Extracranial Non-vestibular Head and Neck Schwannomas: A Ten-year Experience

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Abstract:

Introduction: We present a series of head and neck extracranial non-vestibular schwannomas treated during a ten-year period, assessing epidemiology, presenting signs and symptoms, location, nerve of origin, diagnostic modalities, treatment and clinical outcome. Materials and Methods: Clinical records of all patients with head and neck schwannomas treated at our department from April 1995 to July 2005 were retrospectively reviewed. Results: There was female predominance (67%). The mean age at diagnosis was 48 years. Sixteen (76%) presented with a unilateral neck mass. Eleven schwannomas (52%) were in the parapharyngeal space. The most common nerves of origin were the vagus and the cervical sympathetic chain. The tumour may masquerade as a cervical lymph node and other myriad conditions. Treatment for all but 2 cases was complete excision with nerve preservation. Two cases of facial schwannoma required sacrifice of the affected nerve portion with nerve reconstruction. All facial schwannoma patients suffered postoperative facial palsy with only partial resolution (mean final House-Brackman grade, 3.25/6). Among non-facial schwannoma patients, postoperative neural deficit occurred in 12 with partial to complete resolution in 7. The median follow-up period was 24 months. No schwannoma was malignant and none recurred. Conclusion: Non-vestibular extracranial head and neck schwannomas most frequently present as an innocuous longstanding unilateral parapharyngeal neck mass. Preoperative diagnosis may be aided by fine-needle cytology and magnetic resonance imaging or computed tomographic imaging. The mainstay of treatment is complete intracapsular excision preserving the nerve of origin, but for extensive tumour or facial schwannomas, subtotal resection or nerve sacrifice with reconstruction and rehabilitation are considerations. Surgery on intraparotid facial schwannomas carries considerable morbidity and conservative management has a place in treatment. Early recognition of facial schwannomas is key to optimal treatment.


Keywords: Asian, Extracranial non-vestibular, Head and neck schwannomas, Single institution

Introduction

Schwannomas, also known as neurilemmomas, neuromas, or neurinomas, are uncommon nerve sheath neoplasms that may originate from any peripheral, cranial or autonomic nerve of the body with the exception of the olfactory and optic nerve. Malignant change is unusual. Some 25% to 45%1 of schwannomas are located in the head, and these often present as diagnostic and management challenges.

Despite the fact that a large proportion of head and neck schwannoma studies is devoted to intracranial acoustic neuromas, the majority of head and neck schwannomas are non-vestibular and extracranial. There are few Asian series on head and neck extracranial non-vestibular schwannomas.

Materials and Methods

All patients with head and neck schwannomas treated at our department between April 1995 and July 2005 were retrospectively reviewed from the medical records of the Singapore General Hospital, the largest tertiary hospital in Singapore. Data collected included patient age, sex, race, presenting signs and symptoms, anatomical location of the tumour, tumour size, nerve of origin, diagnostic modality, surgical approach, intraoperative finding, histopathological finding, and outcome after treatment.

The presence of characteristic Antoni A or B histologic patterns with or without S-100 stain was used to identify schwannomas. S-100 immunohistochemical staining is
positive in most Schwann cell-derived tumours. The absence of mitoses, necrosis, invasiveness, and specific features such as nuclear hyperchromia and pleomorphism or large atypical cells confirms the benign nature of the schwannoma. Grossly identified, schwannomas are well-circumscribed encapsulated firm, grey myxoid masses attached to nerve but may have areas of cystic and xanthomatous change.

Results

Twenty-two patients with head and neck schwannomas were identified. One patient with no information other than age and diagnosis of a recurrent head and neck schwannoma was excluded from the study. Our study set of schwannomas was necessarily extracranial non-vestibular and mainly non-trigeminal because the vestibular and trigeminal schwanna patients usually receive tertiary treatment through either otolaryngology or neurosurgery.

The study population consisted of 7 males and 14 females, with a mean age of 47.7 years (range, 26 to 67) and a median age of 48 years. Each had a solitary head and neck schwannoma and none suffered from neurofibromatosis. Fifteen (71%) of the 21 patients underwent preoperative computed tomography (CT) or magnetic resonance imaging (MRI) to facilitate diagnosis and delineate extent and related anatomy. One had only ultrasound imaging of the tumour done preoperatively. All patients underwent surgical excision of the tumour. Table 1 shows the epidemiological data of the population, along with the presenting signs and symptoms, initial diagnosis, and diagnostic modality utilised.

Every patient had at least one presenting symptom or sign, with only 4 patients reporting multiple symptoms or signs. A unilateral neck mass was reported in 16 (76%) patients making it the most common presentation of a schwannoma. Of these 16, 10 had a neck mass that was left-sided. Out of all the cases, 12 (57%) were left-sided schwannomas. There was a singular case of a recurrent right-sided neck schwannoma.

Schwannomas in our case series were distributed over 4 anatomical sites – the parapharyngeal space (11), neck (5), parotid (4) and skullbase (1). The nerve of origin (Table 1) was identified in 20 (95%) of 21 patients. Of those identified, 6 were derived from the vagus nerve; 5, the cervical sympathetic chain; 4, the facial nerve; 2, the brachial plexus; 1, the trigeminal nerve; 1, the accessory nerve; and 1, the hypoglossal nerve.

In all cases, the tumour was completely resected surgically. The surgical approach varied depending on the preoperative presumptive diagnosis, location and size of the tumour, and surgeon preference. Schwannomas of the parapharyngeal space and those limited to the neck were resected via transcervical approaches. All 4 parotid schwannomas were intra-parotid and derived from the facial nerve such that complete resection mandated at least a superficial parotidectomy.

Postoperative neural deficit separate from or worse than that reported preoperatively was documented in 16 (76%) patients. Two patients had more than one operation-related neural deficit. Facial nerve injury was evident in 5 patients. Table 2 displays the postoperative nerve injuries and resolution of neural deficits. None of the patients suffered wound infection, haematoma, or cerebrospinal fluid leak, which are possible characteristic sequelae reported in other series.

The patient follow-up period ranged from 1 month to 108 months (median, 24 months). All remained free of disease at last follow-up consultation. None of the schwannomas was malignant.

Discussion

Non-vestibular extracranial head and neck schwannomas are more common but less researched than their relatively more morbid intracranial acoustic counterpart. Few studies on this specialty subject are derived from Asia and this is the largest Southeast Asian series to the best of the authors’ knowledge.

Two-thirds of the study population were female in line with a study by Torossian et al observing female predominance in extracranial cephalic schwannomas, but at variance with Leu and Chang’s series noting male predilection. More striking in the population was the preferential distribution of the tumour on the left side. This especially held true for schwannomas presenting as non-supraclavicular neck masses – 9 of 14 (64%) were left-sided. All patients in the study were of Chinese ethnicity, apart from 3 (14.3%) ethnic Indians.

Three-quarters of the cases presented with an asymptomatic stable neck mass that caused little concern other than the possibility of malignancy and cosmesis. Many of these neck masses were longstanding and have been manifested in the patient for as long as 3 to 9 years. More than half (52%) of the schwannomas in the study, comprising 6 vagal and 5 cervical sympathetic chain schwannomas, was anatomically located in the parapharyngeal space. This concurs with other series identifying the parapharyngeal space as the most common non-vestibular head and neck schwanna location. Of note, 29% of schwannomas in the series originated from the vagus.

While neurogenic presenting symptoms and signs would be expected to feature more prominently befitting cephalic neurogenic tumours, only 4 (19%) cases had any neurogenic symptoms or signs at all. Possible neurogenic symptoms and signs related to the non-vestibular tumour include pain...
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<td>FNA – blood, degenerate cells, spindle cells CT neck – vagal schwanna</td>
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<td>Left neck mass – many years</td>
<td>Carotid body tumour</td>
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<td>KBL</td>
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<td>Left neck mass – 5 years</td>
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<td>KAG</td>
<td>64/Female</td>
<td>Right neck mass – 1 year</td>
<td>Carotid body tumour</td>
<td>CT neck – enlarged lymph node or carotid body tumour</td>
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<td>CCK</td>
<td>67/Female</td>
<td>Right neck mass – 2 years</td>
<td>Level II or III lymph node</td>
<td>FNA – schwannoma MRI neck – schwannoma from vagal or sympathetic nerve</td>
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<td>SGC</td>
<td>41/Male</td>
<td>Left parotid swelling – 1 year</td>
<td>Parotid tumour</td>
<td>FNA – pleomorphic adenoma or schwannoma</td>
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<td>FKY</td>
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<td>Parotid tumour</td>
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<tr>
<td>GG</td>
<td>48/Female</td>
<td>Left parotid swelling with slight pain – 1 year</td>
<td>Parotid tumour</td>
<td>Nil</td>
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<tr>
<td>TBN</td>
<td>64/Female</td>
<td>Right parotid swelling – 7 months</td>
<td>Parotid cyst</td>
<td>Nil</td>
<td>Facial</td>
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<tr>
<td>LWY</td>
<td>34/Male</td>
<td>Left neck mass, tender – 5 years</td>
<td>Level III lymph node</td>
<td>FNA – schwannoma, solitary fibrous tumour, or other spindle cell lesion CT neck – neuroma</td>
<td>Brachial plexus</td>
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and tenderness, coughing upon exerting pressure on tumour, headache, Horner’s syndrome, cranial nerve palsies (particularly V, VI, VII, XII), tinnitus or hearing loss. It has been noted that patients with neurogenic presenting signs and symptoms often had tumours located in more confined spaces such as the middle ear, sinonasal cavity, or skull base. In our series, one (LWY) with a brachial plexus schwannoma and another (GG) with a parotid schwannoma both complained of pain and tenderness of the tumour. The case of a right skull base trigeminal schwannoma – the only “confined” schwannoma in the series – presented with a multitude of occipital headache and right V, VII and XII nerve palsies, likely resulting from a combination of neuropraxia of the affected nerve and mass compression of adjacent nerves. The fourth case, that of yet another brachial plexus schwannoma initially diagnosed as a possible Virchow’s node, had tenderness and upper limb paresthesiae (Tinel’s sign) upon compression, with upper limb paresthesiae and pains elicited during fine-needle aspiration (FNA).

Schwannomas almost always are diagnostic problems because the history and clinical examination are non-specific and deceptive. Often, as was the situation in many cases of this series, the unilateral asymptomatic neck mass is diagnosed clinically incorrectly as an enlarged lymph node, a carotid body tumour, a brachial cyst, a thyroid cyst or nodule, even a parotid cyst or tumour in the case of parotid schwannomas. Definitive diagnosis and identification of the affected is often difficult up to the time of surgery. However, diagnostic modalities in the form of FNA cytological techniques as well as better imaging in the form of MRI or CT scans have lessened the problem of misdiagnosis to some degree. In addition to facilitating diagnosis, preoperative imaging provides information on tumour size, location, extent and surrounding anatomy, thereby aiding surgical planning.

FNA was performed on 10 patients and only in 2 (20%) was a definitive cytological diagnosis of schwannoma made based on the diagnosis of characteristic Verocay bodies, while in another three the diagnosis was suggested by the presence of spindle cells. FNA was not done in the cases presenting as parotid lumps. The diagnostic accuracy of FNA depends strongly on the specimen quality and the experience of the cytopathologist.

Based on tumour location, morphology and even signal characteristics, the radiological diagnosis of schwannoma was suggested in 11 of 15 (73%) cases in which preoperative MRI and/or CT was performed. Three had MRI alone done, 1 patient had both MRI and CT scanning; the remaining 11 received only CT (Figs. 1 and 2) scanning of the tumour. Contrast-enhanced CT and MRI are respectively useful in assessing bony and soft tissue involvement. MRI characteristics of schwannomas include specific signs (split fat sign, fascicular sign, target sign) and signal patterns (i.e., isointense T1 signal relative to skeletal muscle; increased and slightly heterogeneous T2 signal). MRI also finds application in evaluating schwannomas thought to be highly vascular or closely related to...
vasculature, with angiography used for cases requiring preoperative embolisation. On CT scans, schwannomas appear well-covered, well-defined and fusiform; they show relatively homogeneous contrast enhancement with internal cystic change becoming more prominent as the tumour enlarges. This cystic change is associated with mucinous degeneration, haemorrhage, necrosis, and microcyst formation. Ultrasonic images (Fig. 3) of schwannoma are characterised by a round or elliptical cross-section with a clear border with the internal echo reflective of histology. Patterns may be homogeneous to heterogeneous and cystic change may be seen, as was the case in one patient with a right supraclavicular schwannoma. Ultrasound has greater diagnostic utility when the diameter of the nerve of origin is large and it can be seen that the tumour is connected to the often well-delineated nerve. Even with MRI or CT scans, it was in some instances in our series (Table 1) difficult to distinguish adenopathy from schwannoma. The definitive diagnosis remains that which is derived from tissue.

Schwannomas are management problems as well. Cure entails a complete resection but this conflicts with the surgical instinct to preserve the nerve of origin. Altogether, 16 patients had postoperative neural deficit. For clarity, the cases are divided into non-facial nerve and facial nerve schwannomas.

All 17 non-facial nerve schwannomas in the study were completely excised and while every effort was made to identify and spare the nerve of origin, tumour clearance occasionally necessitated partial nerve sacrifice or...
unwittingly caused nerve compromise. Twelve had postoperative neural deficits caused by iatrogenic injury to either the nerve of origin or adjacent nerves. Analysed by nerve affected, it is noted that vagal schwannomas were associated with vocal cord palsy and voice hoarseness, cervical sympathetic chain with Horner’s syndrome, and hypoglossal nerve with tongue atrophy. One case (BK) of a brachial plexus schwannoma developed postoperative paresis of the ipsilateral upper limb, with weakness maximally evident at the shoulder associated with atrophy of the biceps. Shoulder power decreased from grade 5 before operation to grade 2. This probable iatrogenic neuropraxia, likely of the lateral cord from which the tumour arose, fortunately resolved spontaneously at 6 months.

There were 4 cases of facial nerve schwannoma – all were intraparotid. Intraparotid schwannomas usually present as an asymptomatic slow-growing parotid lump but can cause pain: this was experienced by one of the patients. Complete resection is clearly indicated when they present with moderate to total facial nerve palsy. With mild or no facial palsy, as was the circumstance with these 4 patients (all House-Brackman grade 1 preoperatively), the best course of treatment is controversial. All had a presumptive diagnosis of parotid tumour without preoperative imaging and eventually had tumour excision involving parotidectomy. In all but 1 (TBN) patient, there was intraoperative recognition of a facial schwannoma. All 4 suffered facial nerve dysfunction (mean final House-Brackman grade post-resection, 3.25/6) despite efforts made to preserve or reconstruct neural structures. One patient (GG) had a schwannoma presenting with neural deficits experienced new-onset mild dysphagia post-excision. Excision of the trigeminal schwannoma, located in the infratemporal region between the pterygoids, was via a lateral skull base approach involving the use of an optical tracking system, superior control of the internal carotid artery, zygomectomy and piecemeal tumour removal.

The absence of wound infection, haematoma, or cerebrospinal fluid leak in our series is attributed to careful surgical technique, the generally healthy subset of patients with no major comorbidity, and the accessible cervicoparotid location of most of the tumours.

The singular case (LSC) of recurrent right vagal schwannoma was exceptional in that this was the second local recurrence 16 years to the last resection. The patient had had 2 previous surgeries (one 16 years ago and another 18 years ago) to remove schwannoma in the exact same locality. We postulate that the recurrences stemmed from earlier subtotal resections. After the third surgery, she was well with no recurrence at 14 months.

The case of accessory nerve schwannoma manifested postoperative deficit in the form of wound paresthesiae. The patient with trigeminal schwannoma presenting with neural deficits experienced new-onset mild dysphagia post-excision. Excision of the trigeminal schwannoma, located in the infratemporal region between the pterygoids, was via a lateral skull base approach involving the use of an optical tracking system, superior control of the internal carotid artery, zygomectomy and piecemeal tumour removal.

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large facial schwannoma extending into the mastoid: a total parotidectomy was performed such that the tumour was resected together with the involved facial trunk segment; this was followed by a mastoidectomy and great auricular nerve interposition grafting to reconstruct the facial trunk. She then underwent therapist-guided galvanic stimulation of the reconstructed facial nerve achieving good functional recovery and resting tone (House-Brackman 3/6) at 24 months. Another (FKY) had tumour involving the upper division (temporozygomatic) trunk of the right facial nerve. Likewise the tumour was excised with the involved nerve segment, parotidectomy was done, and the great auricular nerve grafted. He had a postoperative facial palsy as well as sweating over his temporal region when masticating (Frey’s syndrome); partial symptom resolution was attained at 32 months. The other 2 patients (SGC, TBN) underwent superficial parotidectomy with vigilant preservation of facial nerves, in spite of which both went on to develop longstanding eye closure problems.

Reflecting on our morbidity with facial schwannomas, the importance of early recognition of the neurogenic tumour – intraoperatively or otherwise – cannot be overemphasised. It is advisable when operating on parotid tumours to be wary and on the lookout for the possibility of a parotid schwanna. Caughey et al17 recommended a degree of suspicion for facial nerve schwanna if the tumour is intimately associated with the facial nerve, and advocated biopsy when a schwanna is suspected, postponing complete resection until further imaging and options are evaluated in view of the potential disabling palsy. Liu et al18 advised delaying resection of facial nerve schwannomas to allow retention of normal facial function indefinitely, urging nerve decompression or subtotal resection for patients with mild or no facial palsy should surgery be desired.

The mainstay of treatment ideally remains complete surgical excision with preservation of the affected nerve.1,3,8,12 There was no recurrence for all the cases after complete surgical removal. The greatest dimension of the schwannomas excised ranged from 2 cm to 7.3 cm (mean size, 4.4 cm). Most authors3,7 have recommended careful intracapsular excision of the tumour to minimise postoperative neural deficit. Microneurosurgery to facilitate intraoperative microscopic diagnosis of the schwannoma and achieve more superior nerve preservation has been described. This involves microscopic enucleation of the tumour after opening epineurium without disrupting nerve continuity. Schwannomas involving the brachial plexus are best approached with microsurgical assistance to prevent potentially catastrophic iatrogenic injury. Even as all attempts are made to preserve the nerve of origin, structural preservation may not necessarily lead to preservation of its functional integrity – this was apparent among our patients with postoperative neural deficit.

The literature4,18 hitherto appears to be in favour of the concept of subtotal or near-total tumour resection for non-vestibular head and neck schwannomas in situations where tumour is extensive and complete tumour resection cannot be achieved without compromising neural integrity and causing significant morbidity from paralysis and sensory loss. This approach is particularly suited to elderly patients or unfit patients who cannot tolerate prolonged surgery. Malignant transformation is exceptional in solitary schwannomas and evidence suggests that subtotal resection may provide adequate disease control of non-vestibular head and neck schwannoma, although continued follow-up is mandated. Judging from the lag time in our case of recurrent vagal schwanna, recurrence after subtotal resection may take 2 years to 2 decades. Generally, as illustrated par excellence in the aforementioned cases of large facial trunk schwannomas, if the nerve of origin is sacrificed, immediate reconstruction and postoperative rehabilitation should be undertaken in the context of a multidisciplinary management team.12

The role of radiosurgery as an alternative to surgical resection is a matter of ongoing debate. Recent small-scale studies19,20 reviewed outcome following stereotactic radiosurgery of non-vestibular cranial nerve schwannomas and reported good tumour control rates and less morbidity in terms of post-treatment neural deficit compared with surgical resection. The patients treated had, among other types of schwannomas not in our population, jugular foramen region, facial, hypoglossal, and trigeminal schwannomas. Some had undergone previous subtotal resection but most had no prior treatment. While intuition may favour radiosurgery over resection for high-risk patients and residual or recurrent tumour, larger case-control studies are required before final conclusions can be drawn. Certainly, a conservative non-operative approach is a valid option for head and neck schwannomas if the diagnosis has been established and the patient is tolerating the tumour well without neurogenic deficit, mass effect or rapid progression. Surgery, however, remains definitive treatment.

The most important differential diagnosis of a firm defined unilateral upper neck mass so characteristic of neck schwannomas must be an enlarged lymph node. In view of this, the approach entails a detailed history, meticulous examination of the head and neck, chest, abdomen, and select investigations targeted at identifying a source of lymphadenopathy. Flexible nasoendoscopic examination of the nasopharynx, oral cavity and larynx should be done. FNA is most helpful in determining the nature of the mass. If the diagnosis remains unknown, an MRI or CT neck is the further diagnostic investigation of
choice, failing which excision biopsy is indicated, adhering to the surgical concepts highlighted should an intraoperative diagnosis of schwannoma be made.

This single-institution study explored a dedicated series of extracranial non-vestibular schwannomas in terms of clinical characteristics, diagnosis, treatment and its outcome. The most likely presentation of an extracranial non-vestibular head and neck schwannoma is an asymptomatic longstanding left-sided parapharyngeal space neck mass in a middle-aged woman. Neural deficits are uncommon presenting findings. The vagus is the most common nerve of tumour origination. Definitive preoperative diagnosis is difficult to achieve but may be aided by FNA and generous MRI, CT or ultrasound imaging. The treatment of choice for schwannoma in principle is complete excision preserving the nerve of origin, and this can be achieved in most cases with minimal postoperative morbidity, but circumstances such as extensive tumour or facial schwannomas may point to subtotal tumour resection or nerve sacrifice with immediate reconstruction and rehabilitation. Surgery on intraparotid facial schwannomas carry considerable morbidity and conservative management may have a place; timely recognition of facial schwannomas is critical for optimal treatment. None of the tumours in the study were malignant and there was no recurrence during the follow-up period post-surgery.

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REFERENCES