



SCHWANNOMAS OF THE AERODIGESTIVE TRACT – A TALE OF 4 CASES WITH LITERATURE REVIEW

ENT

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ABSTRACT

Background: Schwannomas are benign tumors arising from Schwann cells of peripheral nerve sheaths. While they predominantly involve the vestibulocochlear nerve, approximately one-quarter to nearly half may occur in extracranial regions of the head and neck, where they often pose diagnostic and therapeutic difficulties due to variable presentation. **Objective:** This study aims to present four cases of extracranial schwannomas in the head and neck, highlighting their clinical features, imaging findings, surgical management, and histopathological characteristics, along with a brief literature review. **Methods:** A retrospective analysis was conducted on four patients diagnosed with schwannomas located in the larynx, tongue, and nasal cavity. Comprehensive clinical evaluation, imaging (CT or MRI), and complete surgical excision were performed in each case. Histopathological diagnosis was confirmed with S-100 immunohistochemical staining. Relevant literature was reviewed to support and compare findings. **Results:** Patient ages ranged between 14 and 49 years, with symptom duration from 2 months to 5 years. Three lesions were situated in the nasal cavity, and one each in the larynx and tongue. All tumors appeared well-defined, and a definitive nerve of origin could not be identified in three cases. Histology showed classic biphasic Antoni A and B areas, with positive S-100 staining. There were no operative complications, and no recurrences were observed during one year of follow-up. **Conclusion:** Extracranial schwannomas in the head and neck are uncommon and can mimic other benign masses depending on their location. A combination of clinical suspicion, radiological imaging, and histopathology is essential for accurate diagnosis. Surgical excision offers favorable outcomes, while a conservative approach may be suitable for asymptomatic patients given the indolent nature of the disease.

KEYWORDS

Peripheral nerve sheath tumor, Schwannoma, Head and neck neoplasms, Nasal mass, Laryngeal tumor, Antoni A and B, S-100, Case series

INTRODUCTION:

Schwannomas are benign, well-circumscribed, slow growing nerve sheath tumours arising from the Schwann cells. These tumours commonly occur in the cranial nerves with a predilection for affecting the sensory nerves with the 8th cranial nerve being the most common. 25-40% of the schwannoma are located in the extracranial head and neck region involving cranial nerves V, VII, X, XI, XII and or sympathetic and peripheral nerve sheaths. The presentation of these tumours varies based on the site of origin. Here we describe 4 such cases of schwannomas in the extracranial head and neck region and our experience in managing them.

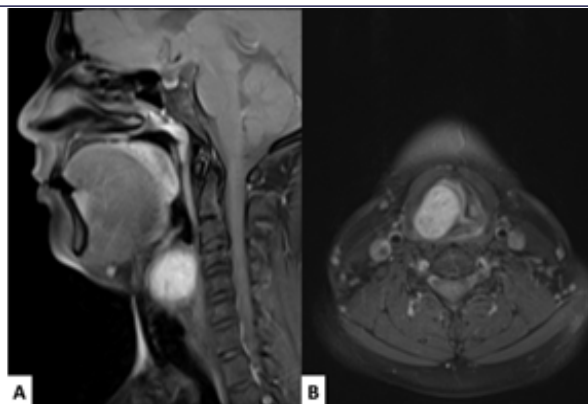
CASE 1:

A 38years old male presented to us with complaints of muffled voice for 5 years, insidious onset and gradually progressive. There was no complaint of breathing difficulty, or swallowing difficulty, and no history suggestive of aspiration. There wasn't history of smoking or alcohol consumption. On examination, laryngeal crepitus was present, and tenderness was absent. On laryngoscopy (Fig.1), a smooth swelling was noted in the right aryepiglottic fold and the false cord, which was obscuring the right true cord. The left vocal cord was mobile. No similar swellings were noted elsewhere in the body.



<Fig 1: Video laryngoscope image>

MRI done showed a single well-circumscribed heterogeneously enhancing T1 hypointense T2 hyperintense ovoid lesion of size 3x2.5x3.4cm in the right aryepiglottic fold extending into the right false cord. No invasion was noted into the surrounding structures. The lesion showed progressive enhancement on dynamic post contrast images (Fig.2)



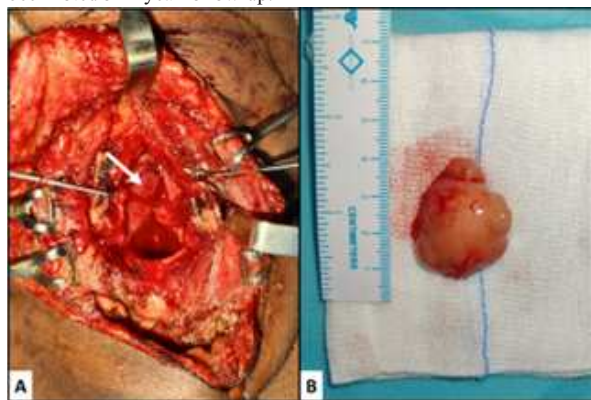
<Fig 2: MRI showing tumour in the supraglottic region.>

Due to the slow progression and the well circumscribed and apparent non-invasive nature of the lesion a schwannoma, a pleomorphic adenoma and a haemangioma were the suspicions. Patient was thus planned for complete excision of the lesion. A laryngo-fissure approach was performed due to the relatively large size and posteriorly based location of the lesion (Fig 3A) During dissection no particular nerve of origin was identifiable. No major intra-operative complication was encountered. Mild oedema was noted in the right hemi-larynx which resolved one-week post-surgery.

On gross examination of the mass, it was found to be firm with a smooth, yellow surface, with multiple dilated blood vessels over it (Fig 3B). Histopathological examination showed regions of hypercellularity and hypocellularity corresponding to Antoni A and B areas respectively. Individual cells were noted to have elongated wavy nuclei with ill-defined cellular borders and scant cytoplasm. Numerous Verocay bodies were seen in Antoni A areas. Immunohistochemical studies revealed that the cells of the lesion were strongly positive during S-100 staining. All these showed that the lesion was a schwannoma.

The patient was placed on regular follow-up and no recurrence has

been noted on 1-year follow-up.



<Fig 3: A: Tumour visualised via laryngo-fissure approach. B: Complete tumour after removal.>

CASE 2:

A 30years old female presented to us with a swelling involving the submental and the submandibular regions (Fig 4) for the 3 years. It had begun insidiously and was gradually increasing in size. Patient had difficulty in chewing and had a muffled voice. On examination, she had a single firm swelling of size 6x5cm with well-defined margins present in the submental and bilateral submandibular regions. It was seen to extend into the floor of mouth, pushing the tongue superiorly and posteriorly. It was firm in consistency and bimanually palpable with the skin superficial to the swelling pinchable. On protruding the tongue, the tongue was found to deviate to the right.



<Fig 4: Swelling noted in the submental and submandibular regions.>

MRI showed a single heterogeneously enhancing T2 hyperintense and T1 hypointense well defined ovoid lesion of size 5.1x6.5x6.3cm pushing the mylohyoid inferiorly and the tongue superiorly and posteriorly with no invasion into these structures. The sub-mandibular glands were normal. (Fig 5)



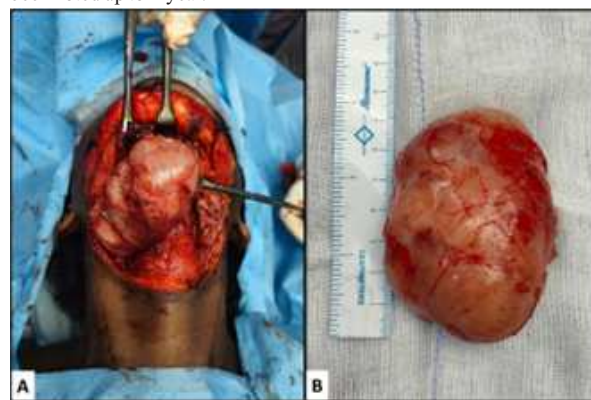
<Fig 5: MRI image of the patient showing a well-defined mass in the sublingual region>

The differentials considered were a pleomorphic adenoma, a

schwannoma, and a rhabdomyoma. Due to the well-circumscribed nature of the lesion a complete excision was planned. Transcervical approach was chosen to avoid unduly large incision on the tongue and floor of the oral cavity. (Fig 6A) The hypoglossal nerve was identified and was found to be free from the tumour. No particular nerve of origin was identifiable during dissection. No major intra-operative complication was encountered. The deviation of the tongue towards the right resolved one month postoperatively.

On gross examination of the mass, it was found to be firm with a smooth, yellowish surface, with multiple dilated blood vessels over it. (Fig 6B). Histopathological examination showed cells arranged in biphasic pattern with hypercellular Antoni A areas and hypocellular Antoni B areas. Many Verocay bodies were noted in the hypercellular areas. Individual cells were spindle shaped with moderate amount of cytoplasm, and wavy nuclei with tapered ends. Based on these features a diagnosis of Schwannoma was arrived at.

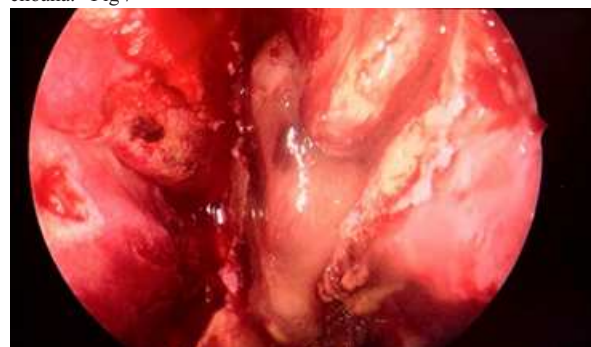
The patient has been kept on regular follow-up and no recurrence has been noted up to 1-year.



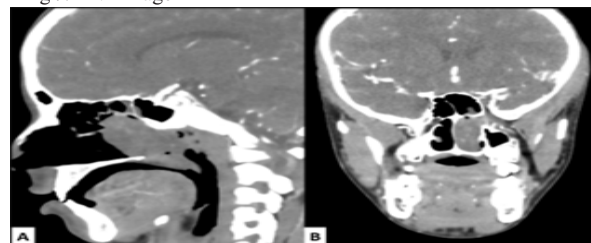
<Fig 6: A: The tumour being approached by a transcervical approach. B: The tumour after excision.>

CASE 3:

A 14years old boy came with complaint of bilateral nasal block for 1 year, which had started insidiously from the left side and gradually progressed to complete bilateral block. There was complaint of occasional nasal bleed from the left nasal cavity which was few drops in quantity. No complaint of ear block or ear discharge was present. On nasal endoscopic examination, a pinkish mass was noted in the left nasal cavity abutting the posterior ends of the inferior and middle turbinates, extending in to the nasopharynx and obstruction the right choana. <Fig 7>



<Fig 7: DNE image>



<Fig 8: Contrast enhanced CT image showing a heterogeneously enhancing soft tissue lesion in the left nasal cavity>

Contrast enhanced CT showed a lobulated, heterogeneously enhancing soft tissue lesion of size 4.8x2.4x3.4cm in the in the left nasal cavity and nasopharynx. Widening of the left sphenopalatine foramen was noted suggesting a probable origin at that site. A few feeders were noted from the external carotid artery system. No bony erosions were noted. (Fig 8)

Considering the age of the patient and the clinical features, a diagnosis of Juvenile Nasopharyngeal Angiofibroma was considered. Patient was planned for complete excision of the tumour. After arterial embolization was performed 24 hours prior to the procedure. A trans-nasal endoscopic approach was chosen due to the tumour being confined to the nasal cavity and nasopharynx. In contrast to the radiological findings, the tumour was not found to be entering the sphenopalatine foramen but was found to be attached to the posterior nasal nerve just below the foramen. The mass was completely delineated using coblator assistance and removed in-toto. No major intra-operative or post-operative complication was encountered.

On gross examination the tumour was found to be pinkish and lobulated with a firm consistency. Histopathological examination showed a fairly circumscribed neoplasm composed of spindle shaped cells and showing a biphasic pattern with hyper and hypocellular areas. The cells had indistinct cell borders, scant to moderate cytoplasm, vesicular nuclei. Considering the intra-operative findings and the histopathological examination, we came to the diagnosis of schwannoma.

The patient was placed on regular follow-up. No recurrence has been noted during a 1-year period.

CASE 4:

A 49 years old female presented with complaints of right sided nasal block for 2 years. There was no complaint of nasal bleed or watery nasal discharge. On examination, a pale smooth mass was found in the right nasal cavity abutting the inferior and middle turbinates extending up to their anterior ends and the floor of the nasal cavity. No cough impulse or pulsation was observed. (Fig 9) On probing, it was found to be firm in consistency and did not bleed on touch. Probing was possible all around the mass except along the septum.



<Fig 9: DNE image>

On CT, a well-circumscribed soft tissue lesion was noted occupying the right nasal cavity extending into the nasopharynx, causing outward bowing of the medial wall of the right maxillary sinus. It wasn't seen to extend to the roof of the nasal cavity. No bony erosions or intra-cranial extensions were noted. (Fig 10)



<Fig 10: CT image>

The lesion was considered to be a schwannoma. Due to the well-circumscribed nature of the lesion, a trans-nasal endoscopic excision of the entire tumour was performed. During the procedure, the tumour was found to be attached to the posterior part of the nasal septum and the anterior wall of the sphenoid. No nerve of origin was identified. No complication related to the surgery was encountered.

On gross examination, the tumour was a firm smooth pinkish mass. Histopathological examination showed a fairly circumscribed lesion composed of spindle shaped cells arranged in a biphasic pattern with hypercellular areas (Antoni A), hypocellular areas (Antoni B) and nuclear palisading. Individual cells are spindle-shaped with moderate eosinophilic cytoplasm and wavy hyperchromatic nucleus. Immunohistochemical staining was present when performed with S-100. These findings confirmed our suspicion of a schwannoma.

The patient was placed on regular follow-up. No recurrence has been noted during a 1-year period.

DISCUSSION:

Schwannomas are benign, slow growing nerve sheath tumours arising from the Schwann cells. 25-40% of the schwannoma are located in the extracranial head and neck region involving cranial nerves V, VII, X, XI, XII and sympathetic and peripheral nerve sheaths with the most common being the sympathetic trunk(33%), vagus(20%).^(1,2) The nerve of origin, however, cannot be identified in all cases. In our series we weren't able to identify the nerve of origin in 3 of the cases. In the series by Liu et al. and Shrikrishna et al. they weren't able to identify the nerve of origin in 17% and 25% of their cases respectively.^(3,4)

Phulware et al. in their meta-analysis had identified the possible locations in the head and neck region where schwannomas can occur as tongue, lower lip, nasal cavity, buccal mucosa, parapharyngeal space, eyebrow, upper lip, lateral canthus eye, infraorbital region, submandibular gland, and external auditory canal. Of these the parapharyngeal region of the neck was identified as the commonest region of occurrence by Biswas et al.⁽⁵⁾ This is in contrast to Phulware et al who had identified tongue as the commonest site of occurrence.⁽⁶⁾ In our series the commonest site was the nasal cavity, while 1 was found in the larynx and tongue each.

Schwannoma are relatively rare tumours with peak incidence in the 20 to 60 years age group but can occur at any age with the youngest patient described being a 7 year old and the oldest a 77 year old.⁽⁶⁻¹⁰⁾ No gender predilection has been identified. Our patients were in the 10 to 50 years age group with the youngest being a 14year old and the oldest being a 49year old. None of the patients had multiple lesions or family history of similar lesions.

Schwannomas are usually sporadic in nature. Multiple authors in their analysis have described the association of schwannomas with genetic syndromes such as neurofibromatosis 2 (1 in 25,000 to 1 in 40,000 cases), schwannomatosis (1 in 40,000 cases), and Carney's complex (prevalence unknown).⁽¹¹⁻¹⁶⁾ They had observed that in these cases multiple schwannomas and associated tumours may be observed and familial history of similar lesions will be present. In our study, all the patients had single identified lesions and no family history of similar lesions suggesting that these were sporadic in nature. Enzo et al. in their study had identified the probability of malignant transformation of head and neck schwannomas to be 8 – 10%.⁽¹⁰⁾

As described by Majumder et al. and Shrikrishna et al., on gross appearance, a classical schwannoma has a well-defined, encapsulated appearance.^(4,17) The tumours which we had excised from the tongue and larynx were well-circumscribed. But the tumours removed from the nasal cavities had a lobulated appearance. Butler et al, Camak et al. and Camak et al., encountered similar findings when the removed tumours from the nasal cavities. In addition they had noticed the absence of a capsule.⁽¹⁸⁻²⁰⁾ This was in contrast to our findings were all the tumours in our series had a capsule.

Histologically schwannoma is described to be covered by a fibrous capsule and residual nerve fibers with the stroma exhibiting two distinct regions: Antoni A regions showing hypercellular spindle cells, which sometimes palisade around eosinophilic areas (Verocay bodies) and Antoni B regions showing a hypocellular myxomatous pattern with a background of loose connective tissue.^(11,17,21-23) We observed similar findings in all the tumours. Cysts, haemorrhage, and fatty degeneration may be present.⁽²³⁾ Calcifications and mitotic figures are rare. Tumours with such changes are known as ancient schwannomas.^(9,24) Few other secondary changes reported in literature include melanotic schwannoma and psammomatous melanotic schwannoma.⁽²⁵⁾ However, no such changes were encountered by us. As S-100, a neural crest marker's helpfulness in identification of the tumour has been described, we used it and found it to be positive in all our cases.^(9,26,27)

The presenting features of these tumours depends on their site and size. Enoz et al. and Phulware et al. in their analysis had identified that the most common presentation is as a painless, slow-growing swelling present for a duration of 2 months to 2 years with the average size at presentation being 1-2cm.^(6,10) Our patients had presented to us with the duration of symptoms ranging from 2months to 5 years with patients having nasal tumours presenting earlier at 2months and 1year. Except for the patient with a lingual schwannoma, in our series no other patient had a externally visible swelling. Most of the symptoms are due to mass effect of the tumour.⁽²⁸⁻³⁰⁾ In our patients, the nasal tumours had presented with nasal block as the primary complaint while the patient with laryngeal tumour had dysphonia and the patient with lingual tumour had dysphagia, snoring, dysphonia. Enoz et al. and Romak et al. had also described similar symptoms in their analysis.^(10,31)

For evaluating the tumour, we preferred MRI for the evaluation of the laryngeal and lingual tumours as it is able to better identify the internal structure and the fat planes around the tumour and the possible nerve of origin. On MRI, the tumours were hypointense on T1 weighted images and heterogeneously hyperintense on T2 weighted images and showing intense post-gadolinium enhancement. Anil et al. had noted similar findings in their review.⁽³²⁾ No flow voids are usually seen. We had performed CT for the patients presenting with nasal tumours for visualizing the associated bony relations and to help during surgical dissection. In our series one tumour was homogenous and iso-dense in relation to the tongue and the other was heterogenous and hyper-dense in relation to the tongue. On contrast administration it was found that the tumour was heterogeneously but intensely enhancing. The variable hypo-, iso-, or hyper-attenuation and variable texture (heterogeneous or homogeneous) and enhancement on CT had previously been described by Silver et al. and Som et al.^(33,34) The heterogeneity which was noted on both CT and MRI can be attributed to cystic degeneration, xanthomatous change, or areas of relative hypocellularity adjacent to densely cellular or collagenous regions.⁽³⁵⁾ In one case the tumour was found to be extending into the sphenopalatine foramen. Similar extensions in nose and paranasal sinus schwannoma have been described by Hasegawa et al. and Hegazy et al. in addition to the possibility of absence of capsule and body erosions.^(20,36) Touati et al. had noted that invasion into the surrounding structures, loss of fat plane and presence of signal voids were suggestive of malignant transformation.⁽³⁷⁾ However, no such features were observed in our series.

Considering that the incidence of malignant schwannomas and the rate of malignant transformation of the benign schwannoma are not available, surgical excision is considered to be the safest treatment. However, due to the non-invasive nature of the tumour and the slow growth rate observational approach could also be considered. Based on the experience from their series, Shrikrishna et al, Valentino et al. and Yasumatsu et al. have advised that surgery should be considered after weighing the severity of preoperative symptomatology with the anticipated postoperative neurological deficit.^(4,38,39)

In our series we had to perform the surgery without opting for observation as these tumours were affecting normal activities of these patients. Of the two described techniques of complete excision and intracapsular enucleation we performed complete excisions considering the well-circumscribed and apparent benign nature of the tumours. We had encountered temporary paresis which resolved spontaneously at 1-month. This is in contrast to Liu et al. experienced nerve palsies in 100% and 67% of the cases respectively in the post-operative period.⁽³⁾ Colreavy et al. in their series too had reported palsy rate at 100% for complete excision and 31% for intracapsular enucleation.⁽²⁾ Electrical nerve stimulation has been advised for identifying the nerves and preservation their function by Ijichi et al.⁽⁴⁰⁾ We haven't noted a recurrence in any of our cases with 1year follow-up. For malignant schwannomas gross total resection is the best possible treatment. It plays an important role in prolonging patient survival.^(2,41) Adjuvant radiation can prevent local recurrence. Though adequate data is not available, adjuvant chemotherapy can also be considered.⁽⁴²⁾

CONCLUSION:

Schwannomas are benign slow growing tumours taking a varying duration to present based on the location of the tumour.

Observation is a possible method of management for these tumour and surgery is advisable only when it impedes normal functions.

Though they are usually well defined, these may have an irregular

appearance when present in the nasal cavity and other differentials should be kept in mind.

Author Contributions:

All authors contributed to patient data collection, manuscript drafting, literature review, and final approval.

Conflict Of Interest: None declared

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Ethical Approval:

The case series was conducted in accordance with the ethical standards of the institutional research committee. Informed consent for publication was obtained from all individual participants included in the study.

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