

Schwannoma of the Submandibular Gland: A Rare Case Report

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ABSTRACT

Schwannoma is a slow growing, benign, encapsulated tumour of Schwann cells, which form myelin in peripheral nerves. It is the most common tumour of peripheral nerve, also known as neurilemoma or neurimoma. Schwannomas of head and neck area is common and may arise from peripheral, central or autonomic nerve but Salivary gland schwannomas are rare extracranial forms. We report a rare case of schwannoma of submandibular gland which presented with painless swelling in submandibular region, treated by total excision of submandibular gland. There was no postoperative nerve deficit or recurrence within 9 months of follow up.

Keywords: Antoni, Neurilemoma/Neurimoma, Tumour

CASE REPORT

A 21-year-old male presented to the Department of General Surgery of Rajendra Institute of Medical Sciences Ranchi with painless swelling in right submandibular region since 3 months. Swelling was insidious in onset and slowly progressive in nature. On physical examination of neck, a single swelling of approximate size 5x4 cm noted in right submandibular region. Surface was smooth, well defined lower margin but poorly defined upper margin, firm in consistency, non tender and slightly mobile. There was no palpable lymphadenopathy and all cranial nerves were within normal limit. There was no history of fever, sore throat or local trauma. Clinical diagnosis of pleomorphic adenoma was made. Ultrasonography neck showed heterogeneous echotexture of submandibular gland with hypoechoic well circumscribed encapsulated mass. Fine needle cytology showed cluster of spindle shaped cells with moderate cellularity, contradict the clinical diagnosis, suggestive of schwannoma of submandibular gland and advised excision biopsy for confirmation. We prepared the patient for surgery under general anaesthesia after taking consent for excision of submandibular gland along with swelling. On exploration of submandibular region a swelling attached to the submandibular gland was found [Table/

Fig-1]. After careful dissection and identification of adjacent anatomical structures total excision of submandibular gland along with swelling done [Table/Fig-2]. Wound healing was normal and no specific medication was used in postoperative period, so postoperative was uneventful. Pathological report showed elongated spindle shaped cells with antoni type A and type B areas [Table/Fig-3]. Immuno-staining consistent with schwannoma with strongly positive for s-100 [Table/Fig-4]. Score was 4+ (immunoreactive in 76-100% cells). These findings confirm the diagnosis and there was no recurrence within 9 months of follow up [Table/Fig-5].

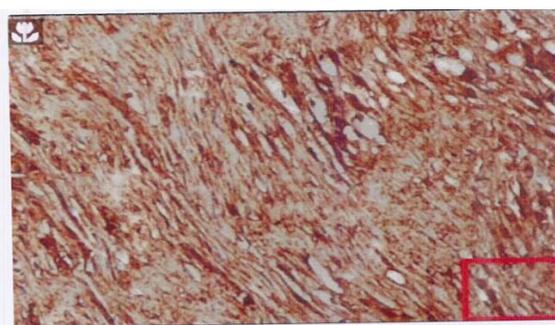
DISCUSSION

Solitary benign schwannomas are common, and association of neurilemmas with von Recklinghausen's disease is well-known, nearly 25-45% of those occurs in head and neck regions [1]. Nerve of origin may be either peripheral, central or autonomic but in about 10 to 40% cases nerve of origin is not identified [2]. These are slow growing tumours rarely shows rapid course with slight female predominance and usually present between second to fourth decades of life [3].

First described by verocay in 1908, these tumours often mimic primary or metastatic disease in the head and neck [4]. Schwannoma of the salivary gland is a particularly rare form of an extracranial neurogenic tumour, with most presenting in the parotid gland originating from a peripheral branch of the facial nerve [5]. Present case is unusual because of involvement of submandibular gland. In the head and neck, schwannomas usually arise from the sensory divisions of cranial nerves, most commonly the vestibular nerve and the vagal nerve [6].



[Table/Fig-1]: Swelling attached to the Submandibular gland (Operative finding).
[Table/Fig-2]: Specimen obtained showing well encapsulated mass.



[Table/Fig-3]: Histopathological report showing elongated spindle shaped cells with antoni type A and type B areas. **[Table/Fig-4]:** Immuno-staining, strongly positive for S-100 (Score 4+). **[Table/Fig-5]:** Postsurgical scar, with no recurrence within 9 months of follow up.



Most common symptom is painless slow growing solitary mass, shows slight female predominance, may be multiple as a part of neurofibromatosis 1 [3]. Pain and malignant transformation is rare.

Pleomorphic adenoma is the most common benign swelling in the submandibular region. Other common benign swellings are inflammatory swelling of the gland and lymph nodes. History and clinical examination of the pleomorphic adenoma may be as similar as schwannoma, so clinical diagnosis is almost impossible. According to report published by Biswas et al., over 10 year experience on schwannomas only 6% of schwannomas is diagnosed by clinical, radiological and fine needle cytology preoperatively [7]. Although MRI is radiological investigation of choice, it may be indistinguishable from Pleomorphic adenomas. So biopsy is diagnostic. Microscopically, two types of tissue distributed randomly in schwannoma. Schwannomas contains both Antoni A type tissue with interwoven bundles of long, bipolar, spindle cells, and Antoni B type tissue with its loose texture. Nuclear palisading and Verocoy bodies in some areas were also typical [8].

Present case is clinically comparable to case reported by Gaffar Aslan et al., both the report highlighting benign, slow growing, non recurrent and good prognostic nature of the disease [9]. Nerve of origin is not defined in present case. So we believe this tumour arise from autonomic nervous system.

Because of chemo and radio -resistance nature of tumour total excision is treatment of choice [10]. Recurrence and nerve deficit after surgical excision is also reported due to iatrogenic nerve injury. In present case there was no nerve deficit or recurrence within 9 months of follow up.

CONCLUSION

Although rare, Schwannoma should be kept in mind in any submandibular gland swelling as total excision provides cure. As in present case with total excision of mass with submandibular gland, there was no nerve deficit or recurrence within 9 months of follow up.

REFERENCES

- [1] Kang GC, Soo KC, Lim DT. Extracranial non-vestibular head and neck schwannomas: a ten-year experience. *Ann Acad Med.* 2007;36:233-38.
- [2] Gallo WJ, Moss M, Shapiro DN, Gaul JV. Neurilemmoma: review of the literature and report of five cases. *J Oral Surg.* 1997;35:235-36. [PubMed]
- [3] Dhupar A, Yadav S, Dhupar V. Schwannoma of the hard plate: a rare case. *Internet J Head Neck Surg.* 2011;4.
- [4] Marx RE, Stern D. Benign soft tissue tumours of mesenchymal origin. In *Oral and maxillofacial pathology: a rationale for diagnosis and treatment. Quintessence Publishing Co Inc;* 2003:395-461.
- [5] Diaz DD, Kennedy KS, Parker GS, White VJ. Schwannoma of the submandibular gland. *Head Neck.* 1991;13(3):239-42.
- [6] Kawakami R, Kaneko T, Kadoya M, Matsushita T, Fujinaga Y, Oguchi K, et al. Schwannoma in the sublingual space. *Dentomaxillofac Radiol.* 2004;33:259-61.
- [7] Biswas D, Marnane CN, Mal R, Baldwin D. Extracranial head and neck schwannomas - a 10-year review. *Auris Nasus Larynx.* 2007;34:353-59. PubMed Abstract | Publisher Full Text.
- [8] Drevelengas A, Kalaitzoglou I, Lazaridis N. Sublingual hypoglossal neurilemmoma. Case report. *Aust Dent J.* 1998;43:311-14.
- [9] Aslan G, Cinar F, Cabuk FK. Schwannoma of the submandibular gland: a case report. *Journal of Medical Case Reports.* 2014;8:231. doi:10.1186/1752-1947-8-231.
- [10] Langner E, Del Negro A, Akashi HK, Araujo PP, Tincani AJ, Martins AS. Schwannoma in the head and neck: retrospective analysis of 21 patients and review of the literature. *Sao Paulo Med J.* 2007;125:220-22. PubMed Abstract | Publisher Full Text.

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Date of Submission: **Aug 08, 2015**

Date of Peer Review: **Oct 03, 2015**

Date of Acceptance: **Dec 03, 2015**

Date of Publishing: **Feb 01, 2016**

FINANCIAL OR OTHER COMPETING INTERESTS: None.