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## Case Report

# Schwannoma (Neurilemmoma) of the Submandibular Gland

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### ABSTRACT

Schwannomas (neurilemmomas) are infrequent, benign mesenchymal neoplasms that pose considerable preoperative diagnostic difficulties. Schwannomas of the submaxillary gland are even less common, with only few cases reported in the literature to date. The present study describes a male with a right submaxillary tumor for the past 9 years; the histopathological study diagnosed schwannoma - no preoperative evaluation having been able to establish the diagnosis.

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### Introduction

Schwannomas (neurilemmomas) are benign neoplasms derived from the Schwann cells, with a predilection for the head, neck and flexor surfaces of the upper and lower extremities [1]. Twenty-five to 45% of all schwannomas occur in the head and neck region in relation to the different cranial nerves, the cervical sympathetic chain or the brachial plexus [2, 3]. However, 50-72.5% of the lesions located in this region are of unknown origin. Isolated schwannomas of the major salivary glands are extremely rare: in the AFIP series, neurilemmomas accounted for 16% of all benign non-epithelial tumors of the salivary glands [4-6]. In turn, 83% of these lesions were located in the parotid gland, and 17% in the submaxillary gland [7]. Schwannomas are asymptomatic, slow-growing tumors without associated neurological manifestations that are not usually diagnosed before surgery [1, 2, 8]. The present study describes an unusual case of submandibular schwannoma and conducts a review of the literature on the subject.

### Case Report

A 52-year-old male without antecedents of interest or toxic habits presented with a painless tumor for the past 9 years. Combined oral and cervical exploration revealed a firm, stony hard mobile growth in the right submandibular region, measuring about 3 cm in major diameter.

The patient referred no symptoms compatible with prior episodes of submaxillitis, and no neurological manifestations. Salivary secretion from the duct of Wharton was normal in both amount and appearance, with no evidence of calculi upon palpation. There were no neck adenopathies or primary oropharyngeal process to account for the presence of the tumor. The overlying skin showed no signs of inflammation. Fine needle aspiration cytology and a computed tomography study were requested.

The tentative clinical diagnosis centered on two possibilities: a pleomorphic adenoma of the submaxillary gland or a Küttner tumor. The fine needle aspiration biopsy discarded a malignant process but was unable to establish a diagnosis. Computed tomography in turn showed a dense tumor measuring approximately 4 cm in diameter in relation to the deepest portion of the submaxillary gland (Figure 1). Tuberculin and blood tests afforded no significant data. The mass was removed under general anesthesia; during the operation a hard and well encapsulated, grayish tumor (Figure 2) was observed located anterior to the posterior belly of the digastric muscle, in relation to the anterior portion of the gland. The lingual and greater hypoglossal nerves were dissected and found to be fully independent of the tumor. The definitive histological diagnosis was schwannoma of the submaxillary gland. There were no postoperative neurological alterations, and no tumor recurrence one year after surgery.

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**Figure 1:** Axial CT scan demonstrating a defined mass that appears in the deep portion of the right submaxillary gland.



2A



2B

**Figures 2A & 2B:** Gross specimen showing the mass with the typical macroscopical aspect of schwannomas.

## Discussion

Neurilemmoma, first described by Verocay in 1910, is a tumor derived from the Schwann sheath of the peripheral, motor, sensory and sympathetic and cranial nerves (with the exception of the I and II cranial nerves) [1, 2, 8]. These tumors may develop at any age but are most frequent between the third and fourth decades of life [1, 2, 8, 9]. Some authors have reported an increased incidence in females, though others consider both sexes to be equally affected [1, 2, 7-9]. In turn, some studies have reported a male predilection [10]. Between 25% and 45% of all schwannomas are located in the head and neck region. In this sense, cases have been reported in relation to the X, XII and VII cranial nerves, though most tumors in this region are associated with the cervical plexus or are of unknown origin. Kun *et al.*, in a series of 49 cases divided into two groups (neck and oral and maxillofacial tumors) found the lesions

to preferentially distribute in the oral cavity (tongue, cheek, mental zone, palate, lip), and in the zone of the carotid triangle [9].

Despite the tendency of these tumors to affect the head and neck region, the preoperative diagnosis of schwannoma poses considerable difficulties: only one case of the 12 reported by Williams *et al.* [10], and 4 of the 40 patients described by Kun *et al.*, were correctly diagnosed before surgery [9]. Schwannomas of the submaxillary gland are very infrequent. A review of the literature revealed only few similar cases published to date [4, 5, 7, 11-14]. In tumors located within the parotid or submaxillary cells, diagnostic confusion centers on pleomorphic adenoma - which likewise involves a long-evolving, hard and painless growth. The facial nerve is usually unaffected. The diagnostic inefficacy of fine needle aspiration cytology in our case coincides with the results of other authors. This technique is thus of questionable utility in such cases where the clinical findings typically provide few clues for the pathologist [4, 9, 15]. Zbieranowski *et al.* proposed the application of immunohistochemical techniques or electron microscopic studies of the aspirate to improve the diagnostic performance of fine needle aspiration biopsies [16]. Other authors have in turn referred cases where fine needle aspiration mistakenly identified the lesion as pleomorphic adenoma [5].

Other differential diagnostic possibilities comprise nonspecific lymphadenitis, tuberculous lymphadenitis (scrofula), metastasis from oral or nasopharyngeal carcinoma, and other benign mesenchymal tumors (lipoma, lymphangioma) [6]. In our case, these diagnostic possibilities were discarded by the exploratory findings or other studies. The topographic location of the lesion and/or clinically manifest neurological defects provide the only clinical clues to the nerve from which the neoplasm originates. When as in most cases no such neurological manifestations are observed, the location of the tumor and the echographic, computed tomography and/or magnetic resonance findings inform of the origin of the growth, which in any case must be confirmed at surgery. In our patient both the location of the tumor and the computed tomography findings suggested an intraglandular neoplasm.

Simple surgical excision is the treatment approach advocated by most authors [1, 2, 4, 10]. When an intimate relation between the tumor and a given cranial nerve is identified (particularly the facial nerve), a number of management approaches may be evaluated. In this sense, some authors advise incomplete excision of the tumor, to avoid damaging the nerve of origin [1, 6, 13], while others recommend removal of the affected portion of the nerve followed by immediate reconstruction with direct microsuture techniques or autologous sural nerve grafting [3, 17]. The radioresistance of the tumor is a contraindication to radiotherapy [3].

Most studies in the literature report the absence of postoperative recurrences, even in the event of subtotal resections. Sporadic recurrences have been reported in some series, however [1, 9]. Similarly, most large series consider malignant transformation of schwannoma to be highly unlikely - though the possibility of malignant transformation has been reported, particularly in recurrent neurilemmomas [1, 9, 18].

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**Conflicts of Interest**

None.

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