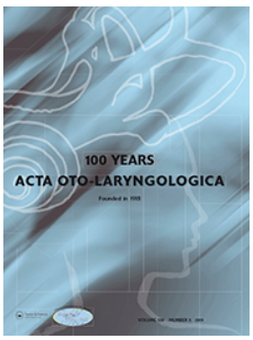

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RESEARCH ARTICLE

Intraparotid facial nerve schwannoma: a 17-year, single-institution experience of diagnosis and management

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ABSTRACT

Background: Intraparotid facial nerve schwannoma (IFNS) is rare and its definite preoperative diagnosis is challenging.

Objective: To improve available knowledge regarding the diagnosis of IFNS and to suggest an appropriate treatment plan.

Material and methods: We retrospectively analyzed medical records of IFNS patients at our hospital. Inclusion criteria were surgery (from January 2000, to December 2016) for a parotid mass, pathologically diagnosed as a schwannoma.

Results: The study included 42 eligible patients who had undergone tumor resection from 5977 parotid tumor patients. Mostly presented hard-textured (18/39) or medium-textured (15/39), with limited mobility (21/39) mass (three tumors were not palpable). Their facial nerve function outcomes were House–Brackmann Grade I ($n = 14$), Grade II ($n = 7$), Grade III ($n = 11$), Grade IV ($n = 5$), Grade V ($n = 3$), and Grade VI ($n = 2$). Significant differences were noted in results based on different surgical methods used ($p = .000$) and tumor involvement ($p = .002$).

Conclusions and significance: A hard-textured tumor with limited mobility mass in the parotid gland should prompt the diagnosis of a schwannoma. Tumors involving main trunk usually lead to unsatisfactory facial nerve outcomes. Facial nerve preservation should always be essential, and stripping surgery or intracapsular enucleation could be the preferred surgical methods of choice.

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KEYWORDS
schwannoma; parotid gland; facial paralysis; reconstructive neurosurgery; nerve sparing surgery

Introduction

Schwannomas, first introduced as 'neurinomas' by Verocay [1] in 1910, are benign tumors originate from Schwann cells, which may grow from any peripheral, cranial, or autonomic nerve. The first complete case of intraparotid facial nerve schwannoma (IFNS) was reported by O'Keefe [2] in 1949. Approximately 9–23% of all facial nerve schwannomas are located extratemporal [3].

The most common complaint of patients with IFNS is a chronic, mostly single, painless parotid mass. Only certain patients experience preoperative hemifacial paralysis [4]. Imaging characteristics are usually nonspecific, and the detection rate with fine-needle aspiration cytology (FNAC) is low. Diagnosis of IFNS is often confirmed when a surgeon discovers the facial nerve course entering the tumor or when the facial nerve cannot be located during a parotidectomy, or after frozen pathological section [5].

The resection of IFNS may cause severe nerve damage. Surgical management of facial nerve schwannoma varies from total resection and reconstruction, stripping surgery, intracapsular enucleation and microenucleation, debulking surgery, and radiosurgery to simple biopsy and conservation [6]. Although several treatment strategies have been proposed [5,7], the relatively low incidence of IFNS makes it difficult to determine the best treatment algorithm, and perform comparisons among different surgical procedures.

This article presents our experience on treating IFNS, with an aim to seek out diagnosis methods, evaluate facial nerve outcomes with various surgical procedures, and contribute to one of the largest IFNS case series to our knowledge.

Methods

We conducted a retrospective chart review of all patients who had undergone resection of parotid region tumor between January 2000, and December 2016, at our hospital. Inclusion criteria were surgical treatment of an intraparotid tumor and a confirmed diagnosis of schwannoma through pathological examination; exclusion criteria were patients in whom tumor seemed to originate in the parapharyngeal space, had another tumor history in the same region, and

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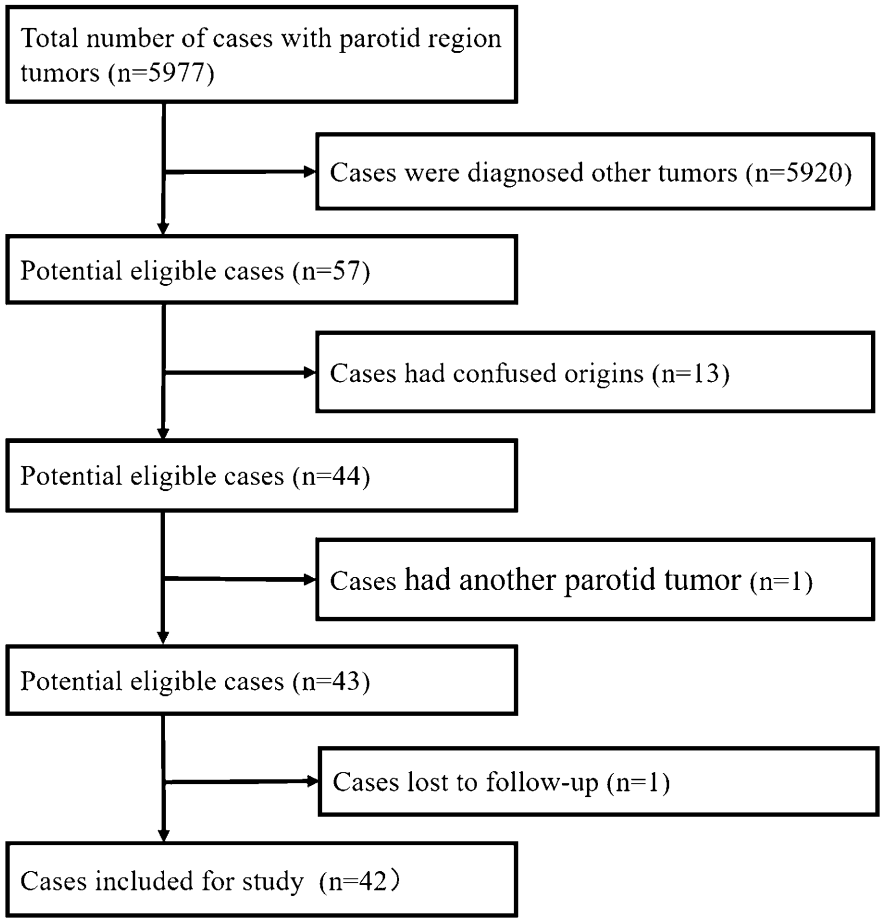

Figure 1. Flow diagram of included and excluded cases.

Table 1. Clinical characteristics and management of patients.

Characteristics	Number
Sex (M/F)	16/26
Side (L/R)	27/15
Median age (years)	44.5 (13–71)
Median duration (months)	8.5 (0.1–240)
Preoperative facial nerve function	
Paralyzed	3
Normal	39
Preoperative diagnosis by	
Surgery history	10
FNAC	2
CT scan	2
False diagnosed	28
Tumor management	
Stripping	17
Intracapsular enucleation	9
Resection with reconstruction	13
Resection without reconstruction	3
Tumor location	
Main trunk or divisionsa	30
Main branches or distal branches	12
Facial nerve management	
Spared	26
Sacrificed	3
Reconstructed	13
Anastomosis	9
Nerve graft	4
Mean size (cm)	2.69 ± 1.14
Mean follow-up (years)	3.37 ± 2.69
Recurrence	1/42

[a]Main division represents the temporo-facial and the cervico-facial division.

patients with incomplete records or lost to follow-up. The study protocol was approved by the ethics committee of the institution. All surgeries were performed by surgeons with relevant experience in the respective fields.

Analyzed data included patients' sex and age, duration of tumor, side affected, texture, mobility, size, involved branch, surgical approach used, facial nerve function before and after surgery, follow-up period, and tumor recurrence. Facial nerve function was classified according to the House–Brackmann (HB) grading system. Most cases were evaluated using enhanced computed tomography (CT) or magnetic resonance imaging (MRI).

Surgical procedures

A stripping surgery required total exposure of the tumor, including the inferior portion. The key point was locating normal facial nerve fascicles under the epineurium around the tumor. Intraoperative nerve monitoring or application of electroneurography could be helpful in searching nerve fascicles. A tumor could be cautiously dissected when the boundary of the nerve and the tumor was verified, so that the nerve could be preserved. Intracapsular enucleation required a longitudinal incision on the capsule in order to avoid conflicting with facial nerve fascicles till the tumor was exposed, and then gently separating the capsule with

Table 2. Comparison between postoperative facial nerve HB grades across different patient characteristics and management technique used.

	Postoperative facial nerve HB grades (%)							
Characteristic	I	II	III	IV	V	VI	Total	*p*
Texturea								.583
Soft	4 (66.7)	0 (0)	1 (16.7)	0 (0)	0 (0)	1 (16.7)	6 (100)	
Medium	5 (33.3)	2 (13.3)	3 (20.0)	3 (20.0)	1 (6.7)	1 (6.7)	15 (100)	
Hard	4 (22.2)	4 (22.2)	6 (33.3)	2 (11.1)	2 (11.1)	0 (0)	18 (100)	
Mobilityb								.109
Good	9 (50.0)	2 (11.1)	3 (16.7)	2 (11.1)	0 (0)	2 (11.1)	18 (100)	
Limited	4 (19.0)	4 (19.0)	7 (33.3)	3 (14.3)	3 (14.3)	0 (0)	21 (100)	
Diagnosis								.053
Schwannoma	1 (7.1)	4 (28.6)	5 (35.7)	1 (7.1)	2 (14.3)	1(7.1)	14 (100)	
Parotid gland tumor	13 (46.4)	3 (10.7)	6 (21.4)	4 (14.3)	1 (3.6)	1 (3.6)	28 (100)	
Tumor involvement								.002
Main trunk or divisionsb	4 (13.3)	6 (20.0)	10 (33.3)	5 (16.7)	3 (10.0)	2 (6.7)	30 (100)	
Main branches or distal branches	10 (83.3)	1 (8.3)	1(8.3)	0 (0)	0 (0)	0 (0)	12 (100)	
Surgical approach								.000
Stripping	13 (76.5)	2 (11.8)	2 (11.8)	0 (0)	0 (0)	0 (0)	17 (100)	
Intracapsular enucleation	1 (11.1)	3 (33.3)	2 (22.2)	1 (11.1)	2 (22.2)	0 (0)	9 (100)	
Resection with reconstruction	0 (0)	2 (15.4)	7 (53.8)	4 (30.8)	0 (0)	0 (0)	13 (100)	
Resection without reconstruction	0 (0)	0 (0)	0 (0)	0 (0)	1 (33.3)	2 (66.7)	3 (100)	
Total	14 (33.3)	7 (16.7)	11 (26.2)	5 (11.9)	3 (7.1)	2 (2.8)	42 (100)	

[a]In 2 patients, tumors were discovered by imaging but were not palpable, and in 1 patient, a residual tumor was found intraoperatively inside the stylomastoid foramen; thus, overall, data recorded from 39 patients be analyzed.
[b]Main division represents the temporo-facial and the cervico-facial division.

the tumor; once the tumor was mobilized, it could usually be removed *en bloc* with the nerve been spared. Proximal and distal portions of the tumor must be meticulously inspected before removal, for any nerve fascicles possibly entering and exiting the tumor. If such fascicles exist, a piecemeal resection might be helpful. Microsurgical instruments, particularly microscopes are also required for manipulation and identification of the facial nerve.

Chi-square test, Student's *t* test, and Fisher exact test were used to test the significance of any differences identified as well as to evaluate and correlations. Calculations were performed using the IBM Statistical Package for the Social Sciences Statistics version 20. A p value $<.05$ was considered statistically significant.

Results

In total, 5977 parotid region tumors were resected during the study period; of these, 5920 were not schwannomas, 13 had confused origins (from the parapharyngeal space or subcutaneous tissues), 1 had another parotid tumor history, and 1 patient lost to follow-up. Overall, 42 patients were eligible for the study (Figure 1).

Patient characteristics

The study consists of 16 men and 26 women, their clinical characteristics were listed (Table 1). The characteristic information of each patient were listed in the Supplementary Table. There were 39 patients who presented a parotid mass, whereas 2 tumors were accidentally detected on MRI scans without any clinical appearance, and in 1 patient, a residual tumor was identified in the stylomastoid foramen during the secondary surgery that was planned for facial nerve reconstruction. Furthermore, three patients suffered preoperative facial nerve paralysis spontaneously, and eight cases appeared facial paralysis after previous surgeries. In addition, 10 patients had already undergone surgeries at other hospitals; of these, 2 had undergone tumor resections (i.e. these were cases of recurrences), 6 had debulking surgeries or biopsy, and 2 patients had simply terminated their surgeries.

Fourteen of the 42 tumor group patients had a preoperative diagnosis of schwannoma: 10 by previous surgical experience, 2 by FNAC, and 2 by CT scans showing enlargement of the stylomastoid foramen. The remaining 25 patients were diagnosed as parotid tumors.

Facial nerve function

The preoperative facial nerve function was HB Grade I ($n=39$), Grade II ($n=1$), and Grade V ($n=2$). Facial nerve function outcomes at follow-up were HB Grade I ($n=14$), Grade II ($n=7$), Grade III ($n=11$), Grade IV ($n=5$), Grade V ($n=3$), and Grade VI ($n=2$). Seventeen patients received stripping surgery and recovered to HB Grade I ($n=13$), Grade II ($n=2$), or Grade III ($n=2$). Intracapsular enucleation was performed in nine patients, who recovered to Grade I ($n=1$), Grade II ($n=3$), Grade III ($n=2$), Grade IV ($n=1$), or Grade V ($n=2$). 13 patients underwent facial nerve reconstruction immediately after tumor resection, including nerve anastomosis ($n=8$) and great auricular nerve graft ($n=5$), and recovered to Grade II ($n=2$), Grade III ($n=7$), or Grade IV ($n=4$). Furthermore, facial nerve was sacrificed without repair in 3 patients, and their postoperative facial function were Grade V ($n=1$) and Grade VI ($n=2$).

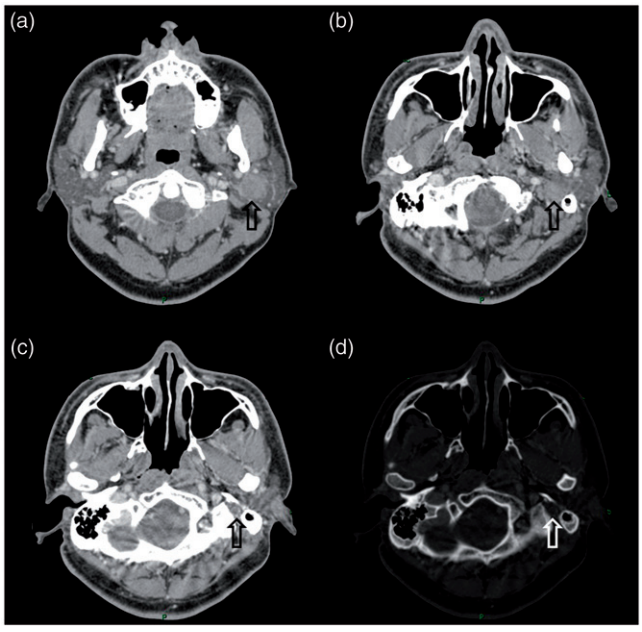

Figure 2. Enhanced CT scan of a patient. Black arrows in (a), (b), and (c) indicate a left-sided parotid schwannoma extending toward the stylomastoid foramen and involved in it. White arrow in (d) shows the widened stylomastoid foramen in the bony window.

Of note, facial nerve outcomes did not differ significantly based on mobility, texture, preoperative paralysis, or accuracy of diagnosis. However, significant differences ($p < .05$) were noted in results based on different surgical methods used ($p = .000$) and tumor involvement ($p = .002$). Stripping surgery seemed to succeed in preserving more facial functions, whereas involvement of the main trunk or main divisions was predictive of unfavorable treatment outcomes (Table 2).

Tumor recurrence

All tumors were benign schwannomas, as confirmed by histopathologic examination. The recurrence rate was 2.38% (1/42).

Discussion

Since 1949 when the first IFNS was reported and discussed, the optimal diagnosis criteria and surgical method have remained unestablished for approximately 70 years and are still unclear [5]. O'Keefe [2] suggested total resection without preservation of facial nerve, which relied on the preoperative appearance of facial paralysis. However, case series proved that most schwannomas from the peripheral nerve appear as benign painless masses without any neurological symptoms, with an extremely low recurrence rate, and hence require nerve-sparing surgeries. Various management strategies have been proposed, among which, conservative treatment has been highly recommended recently by researchers [5,8]. Nevertheless, in most of these cases, diagnosis was made mostly during surgeries rather than preoperatively.

Benign parotid tumors usually present as a soft- or medium-textured, mobile, round, well-defined mass. Although the presentation of IFNS is similar to that of benign parotid tumors, based on our clinical experience, IFNS tend to have a hard texture and limited mobility. Schwannomas usually present soft masses based on gross pathology; however, their development can be limited by the nerve epineurium or surrounded by the compact parotid sheath, and hence, these tumors can also present a hard texture, as observed in the present study (18/39). The track of facial nerve was constantly still, and hence the limited mobility of the tumor was reasonable; it could be moved well perpendicular to the nerve but remained fixed along the nerve, as in the present study (21/39). IFNS usually presents as a round or oval mass, while Zhong et al. [9] considered these tumors to be elongated in shape because their growth was always along the axonal nerve sheath.

The theory regarding multiple facial nerve schwannomas' origin remains unclear. A multicentric hypothesis was proposed and supported [10]. However, contradictorily, bridging of different lobulations, as observed on microscopic examination, were reported to indicate a single origin [11]. Our research supported the multicentric hypothesis, for a patient whose dozens of masses were distributed in different facial nerve branches, without the appearance of 'bridging'.

Based on available literature, the incidence of preoperative facial nerve paralysis ranges 10.5–20% [4,12], in our study, this incidence was 7.1% (3/42). Preoperative facial nerve paralysis happened in reportedly 61.4% of patients with intratemporal facial nerve schwannoma [3]. The marked difference in these incidence rates indicates the importance of surrounding tissue structure. Zhang et al. [4] believed the intratemporal involvement of IFNS was the major cause of facial paralysis; however, in our study, only 1 of 3 patients who experienced facial paralysis showed involvement into the stylomastoid foramen. Since IFNSs are benign tumors, we believed that the pressure exerted on the axons by such tumors, and the lack of internal blood supply of such tumors, was the primary reasons for fiber degeneration; tumor involvement of the fallopian canal could possibly be a high-risk factor.

CT scans of the study usually showed a round, well-circumscribed mass that was isodense with the parotid gland or showed heterogeneous enhancement. Previous studies have suggested that CT scans showing higher peripheral signal intensity on MRI T2-weighted and gadolinium-enhanced T1-weighted images (the 'target sign') [6] prompt the possibility of schwannomas; however, this 'target sign' could barely be found in any of our cases. Tumor involved the fallopian canal and broaden the stylomastoid foramen was showed on five cases (Figure 2). Therefore, we suggest that a parotid tumor involving the stylomastoid foramen detected by any imaging technique should prompt the differential diagnosis of a schwannoma, but only after excluding the possibility of malignancy.

Microscopic characteristics of IFNS are relatively well established. Following are the two most common microscopic patterns of IFNS: Antoni A shows a high density of spindle cells arranged in fascicles, whereas Antoni B shows lower cellular density, often in a reticular pattern, with abundant myxoid and microcystic changes. Typically, recognition of Verocay bodies is the characteristic sign of a

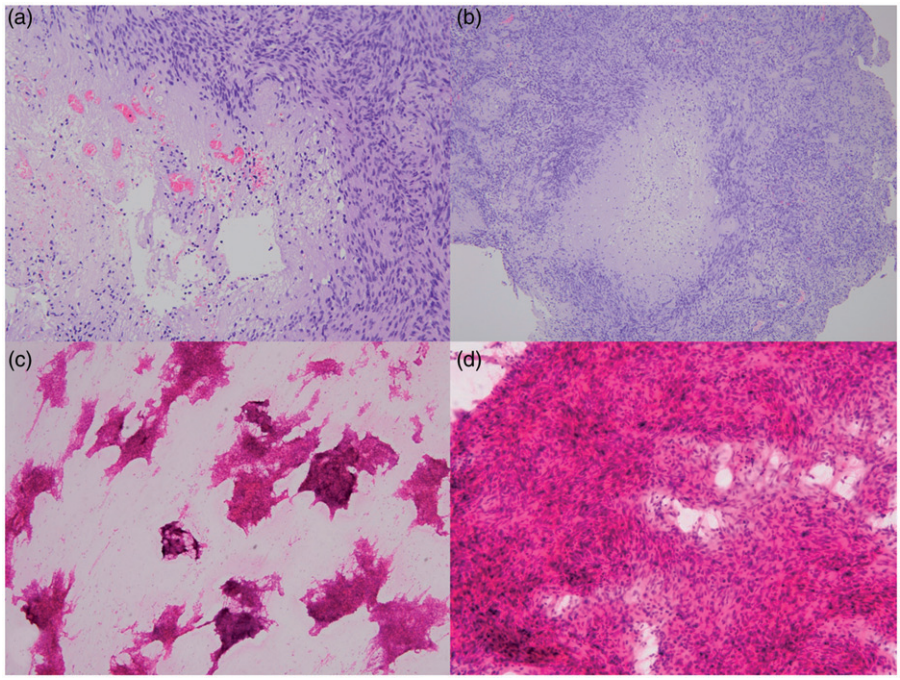

Figure 3. The histopathological findings of a female patient. (a) findings of tumor after resection (HE, ×200); (b) findings of tumor after resection (HE, ×100); (c) findings of fine-needle aspiration (HE, ×40); (d) findings of fine-needle aspiration (HE, ×200).

schwannoma. Immunohistochemistry with strong positive immunostaining for S-100 and GFAP, the lack of calponin, p63, and SMA [13], can be used as differential characteristics against parotid gland tumors. Intraoperative frozen sections have been highly recommended for IFNS by several authors [5,8,14], but the technique also bears the risk of damaging facial nerve axons [4,7]. We believe that intraoperative frozen sections are imperative for decision making during surgery in order to exclude malignant tumors, pleomorphic adenomas or neurofibromas. However, this warrants meticulous observation to identify nerve fascicles or electrical stimulation [14] in order to identify non-nerve region for safe sectioning.

FNAC is recommended as a highly valuable adjunct with a low risk for preoperative differentiation of benign and malignant parotid gland tumors. However, its accuracy in diagnosing IFNS has been only 22–33% [4,6], possibly because of the absence of Verocay bodies or the presence of only noncharacteristic spindle cells. In our study, two patients underwent preoperative FNAC, and both were precisely diagnosed with schwannomas (Figure 3). A part of Verocay body could be spindle cell nucleus arranged in a swirl. Meanwhile, using the fine needle does not mean just acquiring a handful of tissue volume; in fact, adequate cellular samples for microscopy, an accurate sampling site that can be detected via ultrasound guidance [6], and examination by experienced pathologists can increase the diagnosis rate. Taken together, FNAC seems a promising method for preoperative diagnosis.

Fourteen patients were accurately diagnosed of schwannoma – 10 by surgical experience, 2 by FNAC, and 2 by CT scan – but their prognosis of facial nerve function was not ideal. Previous surgical experiences were unquestionable factors for existing nerve injury (8/10), and those terminated surgeries indicated selectively high operation difficulty, predicted poor treatment outcomes as well. Imaging diagnosis relied on the revealed tumor involvement into the stylomastoid foramen, which had increased the risk of degeneration of axons and made resection more challenging. Only two FNAC-diagnosed patients recovered favorable facial function after surgery.

Localization of tumor could not be designated as another category owing to both low incidence and shortage of data for validation from available literature, this category was considered irrelevant either in cases with Type A IFNS (proposed classification by Marchioni et al. [12]) wherein neoplasms were resectable without interrupting the facial nerve. For the remaining Types B, C, and D with tightly adherent schwannomas, the proportion of tumors that involved the main trunk was 69.7–81.5% [12,15]. In the present study, the occurrence of tumors that involved the main trunk, temporofacial division, and cervicofacial division was 71.4% (30/42). In our experience, facial nerve branch schwannomas are easier to manage owing to the possibility of limited damage.

Total tumor resection with nerve preservation is technically possible. Reportedly, the success rate of stripping surgery in cases of intratemporal schwannomas was ideal [16], and intracapsular enucleation has been known to provide better outcomes [15,17]. The gross relationship between the facial nerve and the schwannoma should be cardinally defined. The location of the fasciculus in a transverse section of the schwannoma is grossly classified into two categories: pushed aside eccentrically or separated centrally [5]. According to Marchioni et al. [12], Type A schwannomas account for only 41.3% of IFNS. Hajjaj et al. [18] suggested that 15 of 23 (65.2%) intratemporal schwannomas pushed the nerve trunk peripherally; thus, about half of IFNS tumors loosely touched the facial nerve. In such cases,

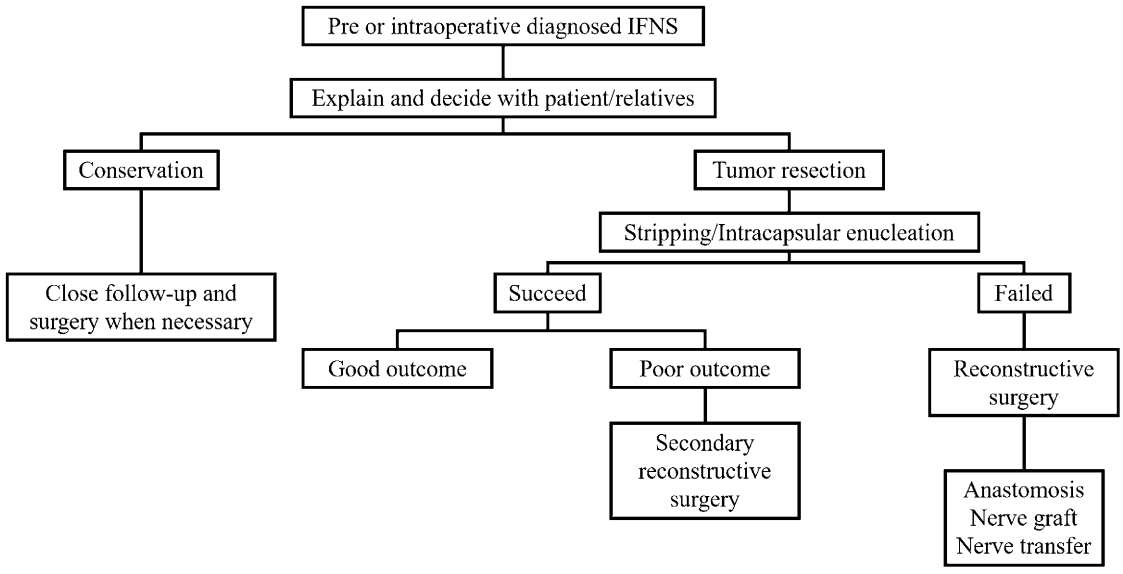

Figure 4. Proposed intraparotid facial nerve schwannoma treatment algorithm based on decision making and the success or failure of nerve-sparing surgery.

stripping surgery is suggested, as long as boundaries of the facial nerve are clear on the tumor surface. In our study, all 17 cases of stripping surgery all recovered to at least HB Grade III.

If locating fasciculus was obstructed by uncertain boundaries formed by the tumor body, intracapsular enucleation was implemented. The fasciculus might be separated peripherally or centrally, but both types presented a tumor that was tightly attached with the facial nerve. After incising the capsule, dissection was performed meticulously and gently, under microscopic guidance as recommended [15], and total dissection could usually be achieved. There was no need to dissect the tumor capsule from the remaining facial nerve fasciculus in such cases as it does not lead to recurrence [17].

Theoretically, flexible application of both stripping surgery and intracapsular enucleation could help to preserve facial nerve integrity. While in practice, surgical resection may lead to injury of fasciculus or even axons due to negligence, or nerve degeneration in extreme cases wherein the fasciculus would become fragile. In any such case of severe damage, the facial nerve should be reconstructed. Several studies have suggested gross tumor resection with primary nerve grafting for patients with a preoperative facial function of HB grade IV or worse because functional recovery after nerve preservation is not better than that after reconstruction in such cases [5,7]. But, in our study, patients with preoperative facial function HB Grade V recovered to Grade II or III after nerve sparing surgery; this might because the environment in the parotid gland differs from that in the temporal bone wherein the facial nerve has better conditions for reanimation. Thus, reconstructive surgery should be performed only after an attempt at nerve preservation has failed.

To our experience and literature, faster recovery and better functional outcomes can be expected in cases with direct end-to-end anastomosis. The suture requires no tension, with both healthy ends placed in end-to-end or end-to-side patterns. Nerve grafts could solve the tension problem, but could not expect better outcomes. The greater auricular nerve is usually the preferred donor nerve, which can be harvested at a single surgical site; reportedly, it is also appropriate for defects in multiple branches. Nerve transfer was required when the proximal end was not achievable and the mimetic muscle needed another motor nerve. The use of a masseteric-facial nerve transfer was recently recommended as it accounted for greater source power and less donor site morbidity. So, in our research, the optimal reconstructive method was determined based on the length and location of a defect, the status of the proximal end, and the duration of existing paralysis.

Secondary stage surgery should be performed as a salvage method. A management algorithm was proposed to determine the best facial nerve outcomes (Figure 4).

The conservative strategy depends on a definite diagnosis, which was rarely achieved before surgery. An IFNS surgery terminated after biopsy, decompression, or even not any further operation, might cause a temporary facial paralysis [8], which was also noted in our patients with previous surgical experience. Researches focusing on intratemporal schwannomas have indicated that subtotal resection could provide better outcomes than total resection [19], but tumor regrowth might be a severe disadvantage [20]. Slow-growing schwannomas might present as enlarged tumors after decades, involving more nerve fibers, and thus present more challenges during surgery; furthermore, involvement of the stylomastoid foramen would evidently increase the risk of paralysis before or after surgery. Therefore, conservative strategy could be appropriate for elderly patients, while in other cases, it should be adopted discreetly after complete explanation and open communication with patients.

Conclusion

A hard-textured tumor with limited mobility mass in the parotid gland should prompt the diagnosis of a

schwannoma. FNAC is likely to be the promising examination technique for diagnosis. The choice of treatment method should be made by patients after information and possible outcomes were provided without tendency. Facial nerve preservation should always be essential, and stripping surgery or intracapsular enucleation could be the preferred surgical methods of choice. Reconstructive surgery should be promptly performed during primary stage surgery and used as a salvage option during additional secondary stage surgery.

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Disclosure statement

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