

Review Article

Schwannoma of the pinna: a narrative review

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ABSTRACT

Schwannomas are uncommon and benign neurogenic tumors that originate from the nerve sheath of the myelinated nerves. It can be found in any peripheral, cranial or autonomic nerves which contain Schwann cells. It is also called neuroma, nerve sheath tumor and neurilemmoma. Schwannomas in the head and neck region often present as painless swelling. The growth of schwannoma of the pinna causes an aesthetic alteration of the pinna. Schwannoma is a rare tumor of the pinna. The diagnosis of schwannoma is confirmed by histopathological study and immunohistochemistry. Histopathologically, it shows encapsulated, well-circumscribed lesions composed of different cellular patterns and arrangements. Histologically, the hallmark of schwannomas is the pattern of alternating Antoni A and Antoni B areas. The treatment of choice for schwannoma of the pinna is complete surgical excision. Recurrence following complete excision of the schwannoma is rare. Because of their rarity, these tumors are often not included in the differential diagnosis of the mass on the pinna. Preoperative suspicion and awareness are very important for schwannoma of the pinna to avoid delayed diagnosis and treatment. There is not much literature on the schwannoma of the pinna, indicating that this clinical entity is neglected. The objective of this article was to provide a comprehensive review of the schwannoma of the pinna including its epidemiology, etiopathology, clinical presentations, diagnosis and treatment.

Keywords: Schwannoma, Schwann cells, Nerve sheath tumor, Pinna

INTRODUCTION

Schwannomas are benign tumors that originate from the Schwann cells of the nerve sheath.¹ Schwannoma is an encapsulated tumor of the nerve sheath that arises from Schwann cells.¹ These tumors are neuroectodermal in origin that are responsible for providing myelin insulation to peripheral nervous system axon.² Schwannoma is a well-circumscribed mass that is attached to the peripheral nerves, cranial nerves or spinal nerve roots. Schwannomas can develop on any cranial nerves except the first and second whereas eight cranial nerves are the most commonly affected. Most of the schwannomas present as vestibular schwannoma.³ These tumors are rarely found in the external ears.⁴ There is difficulty in

obtaining a preoperative diagnosis, as symptoms are often nonspecific.⁵ The commonest clinical presentation of the schwannoma on the pinna is painless progressive swelling. Complete surgical excision is the treatment of choice. The postoperative prognosis is excellent. Schwannoma should be considered during the evaluation of any tumor of the pinna. If the tumor extends into the external ear, computed tomography (CT) scan and magnetic resonance imaging (MRI) are helpful to assess the extent of the lesion.⁶ Wide surgical excision is the treatment option. The schwannomas of the pinna are not widely discussed in the core surgical literature. This review article discusses the details of the epidemiology, etiopathology, clinical manifestations, diagnosis, treatment, and prognosis of the schwannoma of the pinna.

Methods of literature search

Multiple systematic methods were used to find current research publications on the schwannoma of the pinna. We started by searching the Scopus, Pub Med, Medline, and Google Scholar databases online. This search strategy recognized the abstracts of published publications, while other papers were discovered manually from the citations. A search strategy using PRISMA (preferred reporting items for systematic reviews and meta-analysis) guidelines was developed (Figure 1). Randomized controlled studies, observational studies, comparative studies, case series and case reports were evaluated for eligibility. There was a total number of articles 52 (26 case reports; 12 cases series; 14 original articles). This paper focused only on the schwannoma of the pinna. The search articles with schwannomas other than pinna were excluded in this review article. Review articles with no primary research data were also excluded. This paper examined the epidemiology, etiopathogenesis, clinical manifestations, diagnosis, treatment and prognosis of schwannoma of the pinna. This analysis provided a foundation for future prospective trials for schwannomas of the pinna. It also catalysed additional study of schwannomas of the pinna, allowing early detection and treatment.

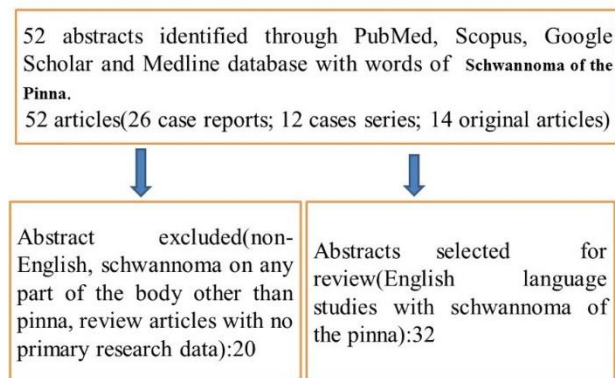


Figure 1: Flow chart showing method of literature search.

Epidemiology

Schwannoma was first documented by Verocay in 1910, who used the term “neuriloma” to describe the benign neurogenic tumor.⁷ The word schwannoma was coined by Del Rio-Hortega in 1942 to indicate this neoplasm from the Schwann cells.⁸ In the outer ear, schwannoma commonly affects the external auditory canal, while pinna and tympanic membrane are rare localization of schwannomas.⁹ Schwannoma was first reported in the pinna by Foder et al in 1977.¹⁰ It is relatively uncommon and accounts for only 5% of all tumors of soft tissues.⁹ About 25 to 45% of the extra-cranial schwannoma originates from nerves of the head and neck region.¹¹

Etiopathology

The exact etiology of schwannomas is not known. Any peripheral, cranial (other than olfactory and optic) or autonomic neuron containing Schwann cells, the cells that make the myelin coating around the nerve fibers, can cause schwannoma.¹² The Schwann cells encapsulate nerve fibers and help fasten the propagation of nerve impulses.¹³ Neurofibromas also originate from Schwann cells but do not have a true capsule and histologically have no Verocay bodies.¹⁴ In the cutaneous region, the histopathological differential diagnosis is a solitary circumscribed neuroma that is partially encapsulated, has ill-defined nuclear palisades and in contrast to the schwannomas, contains no Antoni B areas.¹⁵ The roots of the spinal nerves, the flexor surfaces of the upper and lower limbs, and the tongue are common places where the schwannomas may be found.¹⁶ These tumors can develop alone or in association with neurofibromatosis type 1 (NF1) or type 2 (NF2) as well as schwannomatosis, a commonly inherited lesion. The NF2 gene mediates the Schwann cells and functions as a tumor suppressor.¹⁷ Pinna is innervated by the greater auricular nerve, lesser occipital nerve, auriculotemporal nerve, and partly by VIIth and Xth cranial nerves.¹⁸ So schwannoma can originate from any nerves of the pinna. Schwannoma is often found near the Darwin tubercle of the pinna in the territory of the greater auricular nerve.¹⁹ It is often difficult to determine the nerve of origin of schwannoma of the pinna because of its complex ramification. These tumors usually do not invade the adjacent soft tissues or bone but manifest different symptoms and bone destruction by local pressure effects. The schwannoma is sometimes found in the middle ear usually arising from the facial nerve and chorda tympani nerve.²⁰

Clinical presentations

The mean age of onset of schwannoma is 30 to 60 years, and these are more common in females, although gender predilection is still controversial.²¹ The most common clinical presentation of the schwannoma of the pinna is painless swelling on the pinna with slow progression (Figure 2). It often grows gradually with an increase in size. The consistency of the schwannoma in the external ear is often hard in consistency with the smooth surface covered by normal skin.²¹ The common sites of pinna affected with schwannoma are concha, helix, antihelix, the root of the helix, and auricular tubercle (Table 1).²² These features may produce a few differential diagnoses from some soft tissue tumors like sebaceous adenoma, eosinophilic granuloma, fibroma, chondroma and leiomyoma.²³ Auricular schwannomas are often asymptomatic but pain and paresthesia may be found in about one-third of cases as cutaneous manifestations.²⁴ In pinna, it usually shows as slow growing mass with esthetic deformity. The tumor is often non-tender and soft/soft-firm in consistency on palpation.²⁵ When the schwannoma of the pinna grows sufficiently to block the

external auditory canal, the patient may present with conductive hearing loss or otitis externa due to blockage and accumulation of skin debris such as wax or otorrhea, but rarely reported bony erosion.²⁶ In the case of schwannoma of the external auditory canal presents with hearing impairment, tinnitus, vertigo, aural fullness, recurrent otitis externa, and autophony.²⁷ The preoperative diagnosis is often difficult as most schwannomas do not show neurological-related symptoms.²⁸ Schwannoma may mimic certain diseases such as tumor metastasis or infection.²⁹ The differential diagnosis of the schwannoma of the pinna includes cystic mass (sebaceous cyst, epidermoid), chondroma, fibroma, and neurofibroma.³⁰ Clinicians should keep in mind that any mass on the pinna may be of neurogenic origin although its incidence is low.



Figure 2: Right pinna showing schwannoma near the concha region (yellow arrow).

Table 1: Previously reported schwannoma of the pinna with exact location of the lesion.²²

S. no.	Year	Author	Exact location of schwannoma
1.	2018	Testa et al	Root of the helix
2.	2013	Kuldeep et al	Auricular tubercle
3.	2012	Dong et al	Concha
4.	2010	Lai et al	Concha
5.	2008	Carter et al	Antihelix
6.	2008	Zhonghua et al	Helix
7.	2001	Galli et al	Concha and external auditory meatus

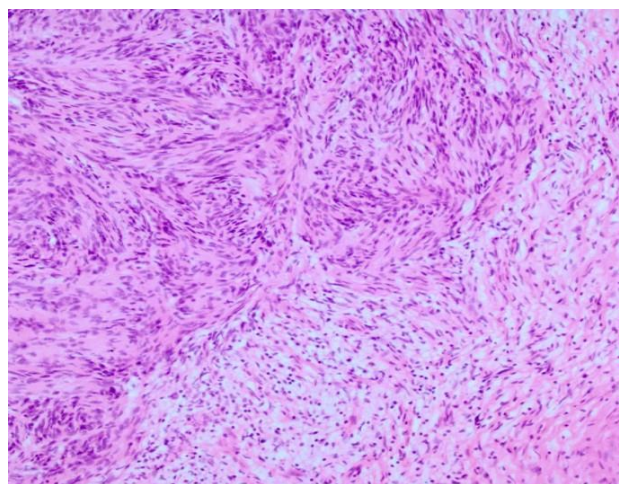


Figure 3: Schwannoma of the pinna showing spindle cells arranged in palisading pattern and forming Verocay bodies (Antoni-A area) (H&E stain, 100×).

Diagnosis

Schwannoma on the pinna is an uncommon clinical condition that can pose a diagnostic dilemma to clinicians. The preoperative diagnosis of schwannoma is difficult and should be suggested by clinical presentations and supported by investigations. The auricular schwannoma is a rare clinical entity and often poses a diagnostic dilemma to clinicians. The use of fine needle aspiration cytology (FNAC) for the diagnosis of schwannoma is controversial and the majority of authors do not advise it for this tumor.³¹ FNAC is usually considered inconclusive in the case of schwannoma. Clinicians should emphasize the biopsy for diagnosis of schwannoma. The diagnosis of schwannoma is confirmed by histopathological study and immunohistochemistry (IHC). The histopathological examination is mandatory for confirming the diagnosis of the schwannoma. The excised mass of the schwannoma is usually oval, smooth surfaced, well encapsulated pinkish white on cut section. The cut section of the schwannoma is firm, uniform parenchymatous consistency and tan to grey, and often reveals focal hemorrhage, necrosis, cystic degeneration, and xanthomatous changes as it increases in size. Schwannoma is classified into seven subtypes such as conventional, cellular, plexiform, melanotic, cranial nerve, ancient and granular cell schwannomas.³² However, there are four important histological types such as conventional, plexiform, cellular, and ancient.³³ All the types mainly have two distinct histological patterns: Antoni A and B. The histopathological study reveals a predominance of Antoni A areas with spindle-shaped cells arranged in a palisading pattern and central acellular areas representing Verocay bodies (Figure 3). Some areas also reveal a hypocellular and less organized arrangement, as seen in the Antoni B type. An IHC study can be helpful for the diagnosis but is more useful to rule out close differentials.³³ The IHC examination with S-100 protein shows intense positivity in the cells of the

peripheral nerve sheath tumors. S-100 is a highly sensitive and most commonly used immunostain for dealing with suspected peripheral nerve tumors. The nerve sheath tumors contain numerous immunoreactive S-100 positive cells.³⁴ Both schwannomas and neurofibromas show moderate to strong reactivity to S-100. However, S-100 shows low specificity toward the diagnosis of peripheral nerve cell tumor. The schwannoma also shows the positive expression of SRY-related HMG box 10 (SOX-10) protein. Both schwannomas and neurofibromas show moderate/strong reactivity to S-100. However, S-100 has low specificity for the diagnosis of peripheral nerve cell tumors. SOX-10 staining is also more sensitive and specific than S-100 for peripheral nerve tumors.³⁵ Calretinin has high specificity for schwannoma and is useful to differentiate it from neurofibroma.³⁶ Intensive staining with CD-57 is seen in traumatic neuromas.³⁷ Schwannomas are also stained positive with glial fibrillary acidic protein (GFAP), Leu-7 antigen, and vimentin.³⁸ A positive neurofilament protein (NFP) staining of the axons differentiates a palisaded encapsulated neuroma from schwannoma.³⁹ CT scans or MRI are helpful for diagnosis and formulating a treatment approach, enabling the characteristics of the lesion and its association with adjacent structures.⁴⁰

Treatment

Surgical excision is the treatment of choice, but the slow growth and noninvasive nature of the schwannomas need an observational approach. The decision of the surgery should be based on the balance between the risk and benefits of the surgery, that is the severity of preoperative symptoms and anticipated postoperative neurological problems/deficits. Schwannoma of the pinna usually presents as slow growing, well-demarcated ovoid mass with a true capsule which improves dissection during surgery.⁴¹ The surgical excisions of the pinna schwannoma are operated with a traditional approach, separating the tumor from the underlying perichondrium.⁴² It has a true capsule forming a smooth surface under the skin which facilitates dissection during surgery.⁴³ The mass should be completely excised and often done under local anesthesia in adult patients with preservation of the overlying skin. Small schwannoma on the pinna required CO₂ laser treatment for a successful outcome with little or no morbidity. The use of CO₂ laser allows for removal of the tumor easily, reduces bleeding and surgical time, and avoids sutures and so unsightly scars on the face.⁹ Open surgical excision is helpful for large tumors. In the traditional approach, the neoplasm is separated from the underlying perichondrium.²² Schwannomas are resistant to radiotherapy and as these are well encapsulated, the treatment of choice is conservative surgical enucleation with periodic follow up.^{44,45} The choice of surgery is mainly decided by the relationship between the tumor and the nerve of origin. The conventional extracapsular excision may damage the normal fascicles at the time of dissection of the capsule. Intracapsular excision with gentle dissection between the

capsule of the tumor and normal fascicles reduces the chance of nerve damage.⁴⁶ The epineural layer covering the schwannoma capsule should be dissected in a pattern similar to peeling an onion to allow for safe removal of the tumor, which should be approached by its proximal and distal pole. The fascicles found in the tumor are often nonfunctional, so excision of these fascicles does not result in neurological problems.⁴⁷ The use of a microscope for surgery and micro-neurosurgical technique has modified the result of conventional extracapsular excision with minimal morbidity.

Prognosis

The prognosis following complete excision of the pinna schwannoma is excellent.⁴⁸ Schwannomas are isolated benign tumors and have a good prognosis with a low risk of malignant transformation. But the primary malignant schwannomas have been documented, about 15% of which are found in the head and neck region.⁴⁹ One study showed the use of CO₂ laser for auricular schwannoma showed no complications and no relapse at five years follow-up period.⁹ These malignant peripheral nerve sheath tumors constitute 5% of all malignant soft tissue tumors and 15% of these tumors found in the head and neck area.⁵⁰ The prognosis after the complete removal of pinna schwannoma is excellent. Post-operative recurrence and malignant transformation of pinna schwannoma are not reported in the literature. However, malignant peripheral nerve sheath tumors are seen in the head and neck region, most commonly in the infratemporal fossa and parotid gland.

CONCLUSION

Schwannoma of the pinna is exceedingly rare. Despite the rarity of the schwannoma in the pinna, this neurogenic tumor of the pinna should be recognized. The most common symptom is a slowly enlarging mass without tenderness. Although there are several imaging options available, the exact diagnosis of schwannoma is done by histopathological study and immunohistochemistry. It can be included in the differential diagnosis for the mass on the pinna. The histopathological study and immunohistochemical investigation are useful for the final diagnosis of schwannoma. The treatment of choice is complete surgical excision. These tumors typically do not recur. An accurate preoperative diagnosis with proper identification of the nerve of origin is often a challenge for a clinician. Sometimes, before a surgical procedure, clinicians should explain details of differential diagnosis, treatment options, and possible nerve damage to the patient.

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