

## Ancient schwannoma of the parotid gland: A case report and review of the literature

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**SUMMARY.** Schwannomas are encapsulated benign tumours arising from nerve sheath cells, of which ancient schwannoma is one of five variants. Since the first description, only a few ancient schwannomas have been reported in different locations in the head and neck region. In the parotid gland, this tumour is very rare. In this report, a 41-year-old female patient with an ancient schwannoma of the parotid gland is presented and the data of this patient compared with other (comparable) cases described in the literature. © 2005 European Association for Cranio-Maxillofacial Surgery

**Keywords:** schwannoma; neurilemmoma; ancient; parotid gland; neck

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### INTRODUCTION

Schwannoma (neurilemmoma) is an infrequent, benign, encapsulated tumour arising from nerve sheath cells (Conley, 1955). They are generally smooth and uniform in gross appearance and are usually surrounded by or attached to their nerve of origin (Toriumi et al., 1986). Although more commonly encountered in the extremities, approximately 25% of the cases originate from the neural structures of the head and neck region (Conley, 1955). In this region, schwannomas comprise a significant number of the tumours of the parapharyngeal space (Maniglia et al., 1979). Histologically, five variants of schwannomas have been described namely common, plexiform, cellular, epithelioid, and ancient schwannoma (Weiss and Goldblum, 2001).

The term ancient neurilemmoma was first suggested by Ackerman and Taylor (1951) in a review of 48 neurogenic tumours of the thorax. They reported 10 cases showed similar features of a typical neurilemmoma, but distinctive because significant portions of these tumours were composed of only few cells within a hyalinized matrix. They clarified that these features occurred in the schwannomas of long duration, and hence coined the term 'ancient schwannoma'. This type is characterized by diffuse areas of hypocellularity, focal accumulations of hyaline material, and fatty degeneration (Eversole and Howell, 1971). In addition, thick capsules, relative absence of necrosis, and being usually infiltrated by large numbers of siderophages and histiocytes plus hyperchromatism are misleading in suggesting malignancy (Ackerman and Taylor, 1951).

Since that first report, a few authors described ancient schwannoma in a variety of locations in the head and neck region (Table 1). The parotid gland location of this tumour is very rare. To our knowledge there has only been one case reported of an ancient schwannoma of the parotid gland in the scientific literature (Jayaraj et al., 1997). In this report, a case of an ancient schwannoma of the parotid gland is presented, discussed, and the literature reviewed for head and neck location of this tumour.

### CASE REPORT

A 41-year-old female patient complained of a painless and progressively increasing mass in the right side of the preauricular region for 5 years (Fig. 1). Clinical and radiological examinations (Fig. 2) identified a firm, oval mass, measuring approximately 5 cm in diameter, covered by normal skin and relatively fixed. The facial nerve functions were normal. The patient denied otalgia, fever, weight loss or night sweats. The remainder of the physical examination including head and neck was normal.

The complete blood count was normal. Ultrasonography and fine-needle aspiration biopsy were non-diagnostic, suggesting benign cystic contents.

The patient underwent surgical removal of the mass by total parotidectomy with preservation of the facial nerve and its branches under general anaesthesia. The 'cystic' mass was dissected from the surrounding tissues. Extirpation was difficult, due to location of the tumour in the deep portion of the parotid. Post-operatively the recovery

was uneventful. Facial nerve function was undisturbed.

### Histologic findings

**Gross features:** The resected specimen consisted of a tan-brown, well-circumscribed parotid gland measuring  $9.5 \times 5.5 \times 3.5$  cm and weighing 45 g. The cut surface showed a  $5.5 \times 5 \times 3$  cm nodular lesion with solid and cystic areas.

**Microscopic features:** On microscopic examination, an encapsulated tumour was seen in the parotid gland

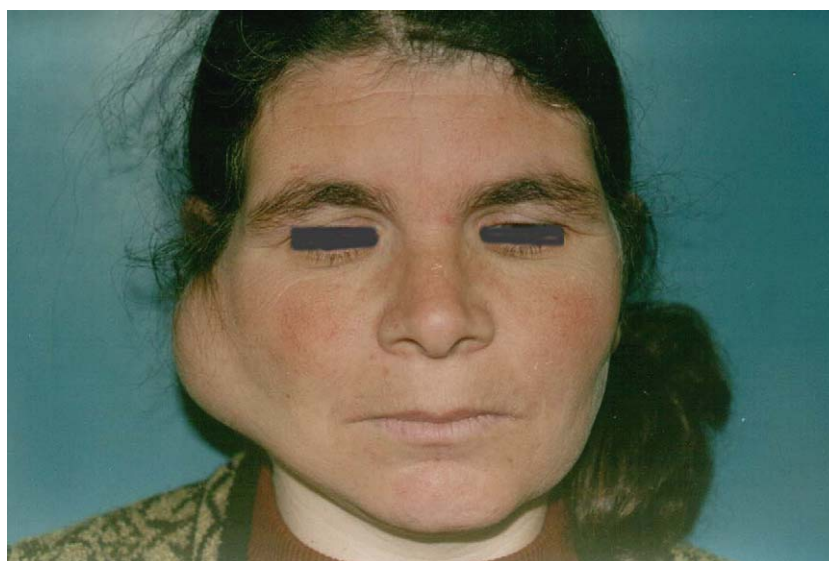
(Fig. 3). The tumour showed solid cellular and hypocellular areas and a large cystic space. The medium and high power fields showed tumour cells that were spindle shaped with twisted nuclei and indistinct cytoplasmic borders. They were arranged in short bundles or palisades. In some areas, the tumour was infiltrated by large numbers of siderophages and histiocytes (Fig. 4). Degenerative changes including nuclear atypia and hyalinization were also seen. Immunohistochemical investigation revealed diffuse, strong positive staining for S-100 protein (Fig. 5). In the light of these findings, the diagnosis of an 'ancient schwannoma' was made.

**Table 1** – Ancient schwannoma cases (1 each) in head and neck region

Literature	Location
<i>Eversole and Howell</i> (1971)	Oral cavity
<i>Marks et al.</i> (1976)	Oral cavity
<i>McCoy et al.</i> (1983)	Oral cavity
<i>Dayan et al.</i> (1989)	Oral cavity
<i>Ogren et al.</i> (1991)	Infratemporal fossa
<i>Nakayama et al.</i> (1996)	Oral floor and tongue
<i>Bondy et al.</i> (1996)	Submandibular gland
<i>Jayaraj et al.</i> (1997)	Parotid gland
<i>Moore et al.</i> (1997)	Posterolateral pharynx
<i>Khawarg et al.</i> (1999)	Orbit
<i>Ledesma et al.</i> (1999)	Oral cavity
<i>Saydam et al.</i> (2000)	Cervical vagus
<i>Walther et al.</i> (2001)	Parapharyngeal space
<i>Zachariades et al.</i> (2001)	Cervical region
<i>Hidaka et al.</i> (2001)	Neck
<i>Longo and Califano</i> (2002)	Auricularis major nerve
<i>Badawi and Scott-Coombes</i> (2002)	Thyroid
<i>Huang et al.</i> (2002)	Infratemporal fossa
<i>Darwish et al.</i> (2002)	Cervical spinal cord
<i>Tzagkaroulakis et al.</i> (2003)	True vocal cord
<i>Moloney et al.</i> (2004)	Orbit
Current case	Parotid gland



**Fig. 2** – CT scan: large, 'cystic' space-occupying mass located in the right preauricular region.



**Fig. 1** – Forty-one-year-old female with a mass occupying the right preauricular region.

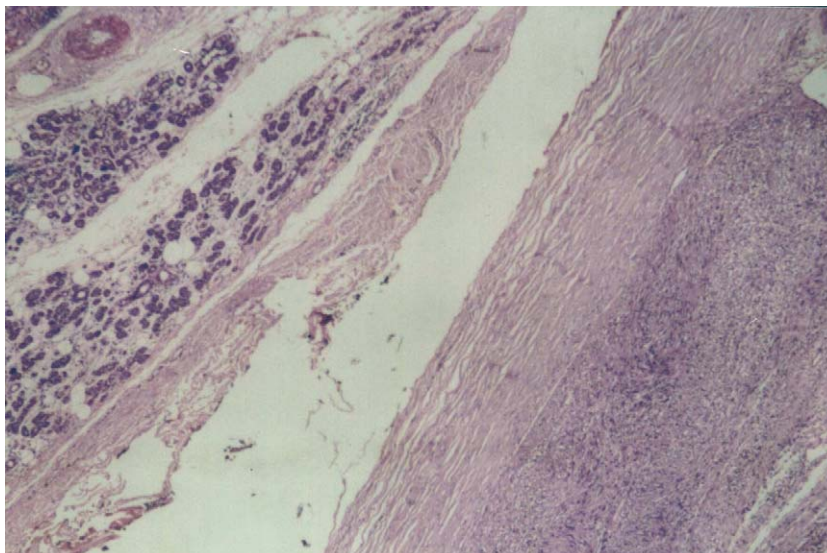


Fig. 3 – Encapsulated tumour in the parotid gland (on the right; HE, original magnification  $\times 100$ ).

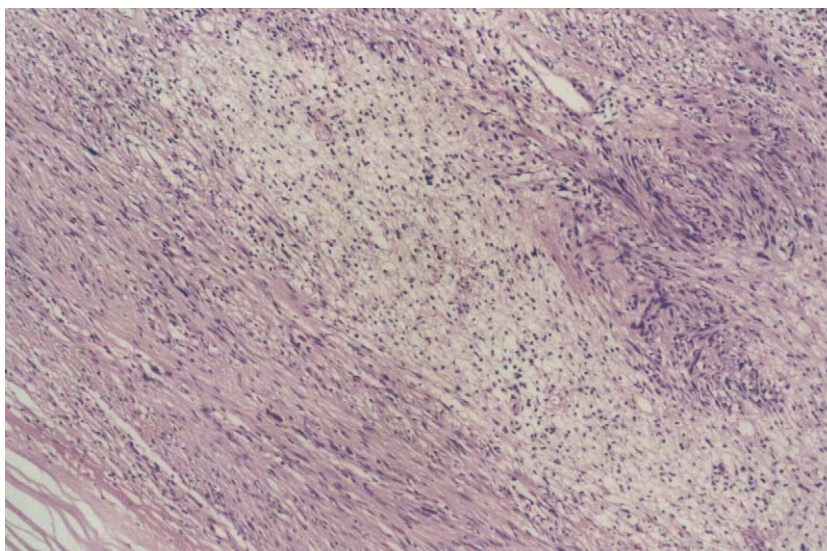


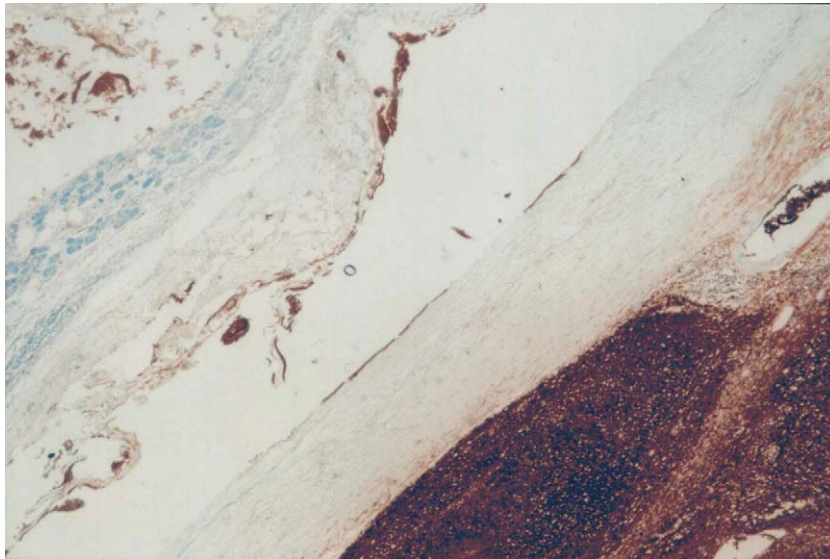
Fig. 4 – Tumour infiltrated by large numbers of siderophages and histiocytes (HE, original magnification  $\times 100$ ).

## DISCUSSION

Schwannoma is a tumour of the neurilemma, i.e. the nerve sheath of Schwann cells, being characteristically solitary, encapsulated and occurring along the course of a peripheral, cranial or sympathetic nerve (Huang et al., 2002). They typically enlarge slowly with minimal symptoms until size and impact on other structures make them evident. These tumours can vary from firm, solid masses to fluctuant cysts. Their characteristic histological appearance is dominated by an encapsulated lesion arising from a nerve and composed of an intimate mixture of spindle cells forming highly cellular so-called Antoni A areas and less cellular, myxoid Antoni B areas. Typically, those lesions that are long-standing may undergo degen-

erative ‘ancient’ changes dominated by large cystic, myxoid areas with variable bizarre spindle cells and even occasional mitoses (Jayaraj et al., 1997). In the case described here, the tumour was encapsulated, and constituted both solid and cystic areas as notified in the previous reports.

Approximately 25–40% of all schwannomas occur in the head and neck area (Saydam et al., 2000) whilst ancient schwannoma rarely affects the head and neck region (Ogren et al., 1991). It is an extremely rare instance to encounter an ancient schwannoma in the parotid gland. Although there are a few ancient schwannoma cases reported in the head and neck region, the literature review yielded only one case in the parotid gland (Jayaraj et al., 1997; Table 1).



**Fig. 5** – Immunohistochemical investigation revealing diffuse, strongly positive staining for S-100 protein (on the right; S-100, original magnification  $\times 100$ ).

In their original description, *Ackerman and Taylor* (1951) proposed that the ancient schwannoma begins as a diffuse cellular overgrowth with increased vascularization, followed by decreased vascularity with resulting hyalinization. Especially in frozen sections, the areas with hypercellularity and atypia may lead the pathologist to favour a diagnosis of a malignant lesion (*Saydam et al., 2000*). *Dahl* (1977) reported 6 out of 11 cases of ancient schwannoma to have been originally misdiagnosed as a malignant tumour (such as sarcoma). Prior to the realization that the nuclear atypia and hyperchromatism observed in ancient schwannomas were not a sign of malignancy but rather a regressive phenomenon, many of these lesions were erroneously diagnosed as sarcomas (*Dodd et al., 1999*). In microscopic evaluation of the current case, cellular and hypocellular areas that can be seen in benign lesions were noted. Nevertheless, nuclear atypia, which arouses a suspicion of malignancy were also seen.

Head and neck schwannomas are frequently misdiagnosed and preoperative investigations are often fruitless (*Badawi and Scott-Coombes, 2002; Jayaraj et al., 1997*). Although aspirates of ancient schwannoma may show some features such as nuclear pleomorphism, nuclear inclusions, perivascular sclerosis, xanthomatous changes, or nuclear atypia (*Dodd et al., 1999*), fine-needle aspiration biopsy has questionable value (*Kun et al., 1993*). It may show unclear histopathological results from these cases (*Krause et al., 1993; Hidaka et al., 2001*), which have the potential to confuse this lesion with a more serious one such as sarcoma (*Dodd et al., 1999*). In the current case, the diagnosis was only evident post-operatively.

These lesions should be treated by surgical excision (with every surgical effort made to preserve the facial

nerve). If the nerve is too thin or splayed out, grafting of the defective nerve segment should be done (*Saydam et al., 2000*). In the case described, the mass originated from the deep portion of the parotid but the facial nerve was not involved, so dissection was done with preservation of the nerve and its branches. Post-operatively facial nerve function was normal. During routine follow-up, the patient was free from disease 2 years later.

## CONCLUSION

A case of an ancient schwannoma is described. This type of tumour is uncommon in the head and neck region and extremely rare in the parotid gland. Fine-needle aspiration biopsy may be histologically misleading. Complete excision leads to complete cure.

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