Original Article

Contemporary management of tonsillar schwannoma—a systematic review

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Background: Tonsillar schwannoma is a rare neoplastic disease representing one percent of all head and neck schwannomas. Current literature on tonsillar schwannoma consists mainly of case reports, with no clear guidelines for clinical workup and management. This systematic review summarises the clinical features, diagnostic workup and management of tonsillar schwannoma with the aim of providing guidance for the treatment of this rare disease.

Methods: A systematic review was conducted of all patients with palatine tonsil schwannoma in the available literature. A search of PubMed, Medline and Embase databases was conducted on 5th May 2023 to identify all published cases. Articles in a non-English language were excluded from the study. The final list of studies included were reviewed by both authors and assessed for bias using the Joanna Briggs Institute (JBI) checklist standardised tool for case reports. Data was reviewed and extracted from cases with a final diagnosis of palatine tonsil schwannoma in both adult and paediatric populations. Extracted data included: demographics, clinical presentation, imaging modality, histopathology, management and follow-up. Data was electronically collated and descriptive statistical analysis and qualitative review of the data was conducted using Microsoft Excel.

Results: A total of thirteen patients from thirteen cases studies were included in this systematic review. We found that this condition was present within a broad age range (eight to seventy-four years of age) and affected females (nine cases) more than males (four cases). There were two main reasons patients presented for review—progressive dysphagia and noticing a mass on intra-oral self-examination. Clinical examination findings were consistent among the case reports, with no evidence to suggest a malignant lesion. In all cases, patients underwent at least one form of imaging, which showed the lesion had features consistent with a schwannoma. Only three patients underwent a fine needle aspiration of the lesion, which produced a non-diagnostic result. Definitive management was unilateral tonsillectomy or tonsillotomy and there was no documented recurrence, although the follow-up period was highly variable.

Conclusions: In clinical practice, unilateral palatine tonsillar masses are treated as malignancy until proven otherwise, and thorough history and examination will determine the level of clinical suspicion. In all cases surgical excision was the definitive management for this condition, but there was high variability in investigations and follow-up among the reported cases. Our management algorithm proposes one form of imaging is sufficient and pre-operatively biopsy is not required. We also aim to standardise follow-up at six to eight weeks post procedure and then again in six to twelve months to rule out recurrence.

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Introduction

Neurogenic tumours make up a small percentage of neoplastic head and neck lesions and mainly comprise neurofibromas and schwannomas (also known as neurilemmomas) (1). Schwannomas are benign, encapsulated tumours of Schwann cells in peripheral nerves (2). Histopathological assessment of schwannomas identifies two classic cellular patterns: (I) Antoni A areas characterised by increased cellularity and spindle nuclei; and (II) Antoni B areas characterised by a loose arrangement of cells that are hypocellular and have variable macrophage infiltrate (3,4). Other features include Verocay bodies, hyalinised vessels with perivascular haemosiderin deposition, cystic spaces and degenerative changes (3).

The risk factors for the development of these lesions are prior radiation exposure and genetic predisposition (2), such as Von Recklinghausen's disease and type I neurofibromatosis (5).

The head and neck region is the most common site for schwannomas (25–48% of cases), but only about one percent occur intraorally (6). They mostly involve the acoustic nerve (7) and but can also occur in the trigeminal, facial and hypoglossal nerves (8). The first case of tonsillar schwannoma was described in the literature in 1975 (9) and since then there have only been twelve other cases reported worldwide in the English literature. Differential diagnoses for unilateral tonsillar hypertrophy include infectious (tonsillitis, peritonsillar abscess), inflammatory (Kawasaki disease, Kimura's disease) and neoplastic lesions, both benign (polyp, squamous papilloma, haemangioma) and malignant (squamous cell carcinoma, lymphoma) (10-12). Current literature on tonsillar schwannoma consists mainly of case reports, with no clear guidelines for workup and management of this rare disease. This systematic review summarises the clinical features, diagnostic workup and management, with the aim of providing guidance for the treatment of palatine tonsil schwannomas. We present this article in accordance with the PRISMA reporting checklist (available at https://www.theajo.com/article/view/10.21037/ajo-23-31/rc).

Methods

Study design

A systematic review of the literature was performed in relation to tonsillar schwannoma.

Search strategies

A review of the published literature was performed on 5th May 2023 (*Figure 1*). Embase, Medline and PubMed databases were searched for eligible studies. Four main search terms were combined with the Boolean operators "AND' and "OR". The keywords within the first two searches were "tonsil" OR "palatine tonsil". The second two searches were "schwannoma" OR "neurilemmoma". Both searches were combined with the Boolean operator "AND". All cases fulfilling the search criteria were reviewed by the author and relevant cases were compiled into a database. The study was not registered prior to completion and a review protocol was not prepared.

Study inclusion/exclusion criteria

Cases with a diagnosis of palatine tonsil schwannoma were included in the study. Both adult and paediatric cases were included. Cases that did not have a diagnosis of schwannoma of the palatine tonsil were excluded, for example schwannomas of other regions in the head and neck or oral cavity. Duplicate and non-English cases were excluded.

Study selection

Search strategies were implemented by author S.A.K. and results were collated into a Microsoft Excel spreadsheet. Duplicate references were removed via manual search. Remaining titles were screened for eligibility against inclusion and exclusion criteria. The bibliographies of the relevant studies were reviewed to identify additional relevant studies. The final list of articles included thirteen

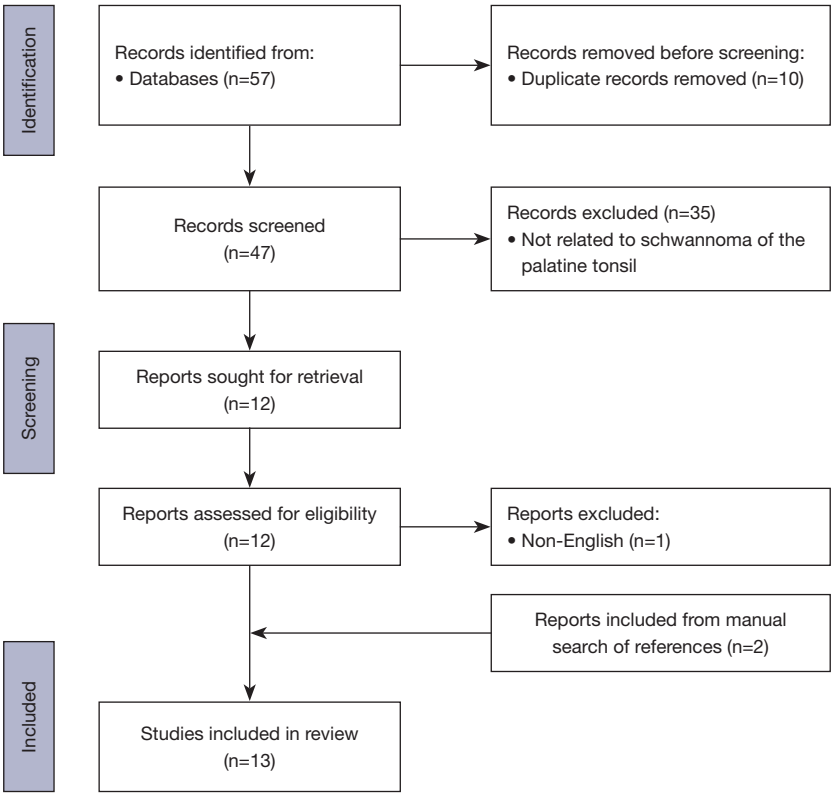

Figure 1 PRISMA 2020 flow diagram describing study selection process and identification of eligible cases.

case reports and was reviewed by C.M., senior author and fellowship trained Otolaryngologist.

Data extraction

Study data included year of publication, age and sex of patient, presenting symptoms, clinical examination findings, imaging modality and results, biopsy results, histopathology results, surgical technique and follow-up interval and outcome.

Quality/risk of bias

Risk of bias was assessed using the Joanna Briggs Institute (JBI) checklist standardised tool developed for case reports, which includes eight questions (13)—the detailed assessment of each study can be found in the supplementary appendix online.

Statistical analysis

Due to the small number of cases, a meta-analysis of the data was not possible. A descriptive statistical analysis and qualitative review of the data collected was conducted using Microsoft Excel.

Results

Patient demographics

The age range of patients presenting with tonsillar schwannoma is between eight to seventy-four years of age, with an average age of thirty-seven years. There were only three paediatric cases and it appears to affect females (nine cases) (6,7,14-20) more than males (four cases) (8,9,21,22) (*Table 1*).

Presenting symptoms

Most patients with tonsillar schwannoma present with either progressive dysphagia due to mass effect of the lesion, n=4 (6,8,17,18) or notice an enlarging unilateral mass on intraoral self-examination, n=6 (7,14-16,20,22). Other presenting symptoms include throat pain, n=2

Table 1 Summary of data from included studies on tonsillar schwannoma

Ref	Authors	Year	Age, year	Sex	Presenting complaint	Side	CL	Biopsy	Imaging	Surgical approach	Recurrence
(8)	Anil *et al.*	2005	38	M	DWS	Left	N	FNA	USS, CT	Tonsillectomy	Nil at 18 months
(14)	Bildirici *et al.*	2002	69	F	IFM	Right	N	FNA	MRI	Tonsillectomy	Nil at 12 months
(6)	Boujguenna *et al.*	2022	74	F	DWS	Left	N	Y	CT	Tonsillectomy	Unknown
(15)	Chaudhary	2011	42	F	IFM	Right	N	FNA	CT	Tonsillotomy	Nil at 12 months
(21)	Datta *et al.*	2020	42	M	Pain	Left	N	N	CT	Tonsillectomy	Nil at 1 month
(16)	Goto *et al.*	2012	44	F	IFM	Right	N	Y	CT	Tonsillectomy	Nil at 11 months
(22)	Joseph *et al.*	2010	24	M	IFM	Right	N	Y	CT	Tonsillotomy	Nil at 2 weeks
(17)	Lall *et al.*	1999	13	F	DWS	Left	N	N	Nil	Tonsillectomy	Nil recurrence
(7)	Lee *et al.*	2007	23	F	IFM	Left	N	N	CT	Tonsillectomy	Nil recurrence
(9)	Naik *et al.*	1975	45	M	Foreign body sensation	Right	N	N	Nil	Tonsillectomy	Unknown
(18)	Pham *et al.*	2013	8	F	DWS	Right	N	N	Nil	Tonsillotomy	Nil at 5 months
(19)	Piplani *et al.*	2011	14	F	Pain	Right	N	N	Nil	Bilateral tonsillectomy	Nil recurrence
(20)	Ruan *et al.*	2008	37	F	IFM	Right	N	N	CT	Tonsillectomy	Unknown

CL, cervical lymphadenopathy; M, male; F, female; DWS, difficulty with swallowing; IFM, initially found mass; N, no; FNA, fine needle aspiration; Y, yes; USS, ultrasound scan; CT, computed tomography; MRI, magnetic resonance imaging; Nil, no recurrence at specified follow-up.

(19,21) and sensation of a foreign body, n=1 (9). Associated symptoms included dysphonia, odynophagia, weight loss, referred pain to the ipsilateral ear, snoring and symptoms of obstructive sleep apnoea (OSA).

Clinical appearance

The general appearance of tonsillar schwannomas on clinical examination is a smooth, spherical, or ovaloid mass that projects into the pharyngeal cavity. It is non-tender and has a firm consistency. It is not fluctuant and there is no ulceration of the mucosa. The tumour is attached to the nerve of origin but remains mobile (17). These findings have been described as unilateral. One case reported bilateral abnormalities with one tonsil larger than the other, however schwannoma was only found in the larger tonsil (19). The lesions have been described as extending into the ipsilateral tongue base, nasopharynx, oropharynx and through the midline. Schwannoma was found in the right tonsil in eight cases (9,14-16,18-20,22) and in the left tonsil in five cases (6-8,17,21).

Cervical lymphadenopathy

There has been no documented cervical lymphadenopathy associated with tonsillar schwannomas.

Biopsy

Three patients had fine needle aspiration biopsies performed pre-operatively and the cytology for each specimen yielded a non-diagnostic result (8,14,15). In the case study by Joseph *et al*. [2010] (22), an incisional biopsy was performed in an outpatient clinic setting under local anaesthetic and yielded a histopathological result for a benign schwannoma.

Histopathology

Histopathological analysis of tonsillar schwannomas showed classical features—spindle cells arranged in Antoni A and Antoni B areas with Verocay bodies. Other features described include degenerative changes, such as perivascular hyalinisation and large dilated vessels. In this review, there was only one case of malignant tonsillar schwannoma

which showed "mitoses of six per ten fields" and local infiltration (6).

Imaging

The majority of cases utilised computed tomography (CT) and described a well circumscribed, heterogenous lesion in the area of the tonsil on plain CT (6-8,15,16,20-22). Contrast-enhanced CT showed a heterogeneously enhanced mass, without enhancement of the capsule (20). There was no infiltration or lymph node enlargement seen on any of the imaging performed.

Other imaging modalities used included transoral ultrasound and magnetic resonance imaging (MRI). On ultrasound the lesion appeared as a "hypovascular mass with or without central necrosis" (8). On MRI, tonsillar schwannomas appear as "well circumscribed, hypointense lesions on T1-weighted images, and hyperintense on T2-weighted images" (14).

Management

Ten out of the thirteen cases reported were treated with tonsillectomy of the abnormal tonsil only (6-9,14,16,17, 19-21). Three cases performed a wide local excision of the tumour only and did not remove the entire affected tonsil (15,18,22). One of these cases reported an incomplete resection of the schwannoma, however there was no evidence of lymphoma or malignancy in the excised tissue, therefore a re-excision was not performed (18). In one case the patient underwent a bilateral tonsillectomy as both tonsils looked abnormal upon clinical examination, however only the larger, right tonsil was found to have schwannoma (19). There were no intra-operative or post-operative complications reported in any of the cases published.

Functional deficits

There were no functional deficits reported immediately post-operatively or at follow-up.

Recurrence

Seven cases discussed follow-up at a specific post-operative interval, ranging between two weeks and eighteen months (8,14-16,18,21,22). Three cases did not specify a particular interval for post-operative follow-up (7,17,19) and three cases did not discuss follow-up at all (6,9,20). There was no recurrence of the schwannoma at follow-up. Patients were only followed-up once post-operatively and there was no regular surveillance as part of their management plan.

Discussion

Based on the management of the thirteen cases of tonsillar schwannoma worldwide we propose a management algorithm as outlined below (*Figure 2*). Presentation of a unilateral tonsillar mass is usually considered malignant until proven otherwise, thus a thorough history and examination will determine the level of clinical suspicion. Malignant transformation of schwannomas is exceedingly rare, however if present, has a poor prognosis, high rate of recurrence and metastases and is fatal (23,24).

Both imaging modalities—CT and MRI—provides useful information and helps to differentiate between invasive, malignant, and benign lesions, however neither are diagnostic for schwannomas. Features of schwannomas on CT in other areas of the body are consistent with the histopathological variations, for example Antoni A are represented as high density areas on CT whilst Antoni B, old haemorrhage and cystic changes are seen as low-density areas (20). This is consistent with the heterogenous appearance of tonsillar schwannomas on CT (25).

The imaging of choice is dependent on the resources available at the institution.

The accuracy of fine needle aspiration cytology in diagnosing other head and neck schwannomas is low, between 18% and 33% and is dependent on the specimen quality and the experience of the Cytopathologist (2). Incisional biopsies have an accuracy rate of 45.8%, however have the risks of: (I) insufficient sample; (II) exposing the patient to at least two procedures (i.e., first incision biopsy and then excision of the tumour); and (III) causing scar tissue to form at the surgical site, potentially making subsequent procedures more difficult (2). Biopsy of lesions yield unreliable results and exposes patients to two procedures and an increased level of associated risk. Therefore, pre-operative biopsy is not required in the workup of tonsillar schwannomas.

Ultimately, unilateral tonsillectomy is both a diagnostic and therapeutic procedure in this condition. All cases employed traditional tonsillectomy techniques to remove the schwannoma. However, transoral robotic surgery (TORS) is becoming a favourable method for removing tonsillar neoplasms due to better access of the pharynx for resection compared to traditional tonsillectomy and better

Figure 2 Proposed management algorithm. CT, computed tomography; MRI, magnetic resonance imaging.

post-operative outcomes compared to open approaches (26). In particular, radical tonsillectomy using TORS is an effective method for removing tonsillar carcinomas that have extended into surrounding areas or if there is limited transoral access and has reduced morbidity compared to open surgical approaches (27).

The surgical management of extracranial head and neck schwannomas in other locations is based on weighing the risks and benefits of surgery, pre-operative symptoms, and the anticipated severity of post-operative neurological deficits. Complete resection of the tumour results in palsy of the associated nerve, however with intracapsular enucleation only 31% of patient have a post-operative nerve palsy at six-month follow up—there is no difference in the recurrence rates (0% at two years) between the two methods (28). The risk of nerve palsy post-tonsillectomy is exceedingly rare, but may affect the glossopharyngeal nerve resulting in referred otalgia and transient dysgeusia (29). Tonsillectomy is the treatment of choice for other benign conditions, such as recurrent tonsillitis, sleep disordered breathing and OSA and therefore is a reasonable surgical approach for the treatment of tonsillar schwannoma.

There is no consensus on the post-operative follow-up interval in the reported cases, however there should be standard post-operative care and further follow-up at appropriate intervals to monitor for recurrence. We recommend standard post-operative follow-up at six to eight weeks and then re-review at six to twelve months to rule out recurrence of disease.

Limitations

The rarity of this disease is evident by the small number of cases reported in the literature since 1975. Thus, the evidence in this systematic review is comprised solely of case reports and hence the quality of evidence must be considered. The bias within included studies was low when assess with JBI checklist (see Appendix 1) as all the

data required to complete this review was available in the published literature. Publication bias cannot be determined as meta-analysis or effect estimate could not be completed due to the small study population. Selection bias was avoided as all the available published cases of palatine tonsillar schwannoma were included in this review.

Possible limitations of this review arose from the variability in post-operative follow-up, which ranged from two weeks to eighteen months. There was no follow-up beyond eighteen months, which can potentially underestimate disease recurrence. There was also variability in demographic data reported for each patient, for example some cases reported patient ethnicity, smoking status, personal or family history of tumours etc., whilst others did not. This could potentially limit the generalisability of the results from this review.

This study is the first systematic review on palatine tonsillar schwannomas and can provide Otolaryngologists with valuable insights on how to investigate and manage this rare disease.

Conclusions

Tonsillar schwannoma is a rare, benign condition found mostly in the adult population. It can cause significant symptoms such as throat pain, dysphagia and odynophagia and can be alarming to a patient and their primary care physician. However, it is a relatively easy condition to treat, once investigated appropriately, with complete excision leading to negligible recurrence risk. This systematic review on tonsillar schwannomas summarises all known literature and provides a management algorithm which can be applied in clinical situations.

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Footnote

Reporting Checklist: The authors have completed the PRISMA reporting checklist. Available at https://www.theajo.com/article/view/10.21037/ajo-23-31/rc

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Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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Supplementary

Appendix 1 Assessment for bias using Joanna Briggs institute critical appraisal checklist for case reports

Questions:

1. Were patient's demographic characteristics clearly described?
2. Was the patient's history clearly described and presented as a timeline?
3. Was the current clinical condition of the patient on presentation clearly described?
4. Were diagnostic tests or assessment methods and the results clearly described?
5. Was the intervention(s) or treatment procedure(s) clearly described?
6. Was the post-intervention clinical condition clearly described?
7. Were adverse events (harms) or unanticipated events identified and described?
8. Does the case report provide takeaway lessons?

Case Report	Questions:								
	1	2	3	4	5	6	7	8	JBI Score
Anil *et al.* 2005									8
Bildirici *et al.* 2002									8
Boujguenna *et al.* 2022									6
Chaudhary 2011									8
Datta *et al.* 2020									8
Goto *et al.* 2012									8
Joseph *et al.* 2010									8
Lall *et al.* 1999									8
Lee *et al.* 2007									8
Naik *et al.* 1975									6
Pham *et al.* 2013									8
Piplani *et al.* 2011									7
Ruan *et al.* 2008									7
KEY: GREEN: Yes (1 point); RED: No (0 point); JBI score: total out of 8.									

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