

Schwannoma of the Submandibular Gland

Dechu Muddaiah,¹ Srinivas Venkatarangaiah,¹ Prashanth V,¹ Mohammed Adil Hussain M¹

ABSTRACT

Introduction

Mesenchymal tumours of submandibular gland are extremely rare. Schwannoma of the salivary glands is a particularly rare form of an extracranial neurogenic tumour.

Case Report

Here, we present an unusual case of schwannoma of submandibular gland in a 16 year old girl, who underwent total excision of mass with submandibular gland excision with no cranial nerve deficits. The details of the histopathologic features are present.

Discussion

Schwannoma of the salivary gland is a particularly rare form of an extracranial neurogenic tumour. Our case indicates good prognosis in a case of submandibular gland schwannoma after surgical excision.

Keywords

Submandibular Gland; Neurilemmoma; Schwannoma

Schwannomas are benign, solitary and well differentiated neurogenic tumour derived from Schwann cell of the neural sheath.¹ Schwannoma is not a common tumour of the salivary gland. Nearly 25-45% of all schwannomas occur in the head and neck area. Schwannomas are usually, unilocular, cystic, symptomless, slow growing, benign, solitary, encapsulated tumours that are attached or surrounded by a nerve. They appeared to push the nerve axions and can often be dissected free, with preservation of the nerve of origin. Histologically, classic schwannomas are characterised by two distinct patterns showing cellular areas of spindle cells with nuclear palisading (Antoni A) and spindle cells scattered in a delicate, fibrillar microcystic matrix (Antoni B).² The histological forms of schwannoma are ancient, cellular, epitheloid, hybrid, melanotic, plexiform and psammomatous variants.³

Schwannoma also known as neurilemmoma was first described by Verocay et al.⁴ In the present report we describe a case of Schwannoma of the left submandibular gland with the image and histological findings.

Case Report

A 16 year old girl was admitted to our hospital with a painless swelling on the left side of the neck for past two months. A thorough physical examination of neck revealed a smooth surface, mobile, firm and painless mass of approximately 5cms in its greatest diameter in left submandibular region, there was no cervical lymphadenopathy. All the cranial nerve examination were normal and ultrasound examination of neck revealed a well circumscribed and heterogenous mass. Pre operative fine needle cytology of the mass was suggestive of chronic sialadenitis. CECT of the neck was done which was suggestive of the encapsulated well circumscribed spherical mass lesion 4.5x3x3 cm mass with heterogenous opacity in the left submandibular region. The mass was located lateral to genioglossus muscle and the submandibular gland was compressed and inferiorly displaced. Contrast enhancement demonstrated weak enhancement with no evidence of cystic or necrotic

1 - Department of ENT, BGS Global Institute of Medical Sciences, Rajiv Gandhi University of Health Sciences, Bengaluru

Corresponding author:

Dr Dechu Muddaiah
email: dechum23@gmail.com

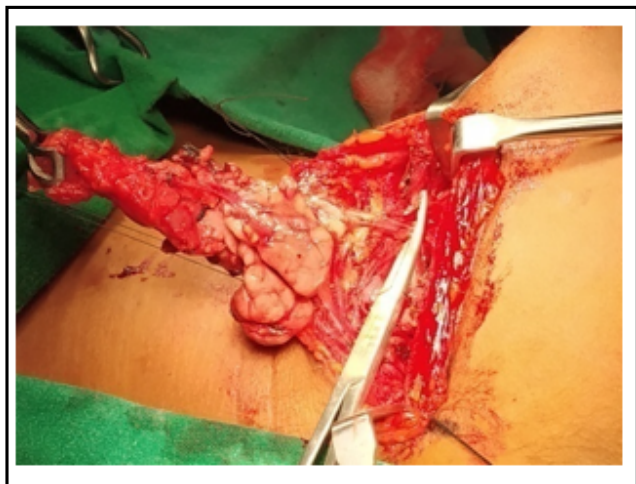


Fig.1. Intra operative picture showing tumour mass attached to left submandibular gland.

degeneration. After the initial evaluation the patient was prepared for surgical evaluation and resection with a presumptive diagnosis of benign tumour of left submandibular gland. The tumour mass was abutting the left submandibular gland and was not attached to any nerves. (Fig. 1) The mass was carefully dissected from the adjacent structures. The lesion was excised completely with the submandibular gland.

Macroscopically the resected mass was encapsulated yellowish in colour; measuring 6 cms. It was oval smooth, firm, fleshy. (Fig. 2) Patient had an uneventful postoperative recovery. Total excision resulted in complete recovery of symptoms with no cranial nerve deficits. Histopathological examination revealed a well encapsulated tumour with alternating hypercellular Antoni A and hypocellular Antoni B areas. (Fig.3) Antoni A areas composed of spindle cells with twisted nuclei and indistinct cytoplasmic borders. Many Verocay bodies formed by two compact rows of well aligned nuclei separated by fibrillary cell process. (Fig.4) Antoni B areas are hypocellular with loose fibromyxoid change.

Discussion

Neurogenic tumours arise from the neural crest which differentiates into the Schwann cells and sympatheticoblasts.⁵ The Schwann cells give rise to

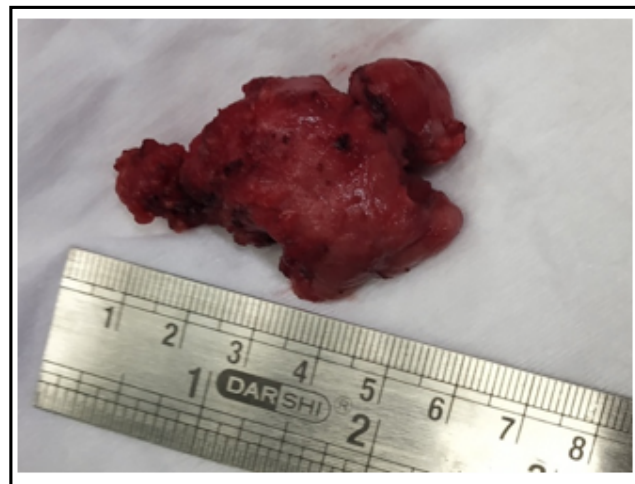


Fig. 2. Excised specimen tumour attached to submandibular gland.

neurofibroma and neurilemmoma (Schwannoma).⁶ Schwannoma is a slow growing solitary and encapsulated tumour attached to a nerve. Schwannoma may arise from any cranial nerve or spinal nerve that has a sheath. Schwannomas occur in the head and neck. The common sites of extracranial neurilemmoma in head and neck are parapharyngeal space, submandibular space, paranasal sinuses, cheek and oral cavity.⁷ Batsakis reported that in the neck the schwannomas can be divided into medial and lateral group, on the basis of nerve of origin. The medial group arises from last four cranial nerves and the cervical sympathetic chain, the lateral group arises from cervical trunk.⁸

Mesenchymal neoplasms are rare in the salivary glands, representing only 2-5% of salivary gland tumours.² Schwannoma in particular are rare in the submandibular gland. After Attie et al,⁹ described a case of Schwannoma arising from lingual nerve and Sutay et al¹⁰ reported a Schwannoma originating from hypoglossal nerve. Sato et al¹¹ published first case of parasympathetic schwannoma of submandibular gland.

A salivary gland tumour of mesenchymal origin usually presents as an asymptomatic, slow growing, well circumscribed mass in the submandibular region. Radiological investigations like contrast enhanced computer tomography is required and MRI may be necessary in a few cases. Our patient underwent preop CECT which revealed well circumscribed spherical

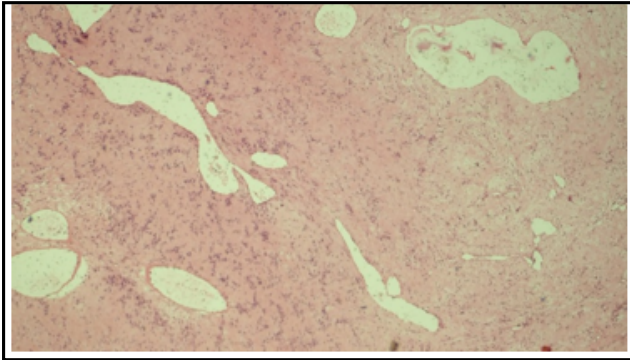


Fig.3. Histopathology of schwannoma showing Antoni A and Antoni B cells (H&E, 4X)

mass with mixed attenuation in the left submandibular region, the mass was lateral to the hyoglossus muscle and the submandibular gland was compressed and inferiorly displaced. There was no evidence of cystic or necrotic degeneration. MRI was not advised as it is not a routine diagnostic aid however, MRI with gadolinium is the better choice because it can diagnose even small intra glandular nerve cell tumour. In head and neck schwannomas fine needle aspiration biopsy is of questionable value, as it may show unclear histopathological results from these cases, which have the potential to confuse this lesion with a more serious one such as sarcoma.¹² Even in the present case FNAC was suggestive of chronic sialadenitis.

The size of Schwannoma arising in the submandibular glands range from 1 to 6 cm in diameter, in our case measured approximately 4.5x3x3 cm. The swelling is most often freely mobile and may be fusiform in shape, but when it is connected to a large nerve or nerve trunk there may be restriction of movements.

Schwannoma shows characteristic histological appearance, dominated by an encapsulated lesion arising from a nerve composed of an intimate mixture of spindle cells forming highly cellular so called Antoni type A areas and less cellular, myxoid Antoni type B. Between palisades of Antoni type A cells are regions that are devoid of nuclei termed verocay bodies, after the person who described schwannomas. The present case contained a mixture of Antoni type A and Antoni type B, Antoni type A being the predominant microscopic pattern. The Antoni A and Antoni B are used for pathological

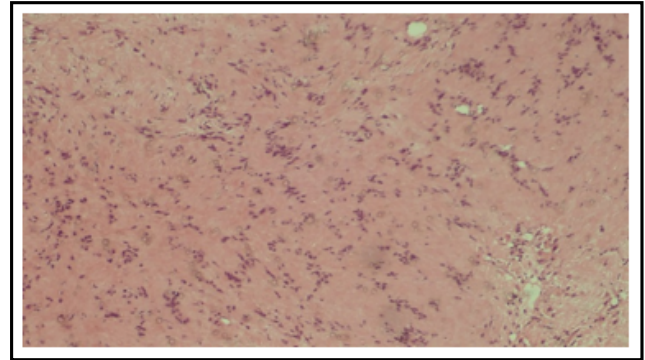


Fig.4. Antoni A areas with nuclear palisading and verocay bodies (H&E, 10X)

determination of the tumour and have no clinical significance. Even though the histological features of Schwannoma are quite characteristic and can be diagnosed light microscopically, in certain problematic cases, immunoperoxidase and ultrastructural studies may be needed. Immunohistochemical evaluation of schwannomas for S100 protein provides consistently positive results, especially in Antoni type A areas.

The treatment of choice for schwannomas are complete surgical excision with preservation of nerve function. Recurrence is uncommon. In our case the patient underwent submandibular gland excision with excision of tumour mass. The patient is on follow up for last 1 year.

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