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Case Report

A Rare Case Sebaceous Adenoma of the Parotid Gland in Patient with Steinert's Syndrome: Case Report

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Abstract
Sebaceous adenoma is an extremely rare tumor located in the parotid gland. In the English literature, 13 cases have been reported. Sebaceous adenoma represents 0.5% of all monomorphic adenomas. The authors are presenting a case of sebaceous adenoma of the parotid gland in a 45-year-old male who presented with a mass in the left parotid area that had been gradually enlarging for two years without symptoms of pain. On imaging, a well-defined mass lesion in the left parotid region was seen eroding the overlying skin. Histopathological findings were consistent with sebaceous adenoma. Surgical excision is curative. The prognosis is excellent, with a low recurrence rate. The present case report will increase awareness and possibility of this rare tumor occurring at an unusual site, thereby avoiding any chance of misdiagnosis.

Keywords: *Salivary gland neoplasm, parotid gland, sebaceous adenoma.*

INTRODUCTION

Sebaceous adenomas are exceptionally rare benign salivary neoplasms, accounting for approximately 0.1% of all salivary gland tumors [1]. Due to their infrequent occurrence, these lesions are often misdiagnosed preoperatively as more aggressive malignancies, necessitating careful histopathological differentiation to avoid overly radical surgical interventions [2].

CASE REPORT

A 45-year-old man presented with a slowly growing palpable mass of the left parotid gland, which was first seen one year earlier. There was no history of pain, bleeding, trismus or discharge from swelling during the course of enlargement, history of fever, or altered salivary flow.

The patient presents as the co-morbidity of Steinert's syndrome, it is a genetically hereditary multisystem and progressive condition, characterized by muscular weakness (more distal than proximal), myotonia (difficulty in muscle relaxation), cataracts, cardiac and endocrine problems.

Physical examination of the left parotid gland disclosed a soft, non-mobile parotid gland tumor of 8 cm in diameter and had no tenderness to palpation. The tumor infiltrated the skin and was attached to deeper tissues. No signs of cervical lymphadenopathy were observed; however, neurological involvement was present: Steinert´s syndrome (A rare genetic multi-system disorder characterized by a wide range of muscle-related manifestations -*muscle weakness, myotonia, early-onset cataracts before age 50*- and systemic manifestations -*cerebral, endocrine, cardiac, gastrointestinal tract, uterus, skin, and immunologic involvement*- that vary depending on the age of onset) [4,5].

Based on the history, clinical examination, and CAT findings, a provisional diagnosis of carcinoma was made. The biopsy report adenoma sebaceus and confirmed by immunohistochemistry.

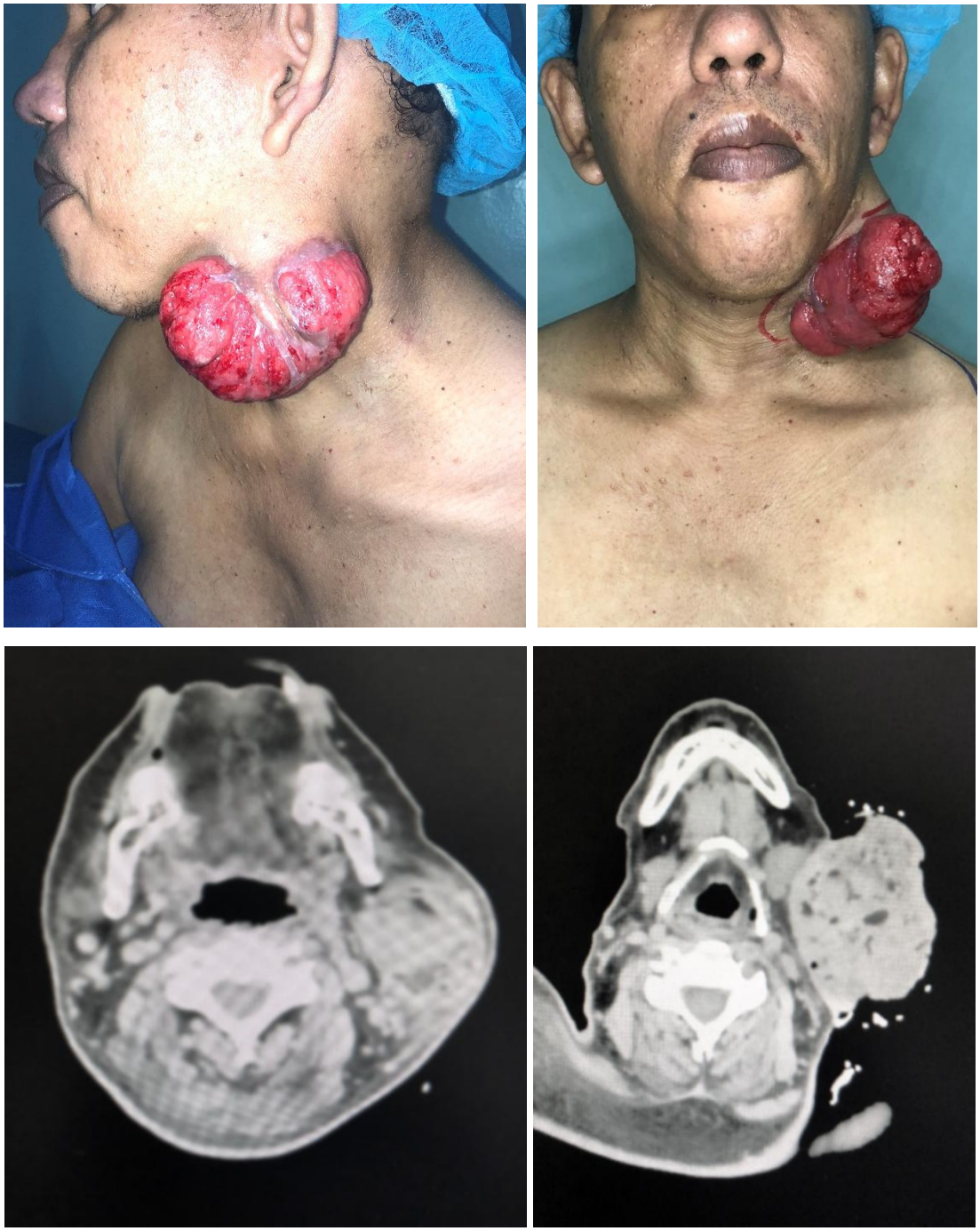


Fig. 1. Computed tomography (CT) with contrast showing a heterogeneous, partially cystic mass encompassing superficial and deep lobes of the parotid gland. Note dark areas with consistency of adipose tissue.

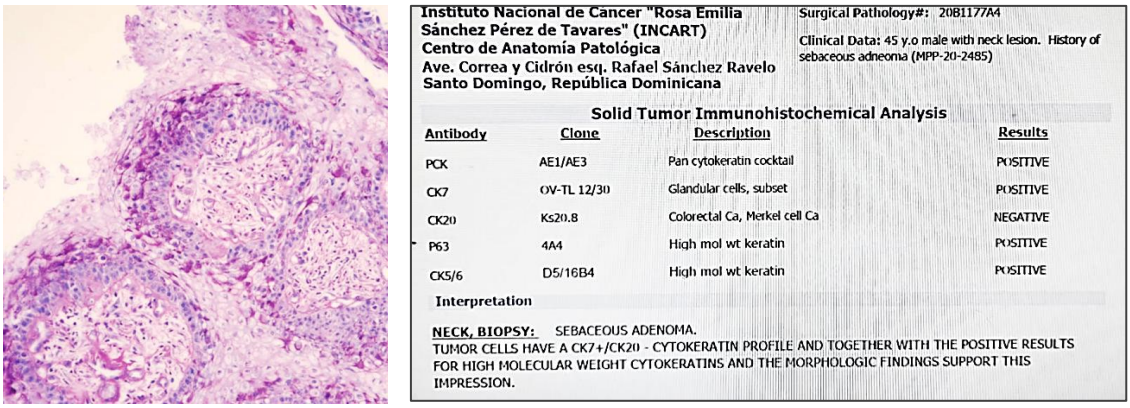
Histopathology reported a well-circumscribed tumor of 9 × 9 × 6 cm, surrounded by normal parotid tissue, white, yellowish color, and some cystic spaces. The cut surface was homogeneous, with small cystic spaces (Figure 2a, b).

Histologically, paraffin sections stained with hematoxylin and eosin showed a microcystic appearance with numerous sebaceous glands with various dimensions embedded in stromal fibrosis (Figure 3a, b). Occasionally, the glandular structures are dilated and contain sebaceous material (Figure 3c, d).

The tumor has a multilobulated appearance and may involve the overlying epidermis (as seen here). The individual lobules attempt to recapitulate the architecture of normal sebaceous glands. At the periphery of the lobules are layers of small germinative cells with basaloid appearance. These cells have small vesicular nuclei and scant cytoplasm. They merge towards the center of the lobules with more mature-appearing sebaceous cells containing abundant pale-staining

foamy cytoplasm and small hyperchromatic nuclei. At least half of each lobule in sebaceous adenoma consists of mature sebaceous cells. Increased mitotic activity is sometimes seen in sebaceous adenomas and should not be over-interpreted as sebaceous carcinoma.

In the immunohistochemistry of tumor cells there is a ck7+/ck20, cytokeratin profile and together with the positive results for high molecular weight cytokeratins P63 and CK5/6.



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Surgical Pathology#: 20B1177A4

Clinical Data: 45 y.o male with neck lesion. History of sebaceous adneoma (MPP-20-2485)

Solid Tumor Immunohistochemical Analysis

Antibody	Clone	Description	Results
PCK	AE1/AE3	Pan cytokeratin cocktail	POSITIVE
CK7	OV-TL 12/30	Glandular cells, subset	POSITIVE
CK20	Ks20.8	Colorectal Ca, Merkel cell Ca	NEGATIVE
P63	4A4	High mol wt keratin	POSITIVE
CK5/6	D5/16B4	High mol wt keratin	POSITIVE

Interpretation

NECK, BIOPSY: SEBACEOUS ADENOMA.
TUMOR CELLS HAVE A CK7+/CK20 - CYTOKERATIN PROFILE AND TOGETHER WITH THE POSITIVE RESULTS FOR HIGH MOLECULAR WEIGHT CYTOKERATINS AND THE MORPHOLOGIC FINDINGS SUPPORT THIS IMPRESSION.

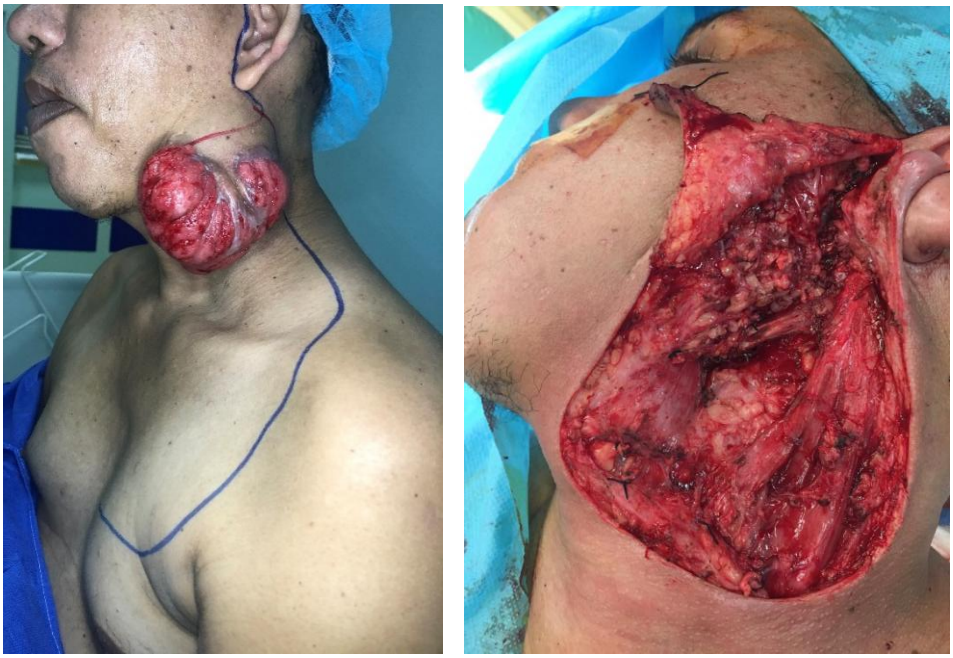
Figure 2. Sebaceous adenoma under magnification; cystic elements of the lesion are lined by both squamous and sebaceous cells. The surrounding stroma is fibrotic (original magnification, X200).

As recommended by the neurology department, caution should be exercised with the use of muscle relaxants like succinylcholine which can trigger a myotonic crisis, causing difficulties in orotracheal intubation [5] and ventilation, and efforts should be made to avoid a lengthy surgery.

As Manzon and Philbert [6] say, although the induction of general anesthesia is extremely safe in a healthy population, severe reaction to anesthetic and neuromuscular paralyzing agents in a myotonic patient can cause organ failure, myocardial infarction, and respiratory failure.

A left superficial parotidectomy with facial nerve preservation and cervicopectoral flap for neck skin reconstruction was performed. Has a general anesthesia controlled and approved by the neurology department. The patient was discharged 4 days after her surgical procedure without any complications.

The patient's postoperative course was complicated by necrosis in the tip of the flap, which healed and was left to heal by secondary intention, causing an unsightly scar on the neck; therefore, scar remodeling will be necessary. Until now (4 years later), he has remained without evidence of recurrence. The patient is still under follow-up.



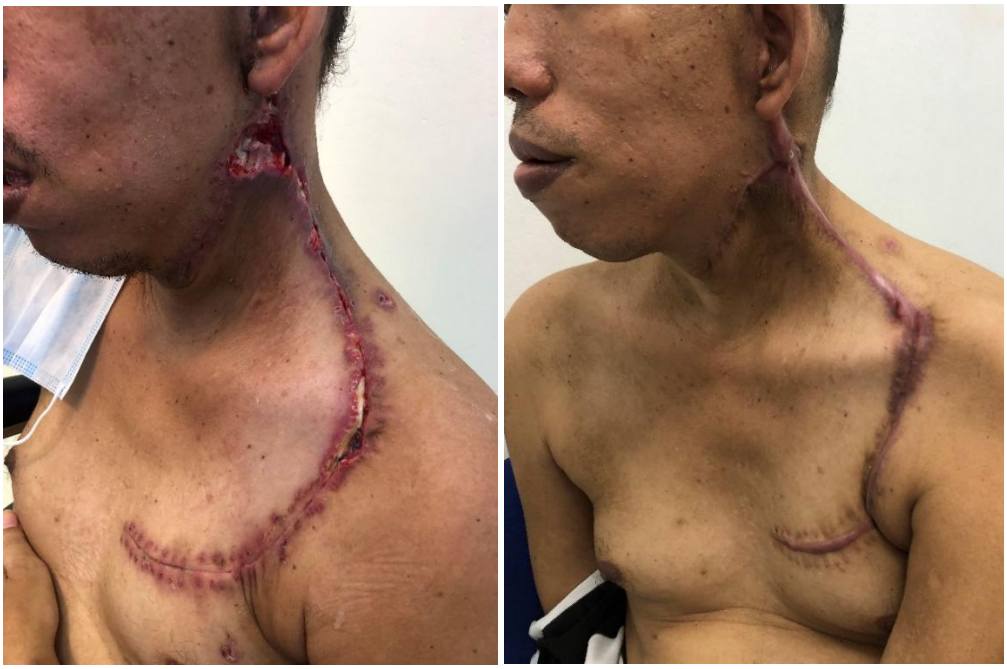


Fig. 4. Immediate postoperative view showing the necrotic area and a late postoperative view showing the healing of the surgical wound.

DISCUSSION

Hermann Hamperl (1899–1979) first noted the presence of sebaceous glands within salivary gland tissue in 1931 [11,14,22]. In spite of this observation, primary sebaceous tumors of the salivary glands are exceedingly rare, with only a few cases reported in the literature.

The parotid gland develops from a tubular outgrowth of the mucosa that lines the oral cavity which appears at about the 45th day of the fetal life. The proximal portion of this outgrowth persists as the parotid duct while the proliferation at the distal end forms the glandular parenchyma [18].

The origin of sebaceous cells or glands within salivary gland tissue is unknown and is not attributable to an ectopia. Gnepp [3] stated that sebaceous foci could be congenital in origin, naturally develop later in life or be due to a metaplastic process secondary to ductal obstruction. Thus, sebaceous cells represent another potential differentiation of the pluripotential salivary gland duct epithelium.

Although sebaceous foci can be found in neonatal glands, their prevalence in children is lower than in later life which argues against their congenital presence [13].

Moreover, the presence of sebaceous glands and cells in major salivary glands seems to increase its incidence after puberty [10]. Gnepp and Brannon [11] believe that possibly the same factors that “activate” sebaceous glands in the skin during puberty also activate those in the salivary glands.

Although most commonly associated with hair follicles, ectopic sebaceous glands can be identified independently. These lesions can be found mainly on the eyelids and oral mucosa [7]. Occasionally it may arise in the major salivary glands [12].

Benign tumors can rarely originate from these sebaceous glands, such as sebaceous adenoma, sebaceous lymphadenoma, or sebaceous carcinoma [9].

Despite the common occurrence of sebaceous differentiation in salivary glands [13], sebaceous adenomas are very rarely encountered in major salivary glands, accounting for 0.1 percent of all salivary gland tumors [9] and less than 0.5 percent of all salivary adenomas [14]. Most of them develop in the parotid gland, but they can rarely develop in the submandibular and minor salivary glands [10]. There was no impact on sebaceous adenomas of the parotid gland in either gender [15]. The majority of cases are diagnosed in patients 60 or older [13].

We performed a search on two databases, PubMed® and Web of Science®, published during the all-time topic, screening for the keyword “sebaceous adenoma of the parotid gland”. After the screening, we found a few reports concerning sebaceous adenoma located in the parotid gland (Table 1).

Table_1: Characteristic of sebaceus adenoma involving the parotid gland reported previously published literature.

Cases	Autor/year References	Age (Years)	Sex	Clinical features of the tumors	Follow-up
1	Foote & Fazell (1953)			3.5 cms mass	
2	Bad & Ulmansky (1959)	57	F		6 Month,NR
3	Pieter & Seymour (1981)				o
4	Gnepp & Brandon (1984)	71	F	14mm diameters mass	1.5 Years, NR
5	Gnepp & Brandon (1984)	74	M	23 mm diameters mass	6 Years, NR
6	Shen (1994)	39	M	5X6X4cms mass	19 months NR
7	Derias (1994)	73	F	-	-
8	de Vicente Rodríguez (2006)	59	F	3.5 cms mass	5 years NR
9	Welch (2007)	2	M	5 cms mass	-
10	Apple (2009)	29	F	4 cms mass	
11	Farahnik et al. (2016)	13	F	2.5X2cm mass	
12	Dinka et al. (2022)	65	F	3.2x2x1.6 cms mass	4 months NR
13	Singh et al. (2024)	67	M	4cm mass	
14	Our Case (2026)	45	M	8x8x4 cm	4 years NR

Following the analysis of the all-time literature, the current case is the tenth reported case of sebaceous adenoma with localization in the parotid gland. The case presented showed differentiated sebaceous lobules accompanied by a fibrous stroma.

Thus, de Vicente-Rodríguez reported the histopathological appearance in a case of sebaceous adenoma located in the parotid gland [22]. Foote & Frazell reviewed 877 tumors of the major salivary glands and revealed only one case of sebaceous adenoma of the parotid gland [17].

Bab & Ulmansky reported a case of an adenoid cystic carcinoma associated with a sebaceous cell adenoma [18]. Amongst the 183 cases reported by Pieterse and Seymour, there was one case of sebaceous adenoma [19]. Gnepp and Brannon reviewed 21 cases of primary salivary gland sebaceous tumors and found three sebaceous adenomas located at the parotid gland [11].

Due to the low prevalence of the sebaceous adenomas of the parotid gland and the low number of published cases, obtaining an exhaustive description of the characteristics of the disease is difficult.

Sebaceous adenomas of the skin sites may be associated with nonpolyposis colorectal cancer, a condition known as Muir–Torre Syndrome [25,26], an autosomal-dominant disorder characterized by a combination of at least one cutaneous neoplasm and at least one visceral malignancy [27]. No such relationship between these tumors within the major salivary glands and Muir–Torre Syndrome has been documented [28], but it has not been associated with Steinert's syndrome.

Generally, sebaceous adenomas in the parotid gland are painless and slow-growing well, encapsulated lesions that do not infiltrate the surrounding normal tissues, with no facial nerve involvement. The tumor size typically ranges from 0.5 to 5 cm in diameter. Farahnik *et al.* to note an intraparotid sebaceous adenoma that demonstrated hypervascularity in need of embolization [29].

They histologically appear as white-yellowish nodules that have a microcystic pattern, with abundant well, differentiated sebaceous glands of variable size [9,29]. The absence of the lymphoid component did not support a diagnosis of sebaceous lymphadenoma [21].

Besides clinical examination, ultrasonography, computed tomography, and magnetic resonance imaging (MRI) are the most common radiological procedures for the diagnosis of tumor-like lesions of the salivary glands. According to Haynes et al., the initial diagnostic test for salivary gland tumors should be computed tomography, which offers information regarding the mass's size, extent, location, content, and consistency [30].

However, ultrasound is accepted as the first imaging method for assessing soft-tissue diseases in the head and neck, including the major salivary glands, being beneficial in distinguishing cystic from solid lesions [31]. The ultrasonography can be used to establish the need for imaging procedures, particularly in those lesions showing malignant features on ultrasonography or large masses located in the deep lobe [32]. Due to poor economic status, our patient was not willing for CT; hence, the ultrasonography was conducted.

There is no consensus about using fine-needle aspiration biopsy (FNAB) in major salivary gland tumors [33]. According to some researchers, parotid tumors other than pleomorphic adenomas are uncommon, and pathologists may misdiagnose FNAB if they are not specialized in parotid tumors [34].

In the current case, no FNA was performed as that was a frankly benign parotid tumor, and the plan was to proceed for superficial parotidectomy directly. Differential diagnosis should include other benign parotid tumors with solid mass with cystic change patterns [20].

Usually, these benign tumors can be removed, and they are not likely to recur if adequately excised. Since there is no evidence for efficacy, radiation therapy should not be considered an alternative to surgery in sebaceous adenoma. Thus, curative surgical excision is the treatment of choice.

CONCLUSIONS

The authors described a sebaceous adenoma of the parotid gland is a very rare benign entity that can clinically mimic a malignant neoplasm, especially when it is large, fixed, or involves superficial tissues. Therefore, it should be included in the differential diagnosis of parotid masses, and its confirmation depends on histopathological and immunohistochemical examination.

The preferred management is complete surgical excision, usually with parotidectomy and preservation of the facial nerve when possible. If the resection is adequate, the prognosis is favorable, and recurrence is unlikely. In patients with comorbidities such as Steinert's syndrome, careful anesthetic planning is essential to avoid serious complications.

In this paper, the clinical presentation, pathological findings on the biopsy and surgical resection specimen, and review of the literature will be discussed.

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