CASE REPORT

Angiomyolipoma of the Parotid Gland: A Rare Presentation

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SUMMARY

We present the case of a girl in her late teens who presented with a slowly enlarging, painless mass in her right parotid gland over four years. Initial investigations, including ultrasonography and MRI, suggested a vascular malformation. Fine needle aspiration supported a benign tumour. Histological analysis following excision confirmed the diagnosis of angiomyolipoma (AML) of the parotid gland. This case highlights the importance of considering rare benign mesenchymal tumours like AML in the differential diagnosis of parotid gland masses, even in the absence of typical associations such as tuberous sclerosis.

BACKGROUND

Angiomyolipomas (AMLs) are benign mesenchymal tumours composed of mature adipose tissue, smooth muscle cells, and thick-walled blood vessels. While predominantly found in the kidney, where they are often associated with tuberous sclerosis, extrarenal AMLs are exceedingly rare. Documented extrarenal sites include the liver, lung, and various other organs, but involvement of the salivary glands, particularly the parotid gland, is exceptionally uncommon, with only a handful of cases reported in the literature.[1] This report aims to describe a unique presentation of AML in the parotid gland, emphasizing its diagnostic considerations and management in a young patient.

CASE PRESENTATION

A girl in her late teens presented to the Ear, Nose, and Throat (ENT) Outpatient Department with a history of a swelling in front of her right ear for the past four years. The swelling had been constant in size for the last two to three years and was associated with intermittent mild pain. The onset was insidious, with gradual progression. There were no aggravating or relieving factors, and no history of trauma. She denied any difficulty in opening her mouth, swallowing, or breathing. Clinical examination revealed a soft, rubbery mass measuring approximately 2×1 cm located anterior to her right ear. The swelling was inconspicuous on inspection (Figure 1).

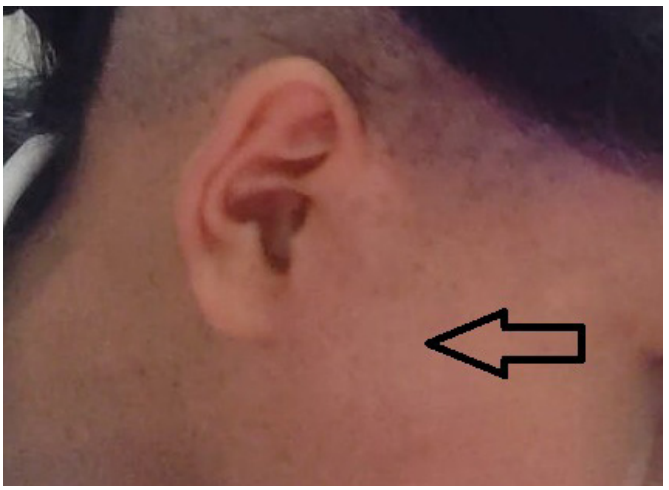

Figure 1: Clinical photograph showing the swelling anterior to the right ear of the patient

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INVESTIGATIONS

Initial ultrasonography of the mass revealed thickening in the fat plane with Doppler flow, suggestive of an arteriovenous malformation. A subsequent Magnetic Resonance Imaging (MRI) of the parotid gland identified vascular malformations with mild involvement of the right parotid gland, possibly connected with the retromandibular vein. Fine-needle aspiration cytology was performed, which supported the diagnosis of a benign tumour.

Following excision of the mass by partial parotidectomy under general anaesthesia, the tissue was sent for histopathological examination. The specimen received consisted of multiple grey-white soft tissue bits, the largest measuring 2×1.7×1.2 cm. Histological sections revealed lobules of mature adipocytes interspersed with vessels, many of which had thick, hyalinized and sclerosed walls. Myotic spindle cells were also observed. Importantly, no atypical mitoses or necrosis were noted. Immunohistochemical analysis showed negativity for Human Melanoma Black-45 (HMB-45).

To exclude the potential presence of tuberous sclerosis, often associated with renal AMLs, additional examinations were conducted. A thorough physical examination did not uncover characteristic skin abnormalities such as hypopigmented spots or angiofibroma-like nodules. Abdominal ultrasound scans also did not reveal any intra- or extra-cranial lesions.

DIFFERENTIAL DIAGNOSIS

The initial presentation of a slowly growing parotid mass in a young patient necessitates a broad differential diagnosis including common benign tumors such as pleomorphic adenoma and Warthin's tumor, as well as rarer vascular malformations or other mesenchymal lesions. Given the ultrasonography findings of fat plane thickening with Doppler flow, an arteriovenous malformation was initially considered. However, the fine-needle aspiration suggesting a benign tumor narrowed the possibilities. The final histopathological diagnosis of angiomyolipoma distinguished it from other common parotid lesions and emphasized the importance of considering uncommon benign mesenchymal tumors in the head and neck region.

TREATMENT

The definitive treatment for the parotid mass was surgical excision. A partial parotidectomy was performed with meticulous identification and careful dissection of the facial nerve to preserve its function.

OUTCOME AND FOLLOW-UP

The partial parotidectomy successfully removed the lesion without complications. At two months post-surgery, a well healed surgical site was demonstrated (Figure 2). The patient was followed up till 18 months after surgery with no signs of recurrence and normal facial nerve function.

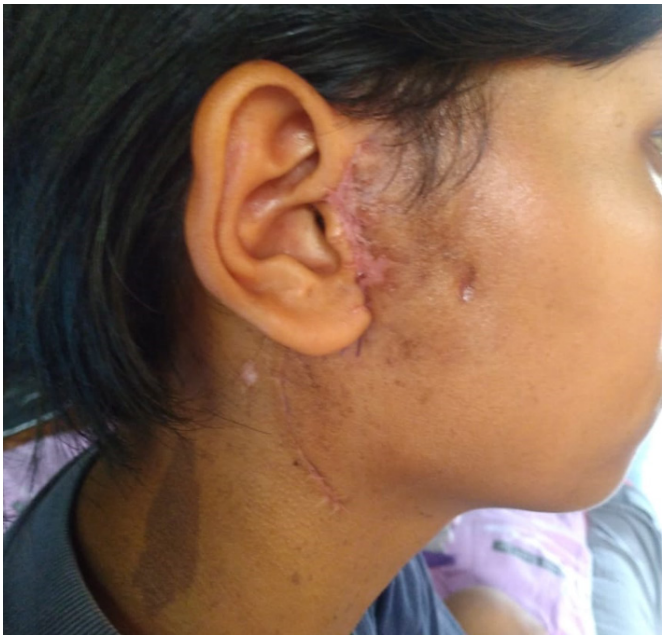

Figure 2: Post-operative images showing a well healed surgical scar

DISCUSSION

Salivary gland tumors constitute approximately 5% of all head and neck neoplasms, with the parotid gland being the most common site.[2] among which the liver is the most frequently recorded. Only rare cases of angiomyolipoma located in the head and neck region (ear and oral and nasal cavity While the majority of parotid tumors are benign, angiomyolipoma (AML) is an exceptionally rare entity in this specific anatomical location, with only a few documented cases reported in the global literature. This extreme rarity makes each reported case, such as the one presented here, a significant contribution to the collective understanding of its clinical presentation, diagnostic challenges, and management. To the best of our knowledge this is the fifth reported case of AML of the parotid gland. It underscores a critical need for pathologists and clinicians to maintain a high index of suspicion for such uncommon benign mesenchymal tumors, even when faced with presentations that might initially suggest more common pathologies (Table 1).

Historically, AML was first meticulously described by Morgan in 1951. For a period, it was broadly considered a hamartoma, a benign focal malformation resembling a neoplasm but resulting from an overgrowth of mature cells and tissues normally present in the affected part. However, contemporary understanding has evolved, and the current consensus classifies AML as a true neoplastic entity, meaning it arises from uncontrolled cell growth, albeit benign. This shift in classification reflects a deeper understanding of its

Table 1: Literature review of all reported cases of angiomyolipoma of the parotid gland

S.No	Author (Year)	Age	Gender	Presenting Symptoms	Degree of Parotid Gland Involvement	Treatment	Follow up
1	Foschini et al. (1999)	68	Female	Slow-growing nodule in right parotid	Well-defined nodule, superficial lobe (2.8 cm)	Total parotidectomy	12 months
2	Guevara et al. (2008)	43	Male	Slow-growing nodule in parotid	Superficial lobe	Partial parotidectomy	Not available
3	Rosado et al. (2010)	73	Female	Painless enlarging parotid mass	Superficial lobe (1.5 cm)	Partial parotidectomy	12 months
4	Uzunoglu et al. (2013)	38	Male	Slow-growing, rubbery mass; 7-year hx	Mass in below superficial lobe (2.5cm)	Partial parotidectomy	18 months
5	Current case (2024)	17	Female	Noted parotid swelling, slow onset, no other symptoms	Involving superficial lobe, well-circumscribed (2 cm)	Partial parotidectomy	18 months

cellular origins and growth patterns. While renal AMLs are frequently associated with tuberous sclerosis (affecting about 50% of patients and often involving multiple organs), extrarenal AMLs, including those found in the parotid gland, are typically not linked to this systemic genetic disorder, as strikingly observed in our case.[2] among which the liver is the most frequently recorded. Only rare cases of angiomyolipoma located in the head and neck region (ear and oral and nasal cavity Nevertheless, due diligence mandates a thorough evaluation to rule out tuberous sclerosis in all AML cases, given its potential multi-systemic implications and the need for long-term genetic counselling and follow-up for affected individuals.

The negative HMB-45 staining in our patient is a notable finding, especially considering that renal and hepatic AMLs frequently exhibit strong HMB-45 positivity in their epithelioid cellular components.[2,3] among which the liver is the most frequently recorded. Only rare cases of angiomyolipoma located in the head and neck region (ear and oral and nasal cavity This differential staining pattern is crucial for diagnosis, as HMB-45 is a marker for melanocytic differentiation, which some AML cells can exhibit. However, it is also well-documented that HMB-45 negativity can occur, particularly in cases where spindle cells predominate histologically.[3] In our specific case, the histological examination revealed only rare epithelioid cells, consistent with the observed HMB-45 negativity and not contradicting the AML diagnosis, but rather providing a more precise sub-classification of this rare parotid variant. Furthermore, it has been reported that HMB-45 positivity in renal AML is often directly related to the presence of tuberous sclerosis[1] and can even serve as an indicator for an increased risk of recurrence[1]. The absence of both HMB-45 positivity and evidence of tuberous sclerosis in our patient aligns with the generally more favorable prognosis seen in isolated extrarenal AMLs.

Preoperative imaging, such as ultrasonography and Magnetic Resonance Imaging (MRI), plays an undeniably crucial role in precisely localizing the tumor and meticulously guiding subsequent surgical planning. However, in rare presentations like parotid AML, these imaging modalities may offer non-specific findings, potentially leading to a broader differential diagnosis, as seen with our patient's initial suggestion of a vascular malformation. Brennan et al. suggested that additional sophisticated imaging, beyond initial assessments, may not be universally required for most parotid gland tumours;[3] however, in diagnostically challenging cases, comprehensive imaging becomes invaluable. Histo-pathologically, the definitive diagnosis hinges on the characteristic features of AML, comprising mature fatty tissue, smooth muscle intermingled with numerous thick-walled vessels[1] (Figure 3 and Figure 4).

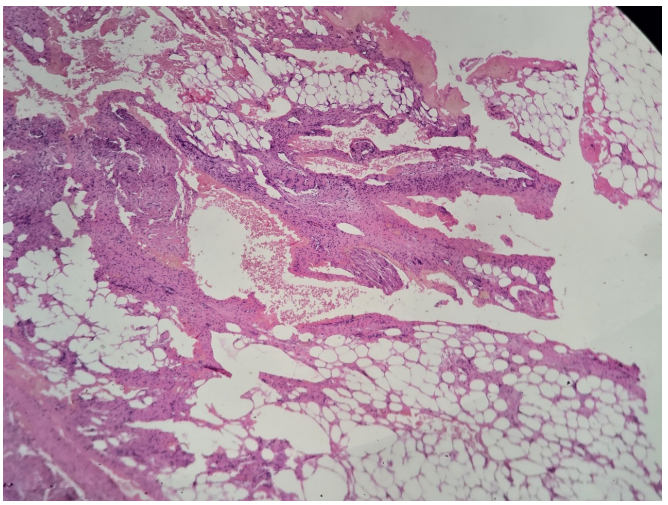

Figure 3: Histopathology image of the resected parotid gland specimen, haematoxylin and eosin stain, showing mature adipocytes, thick-walled vessels, and spindle cells. (Original magnification: ×200)

demonstrates the histopathological findings in our case. It is critical to note the absence of mitosis and necrosis in our specimen, which are important features distinguishing benign from malignant lesions. Although fine-needle aspiration cytology (FNAC) serves as a primary, minimally invasive investigation for differentiating benign from malignant parotid masses[1-3]among which the liver is the most frequently recorded. Only rare cases of angiomyolipoma located in the head and neck region (ear and oral and nasal cavity, its false-negative

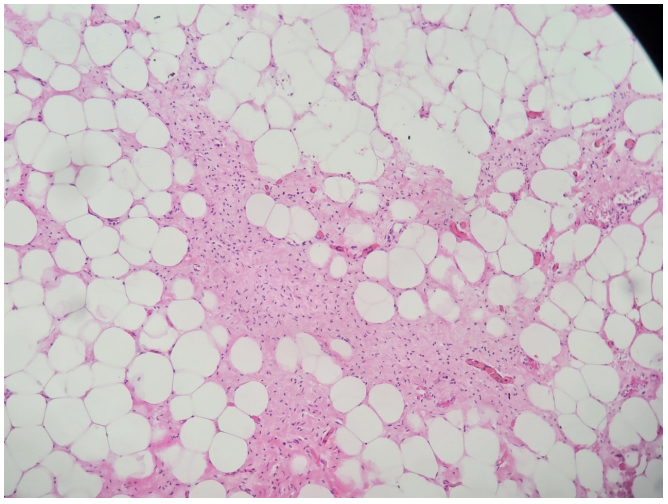

Figure 4: Histopathology image showing mature adipocytes (x400 magnification)

rate, documented to be around 10%, critically highlights the indispensable importance of subsequent histopathological confirmation and a complete immunohistochemical panel in equivocal or unusual cases. Previous reports, including immunohistochemical analyses, have consistently shown AMLs to be strongly positive for smooth muscle actin (a general marker for muscle differentiation) but, as noted, potentially negative for human melanoma black-45 (HMB-45) depending on the cellular phenotype. The prognosis for extrarenal angiomyolipomas, including those situated in the head and neck region, is generally excellent following complete surgical removal. Reassuringly, the literature reports no recurrence after surgical excision for this specific type of tumor in the head and neck, which strongly supports a conservative yet comprehensive surgical approach aimed at achieving complete resection without resorting to excessively radical or disfiguring procedures. Our case robustly aligns with this established management paradigm, demonstrating a successful outcome achieved through partial parotidectomy combined with painstaking facial nerve identification and meticulous dissection, ultimately leading to full preservation of facial nerve function.

LEARNING POINTS/TAKE HOME MESSAGES

- Angiomyolipoma should be considered in the differential diagnosis of parotid gland masses, despite its rarity.
- Histopathological examination with immunohistochemistry is crucial for definitive diagnosis, especially when imaging and fine-needle aspiration results are inconclusive or atypical.
- Extrarenal angiomyolipomas, including those in the parotid gland, generally have a good prognosis with complete surgical excision.
- Careful surgical planning, including facial nerve identification, is essential during parotidectomy for AMLs to ensure complete removal while preserving function.

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