

## A Hypervascularized Sebaceous Adenoma of the Parotid Gland in a 13 Year-old Female: A Case Report

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### Abstract

We describe the rare case of a 13 year-old girl with a hypervascularized sebaceous adenoma of the parotid gland. Our case is one of only thirteen other cases of parotid gland sebaceous adenoma to be reported in the literature. The difficulty in diagnosing sebaceous adenomas of the parotid gland preoperatively has been reported previously and as such we present a literature review to typify the clinical and histological characteristics of sebaceous adenoma when present in the parotid gland.

**Keywords:** Sebaceous adenoma; Salivary gland; Parotid gland; Benign mass

### Introduction

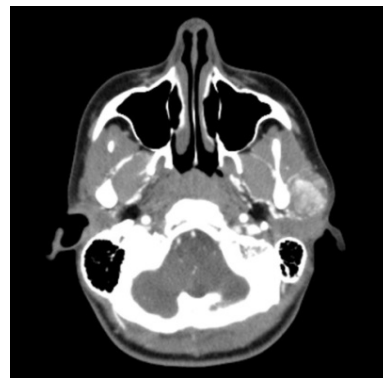
Despite the rather typical finding of sebaceous elements in salivary gland tissue, sebaceous differentiation in salivary gland lesions occurs very rarely [1] with sebaceous adenomas representing about 0.1% of all salivary gland tumors and less than 0.5% of all salivary adenomas [2]. A sebaceous adenoma is a rare, benign, solitary epithelial tumor composed of cells showing sebaceous differentiation without cellular atypia and with minimal pleomorphism [1,3,4]. Sebaceous adenomas are typically well-circumscribed or encapsulated and can be of solid or cystic forms [4]. There have been about 30 cases of salivary gland sebaceous adenomas reported in the English medical literature. This case report describes one of only thirteen other reported cases of a sebaceous adenoma in the parotid gland and it describes the first noted case of sebaceous adenoma associated with hypervascularity in the parotid gland. The major problem in dealing with sebaceous adenomas is primarily in recognizing them: proper identification of these benign tumors is important to avoid confusing them with more aggressive neoplasms whose presence would lead to more radical surgeries [2]. We describe our case and also review past cases of other sebaceous adenomas of the parotid gland to better characterize these relatively under-reported mass lesions.

### Case Presentation

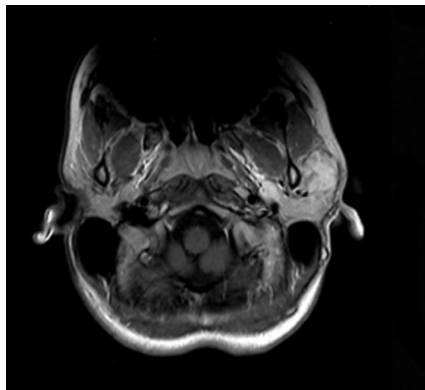
A 13-year-old female presented in clinic for evaluation of a left preauricular mass that she had first noticed about two years prior. The patient reported that the mass had doubled in size in the past two months. She had not experienced any recent weight-loss, constitutional symptoms, facial weakness, or difficulty in mastication or swallowing. She had no significant past medical history except for an operation to repair a traumatic finger fracture. In clinic, physical examination was notable for a 2.5 × 2 cm mass in the left pre-auricular region with no overlying skin changes. The mass was noted to be firm, non-pulsatile, and tender to palpation by the physician. The facial nerve was intact

and symmetric bilaterally with no signs of weakness. She was advised on further necessary studies and the possibility for surgery.

The patient elected to pursue any necessary studies preoperatively. A CT angiogram was performed and revealed a hypervascularized mass in the left preauricular region with a surrounding 2.5 × 2 cm dense mass in the superficial lobe of the parotid gland, without involvement of the deep lobe (Figure 1). The scan revealed that the mass posteriorly displaced the retromandibular vein and facial nerve. Both CT and MR angiograms (Figures 1 and 2) were suggestive of a possible hemangioma; however, an ultrasound guided fine-needle aspiration (FNA) biopsy revealed a parotid gland tumor with benign features. The fine-needle aspiration cytology, as well as the CT and MR images, were jointly reviewed by the pathologist and radiologist at a multidisciplinary tumor board conference with the conclusion that the mass was most likely a hypervascularized pleomorphic adenoma. The patient elected to undergo interventional radiology embolization due to the hypervascular character of the mass, followed by surgical excision.



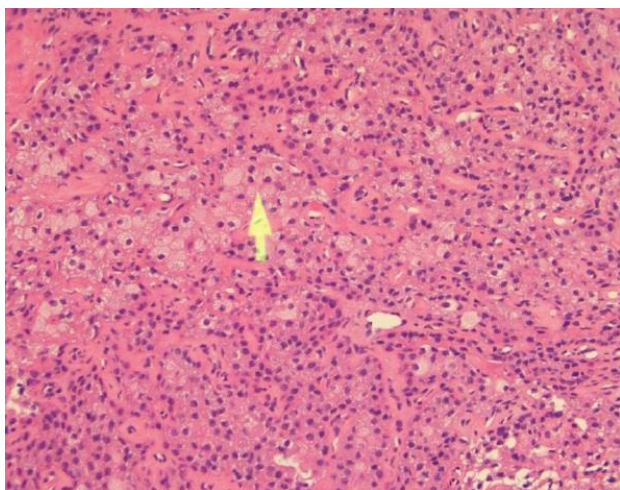
**Figure 1:** CT angiogram revealing a vascular mass in the left preauricular region



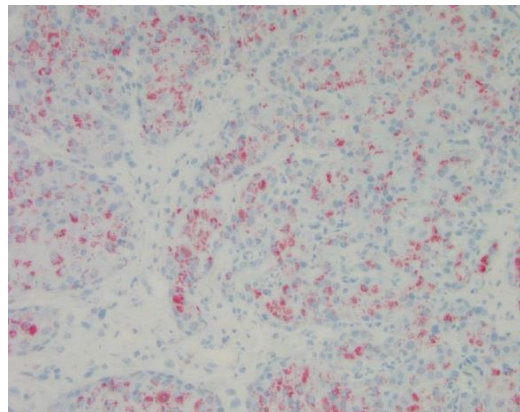
**Figure 2:** MR angiogram suggests a well circumscribed mass in the left parotid region

During the surgical excision, the previously anticipated superficial parotid lobe tumor was actually found to be a deep lobe tumor necessitating a total parotidectomy. An intra-operative frozen section analysis was performed and sent for pathologic examination.

Histologically, the specimen was noted to be a complete intraparotid tumor and appeared well-circumscribed, although no definite capsule was seen. Pathology identified numerous nests and islands of tumor cells surrounded by a fibromyxoid stroma. Hematoxylin and Eosin stain showed tumor cells that were composed of squamous cells admixed with vacuolated cells (Figure 3). No significant cytologic atypia, mitoses, or necrosis were seen. Oil-red-O stain on frozen section tissue highlighted intracellular lipid droplets in the vacuolated cells (Figure 4). Mucicarmin stain demonstrated extracellular mucin only. All these features combined led to the final diagnosis of a hypervascularized sebaceous adenoma presenting in the parotid gland.



**Figure 3:** Hematoxylin and Eosin stain shows numerous nests and islands of tumor cells surrounded by fibromyxoid stroma. No significant cytologic atypia, mitoses, or necrosis are seen



**Figure 4:** Oil-red-O stain on frozen section tissue highlighting intracellular lipid droplets (LD) in the vacuolated cells (VC). Mucicarmin stain demonstrates extracellular mucin

## Discussion

A sebaceous adenoma of salivary gland origin is a rare entity characterized as a benign, solitary epithelial tumor composed of cells showing sebaceous differentiation without cellular atypia and with minimal pleomorphism [5-7]. Sebaceous adenomas are typically well-circumscribed or encapsulated and can be of solid or cystic forms and are painless, slow-growing masses [6]. The tumor presented in our case demonstrated these qualities that distinguished it from other potential diagnoses.

The histopathological differential diagnosis of salivary gland sebaceous neoplasms to be aware of includes: (1) sebaceous adenoma, (2) sebaceous lymphadenoma, (3) sebaceous carcinoma, (4) sebaceous lymphadenocarcinoma, and (5) other tumors that present with sebaceous differentiation, including pleomorphic adenomas with sebaceous elements or mucoepidermoid tumors with sebaceous elements, or a Warthin's tumor with sebaceous elements [4,8-10].

A sebaceous lymphadenoma is a rare, benign, encapsulated tumor typically presenting as a slowly enlarging mass consisting of well-differentiated sebaceous cells, with varying shaped and sized nests of sebaceous glands and ducts, lying in a stroma of lymphocytes [3]. In sebaceous lymphadenoma, cellular atypia is minimal and there is typically no local invasion exhibited [4]. The sebaceous adenoma in our case was distinct from its lymphadenoma-variant due to its lack of lymphocytic components.

Sebaceous carcinomas, the malignant counterpart of sebaceous adenomas, typically present as painful, well-circumscribed masses causing varying degrees of facial nerve paralysis and occasional fixation to the skin. Sebaceous carcinoma is composed mostly of sebaceous cells with varying degrees of maturity that are arranged in sheets, with varying degrees of nuclear atypia, pleomorphism, and invasiveness present [4-6]. In sebaceous carcinomas, areas of cellular necrosis and fibrosis are commonly found [6], unlike in the tumor presented here. Nuclear atypia and pleomorphism varies from mild to severe, but is more prevalent among sebaceous carcinomas than sebaceous adenomas [4].

Sebaceous lymphadenocarcinomas are focally encapsulated and locally invasive with foci of sebaceous lymphadenoma intermixed with or adjacent to regions of pleomorphic carcinoma [4,6,11], all of which were not seen in the tumor presented in our case.

Sebaceous adenomas can sometimes be confused with mucoepidermoid carcinomas histopathologically [4,11]. The two can be distinguished because mucoepidermoid carcinomas can demonstrate intracellular mucin in some clear cells, while in sebaceous adenomas mucin positivity is never found in the clear sebaceous cells (although mucin can sometimes be detected within ducts adjacent to sebaceous cells) [4-7,10]. Further, unlike the sebaceous differentiated tumors that can show luminal holocrine secretions and vacuolated cytoplasm, mucoepidermoid carcinomas typically exhibit clear cytoplasm that focally contain the intracellular mucin [4,6].

Overall the following criteria were adhered to in establishing the diagnosis of a sebaceous adenoma for the tumor in our case: (1) a sharply circumscribed lesion with an oranoid pattern, (2) irregularity of size and shape of the sebaceous lobules, (3) appearance of both mature sebaceous cells and small germinal cells that may be arranged in an irregular pattern, and (4) lack of dilated excretory duct or a common excretory duct after serial sections [1]. In other words, microscopically, sebaceous adenoma is at least partially encapsulated and tumor cells reveal both squamous and sebaceous differentiation in variable proportions [4-6]. Sebaceous adenomas are made of undifferentiated peripheral basoloid (germinal cells) and mature sebaceous cells in the lesion center [5] typically embedded in fibrous stroma [4]. Lymphoid follicles, necrosis, and mitoses are not typically observed in sebaceous adenomas [4] and this tumor-type has occasionally included mucus-containing cells or oncocyctic cells [6]. The findings of oncocyctic cells combined with findings of sebaceous tumors with mucin-producing components in ductal components (but not in sebaceous cells) demonstrate the pluripotency of these tumors; thus, sebaceous tumors must be carefully evaluated to determine malignancy status [1,6]. The diagnosis of benign versus malignant sebaceous differentiated tumors can be made by checking the lesion for the lack of oncocytes by the indications of a Sudan positive fat deposit in the cytoplasm, almost complete lack of mitochondria, and a content of lipid drops and desmosomes [8,6,7,10], as in our benign case.

Sebaceous glands are prominent cutaneous adnexal components [3]. Although the presence of sebaceous glands in the parotid and submandibular glands is a typical and normal finding (sebaceous glands are present in about 10–42% of normal parotid glands and 5–6% of normal submandibular glands), sebaceous glands can be so few in number that they are sparsely encountered in pathological analysis [4,11]. The etiology for sebaceous differentiation in salivary glands is unknown; however, most authors agree that sebaceous differentiation develops naturally in late life since sebaceous components are rarely encountered in children before puberty [1]. Despite the rather typical finding of salivary gland tissue sebaceous differentiation, primary salivary gland sebaceous differentiated lesions occur very rarely [1] with sebaceous adenomas representing about 0.1% of all salivary gland tumors and less than 0.5% of all salivary adenomas [9]. When present, sebaceous adenomas are most often located in the parotid gland, submandibular gland, sublingual gland, buccal mucosa, or minor salivary glands [3,4,8].

Of the 13 cases of sebaceous adenomas found in our literature review of the English medical literature, sebaceous adenomas of the parotid were always painless and noted as slow-growing, with time to presentation varying from 5 days to 15 years. The masses have been

noted to be encapsulated or well-circumscribed, and non-tender, soft, to firm on palpitation. On gross analysis, the masses have appeared as yellow to yellowish-gray to pinkish-white in color [4]. Intraparotid sebaceous adenomas have ranged in size from 1 to 5.0 cm and have occurred more commonly in older patients, although cases have been noted of a 2 year-old boy [12], 16 year-old girl [13], and our case of a 13 year-old girl. Our case was the first to note an intraparotid sebaceous adenoma that demonstrated hypervascularity in need of embolization. No previously noted intraparotid sebaceous adenomas have revealed this quality, although rare instances of oncocyctic features of these masses have been noted.

The work-up and management of sebaceous gland parotid tumors is like that of other benign parotid masses. Preoperative workup may reveal a benign tumor that is otherwise difficult to distinguish from a pleomorphic adenoma. Fine-needle aspiration cytology may be followed by surgical excision for definitive diagnostic pathologic evaluation. Complete surgical excision is considered curative. Our literature review also demonstrated no evidence of recurrence after tumor excision with clear margins and this has been shown for follow ups ranging to 16 years. Deeply rooted tumors of the parotid gland (as in this case) may necessitate a total parotidectomy, but do not need to sacrifice the facial nerve.

In sum, sebaceous adenomas are benign, slow growing, painless, well-circumscribed tumors that are uncommon among adults and even rarer in children. Optimal management includes diagnosis by fine-needle aspiration cytology and imaging, followed by en bloc excision. Although the tumor-type we presented and described here is exceedingly rare in the pediatric population, otolaryngologists should be aware of the possibility of sebaceous adenomas in the diagnosis of parotid masses, even vascularized ones as we describe, and should tend toward treating these tumors conservatively.

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