95 mm (Fig. 4A, 4B); Postoperatively, the patient presented CSF leakage and stable symptoms of hypoglossal nerve palsy, hoarseness and swallowing disturbance. MRI of 3 months after operation showed the tumor was removed totally (Fig. 4C); MRI of 1 year after operation showed tumor had a recurrence (Fig. 4D).

Discussion

It is well known to us that the first-line treatment for JF tumors is surgical resection. Due to the complex anatomical structure of JF, the optimal result of the surgery in the JF tumors is to remove the tumor completely without any cranial nerve and vessel injuries. With advances in intraoperative monitoring, surgical techniques and rich experience of skull base surgery, these tumors could be removed safely. A previous study enrolled 53 patients with JFSs indicated that vague nerve deficit served the most common presentation, followed by VIII nerve and IX nerve deficits. Besides, 5.7% of the total cases experienced tumor recurrence postoperatively [3]. Sedney et al. manifested that hearing loss was the most common complaint in patients with JFSs (51/81, 63%), followed by vagal nerve deficit (46/81, 56.8%) and glossopharyngeal nerve deficit (30/81, 37%) [15]. In the present study, hoarseness and/or dysarthria were the most common symptoms preoperatively (41/77, 53.25%), followed by swallowing disturbance (37/77, 48.05%), hearing disturbance and/or tinnitus (32/77, 41.56%).

The surgical approach for the tumor removal is based on the surgeon's experience, location and extension of the tumor. It was reported that the infratemporal fossa type A (IFT-A) approach was an optimal approach for removing the JF vascular tumors [7, 14]. Petro-occipital transsigmoid (POTS) approach was applied to remove the intra- and extracranial extension lesions effectively, offering well exposure of the JF region [12, 13]. Besides, retromastoid approach remained an optical approach for intracranial JF lesions without osseous invasion. Extreme lateral infrajugular transtubercular (ELITE) approach could be used to the intracranial JF lesions with osseous invasion or dumbbell-shaped JF lesions. An intradural and transjugular approach was used to remove the dumbbell-shaped JF lesions with involving the internal jugular vein and jugular bulb [1, 3]. Another previous study performed the surgery on Samii classification [11] type A and B tumors through suboccipital approach, and used transjugular approach for type C and D tumors [16]. In our center, most patients with JF tumors underwent the transjugular approach for the tumor removal except ten cases (including 8 type A tumors and 2 type D tumors) via an EA-RS approach. Among the nine patients with recurrent tumor, two of them were treated with EA-RS approach and the rest were via a transjugular approach. Through these two approaches and the vascularized flaps indicated by Ramina et al [9], only 9 patients (11.69%) occurred with postoperative CSF leakage in the present study. Furthermore, just 10 cases (12.99%) remained residual.

Recently, increasing studies have showed that radiosurgery could effectively control the growth of residual or recurrent JFSs. A multiinstitutional study reviewed the outcomes of 117 JFSs patients underwent Gamma Knife surgery and showed that 53% of the patients had partial remission and 36% had stable tumors. Tumor progression was found in 11% of the patients. The 3-year

progression-free survival (PFS) rates was 91% and the 5-year' was 89% [4]. Another retrospective study indicated that 51% of the 92 enrolled patients underwent Stereotactic radiosurgery had a regressed tumor and 13% presented with progressed tumor. Besides, dumbbell-shaped JFSs were at higher risk of progression [6]. In the present study, a total of 77 patients with JF tumors experienced surgical resection in our center. The most frequent type of JF tumor was type D (51.95%), followed by type A (35.06%), type B (9.09%) and type C (3.9%). Postoperatively, 67.53% cases were without significant symptoms postoperatively. Except one patient (1/77, 1.3%) with type D JFSs dying for the surgery, there were no mortalities during the follow-up. Nine patients (12.68%) occurred with tumor recurrence. Therefore, surgical resection was an effective treatment of JF tumors with improvements of symptoms and good control of tumor. For patients with residual/recurrent tumors, postoperative radiosurgery serves as an optical treatment for the long-term control of tumor.

Limitations

Many limitations existed in this study. First of all, this is a single-center study and the sample size is limited, which is necessary to enlarge to decrease the bias. Besides, our study is a retrospective study and bias may exist during the follow-up. What's more, due to the low incidence of tumor recurrence rate, the factors affecting the tumor recurrence hasn't been analyzed. Lastly, this study didn't compare the difference between the efficacy of radiosurgery and surgical resection.

Conclusion

A total of 77 patients with JF tumors treated with surgical resection in our center were retrospectively reviewed. Our results manifested that surgical resection was an effective treatment of JF tumors with improvements of symptoms and good control of tumor. However, operation on the JF region was at the risks of CNs deficits and death, which needed advance in skull base surgery and rich experience.

Declarations

Author contribution Conception and design: WS, XL and LY. Acquisition of data: HT and ZD and HW. Follow-up: WS and HW. Composing the article: WS and XL. Approved the final version of the manuscript for publication on behalf of all authors: ZH. Study supervision: NZ and ZH.

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Data availability The data of this study are available on request due to local (academic) restrictions.

Code availability Not applicable.

Ethics approval This study is approved by the Ethics Committee for Clinical Research and Animal Trials of the First Affiliated Hospital of Sun Yat-Sen University (Guangzhou, China).

Consent to participate Not applicable.

Consent to publication All authors are consent to publish in Neurosurgical Review.

Conflicts of interest The authors declare that they have no conflict of interest.

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Figures

Figure 1

Jugular foramen tumors were classified into 4 types based on the tumor location (red arrows). A: Type A: primarily intracranial with minimal enlargement of JF. B: Type B: primarily JF lesions with intracranial extension. C: Type C: a primarily extracranial tumor with extension into the JF. D: Type D: a dumbbell-shaped tumor with both intra- and extracranial extension.

Figure 2

68-years old women with a type D JFSs. A&B: preoperative MRI showed a right dumbbell-shaped jugular foramen tumor; C: MRI of 3 months after operation demonstrating gross-total resection; D: MRI of 1 year after operation showed the tumor was without recurrence.

Figure 3

21-years old man with type D JF low-grade malignant tumor of unknown origin. A, B: preoperative MRI showed a right dumbbell-shaped jugular foramen tumor; C: MRI of 3 months after operation showed the tumor had residual (red arrow); D: MRI of 1 year after operation showed the residue tumor didn't enlarge significantly (red arrow).

Figure 4

57-years old man with malignant solitary fibrous tumor involving JF, WHO III. A, B: preoperative MRI showed a left dumbbell-shaped lesion involving jugular foramen; C: MRI of 3 months after operation showed the tumor was removed totally; D: MRI of 1 year after operation showed tumor had a recurrence (white arrow).