

Membranous-Type Basal Cell Adenoma of the Parotid Gland in a 16-year-old Male: A Case Report and Review of the Literature

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Abstract

Salivary gland tumors (SGTs) encompass a broad category of neoplasms that can be benign or malignant with significant variation in clinical and histopathological presentation. Among SGTs, basal cell adenomas (BCAs) represent a rare subset that is predominantly found in the parotid gland of women over the age of 50. In this case report, we describe an atypical case of a membranous-type BCA identified in the left parotid gland of a 16-year-old male. Diagnostic measures included contrast-enhanced computed tomography (CT) imaging and fine-needle aspiration (FNA) biopsy which ultimately led to a left superficial parotidectomy. Surgical pathology confirmed a 1.7 cm membranous-type BCA with negative margins. Due to the potential for recurrence associated with membranous-type BCA, long-term monitoring was recommended. Here we discuss the rarity of this diagnosis in a male adolescent, review current practices for management of BCAs, and identify gaps in knowledge and standardized clinical guidelines for atypical presentations.

KEYWORDS: basal cell adenoma, membranous-type basal cell adenoma, parotid gland tumor, parotid neoplasm, pediatric salivary gland tumor, salivary gland neoplasm, salivary gland tumor

1 Introduction

Salivary gland tumors (SGTs) are rare, accounting for fewer than 3% of neoplasms of the head and neck [1]. Incidence of SGTs is particularly low in the pediatric population, in which they represent fewer than 5% of reported cases [2]. In pediatric and adolescent populations, parotid tumors account for approximately 90% of salivary gland masses [3], and the differential diagnosis is broad, including Warthin's tumor, adenoid cystic carcinoma, basal cell adenocarcinoma, and pleomorphic adenoma [4-6]. A small Brazilian study of pediatric SGT cases found that 50.9% of tumors were benign with pleomorphic adenomas accounting for the majority of those cases. The utility of this study is limited by its age, as the reported cases occurred in the 1950s [2]. A more recent U.S. study identified a higher percentage of cases as benign (78%), but also found pleomorphic adenomas to be the most common neoplasm (92.3%) [7]. Nevertheless, clinicians should be thorough, open to alternative diagnoses, and have a low threshold for suspicion of malignancy in pediatric and adolescent patients.

It is important to note that while many pediatric SGTs are benign, such tumors are five times more

likely to be malignant than in the adult population [7-9]. Overall, approximately 65% of SGTs are benign, while 35% are malignant, a finding that is generally consistent with historical data and previous studies [10]. Among SGTs, basal cell adenoma (BCA) is a rare subset of tumor that accounts for only 1-6% of all SGTs [10,11]. BCAs frequently present as firm, mobile, painless, and slow-growing masses [12,13]. They arise predominantly in the parotid gland, although they can also occur in the submandibular glands and other intraoral locations [4]. BCAs are classified into four histopathological types: solid, tubular, trabecular, and membranous [14]. The solid type is the most common, but it is not unusual for BCAs to present as a mixture of two or more histological subtypes [4].

While BCAs are benign, approximately 5% can undergo malignant transformation, and this phenomenon is most often observed in the membranous-type [15]. Membranous-type BCAs are non-encapsulated, multinodular, and intermittently multifocal [4], with a notably high recurrence rate of 25-38% [16,17]. The potential for recurrence further distinguishes membranous-type BCAs from other BCA subtypes, which typically do not recur [4,15]. BCAs most often occur in women, with estimates indicating that women account for up to 7% of re-

ported cases [4,5]. However, some studies report nearly equal incidence among the sexes, particularly for membranous-type BCAs [4]. Typically, BCAs occur in patients in their late 50s to early 60s, and they are relatively uncommon in children and young adults [4,5].

While salivary gland masses can generally be identified on physical exam or imaging, diagnosis requires biopsy, which can often be achieved by fine needle aspiration cytology (FNAC). FNAC is a cost-effective and minimally invasive method for diagnosing SGTs, though in some cases excisional biopsy may be required for definitive diagnosis. Treatment generally involves surgical excision. Due to relatively high rates of recurrence in membranous-type BCA, aggressive surgical management followed by close observation and regular clinical monitoring is recommended [4,15].

2 Case Description

Here, we report the unusual occurrence of membranous-type BCA of the left parotid gland in a 16-year-old Caucasian male. On October 9, 2023, he presented to the otolaryngology office with the chief complaint of a mass on his left cheek. He reported that he first noticed the mass two years prior, and that it had been growing slowly since that time. He complained of increasing pain with mastication and stated that he had tried using sialagogues for symptom improvement, without relief.

Physical exam findings at the time of presentation were notable for a smooth, slightly irregular, firm, and mobile mass that was palpated in the left parotid gland, just anterior to the tragus. Physical exam findings suggested that the mass was superficial, and estimated it to measure approximately 2 cm x 1.5 cm. Some facial asymmetry was noted around the left corner of the mouth, but muscle strength was normal. The remainder of the patient's physical exam and his social and medical history were unremarkable. Family medical history was notable only for breast cancer in a secondary relative. Given the patient's symptoms and physical exam findings, the decision was made to order computed tomography imaging and perform FNA biopsy.

Computed tomography imaging revealed a 1.7 cm mass in the left parotid gland, as seen on transverse (Figure 1) and coronal (Figure 2) sections. FNA biopsy was performed on October 24, 2023, with results subsequently showing atypical cells suspicious for a neoplastic process without overt malignant features. Given the uncertain diagnosis at the time of his surgical consultation on November 2, 2023, the patient and his family elected to schedule the left

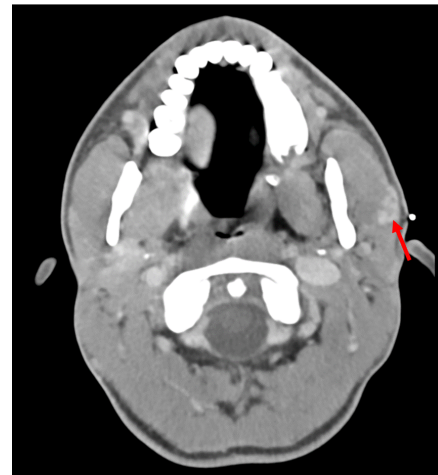


Figure 1: Contrast-enhanced CT image (transverse section). Left parotid mass identified by arrow.



Figure 2: Contrast-enhanced CT image (coronal section). Left parotid mass identified by arrow.

parotidectomy for February 2, 2024 (approximately four months after initial consultation).

The patient was taken for surgery where the mass was found to be deep to the mid facial branches of the facial nerve, making this mass technically within the deep lobe of the parotid. Resection of the mass and preservation of the facial nerve was successful. Since pathology was unknown, total parotidectomy was not performed.

Gross examination of the tumor by pathology revealed a 1.7 x 1.7 x 1.2 cm tan, firm, and partially well-defined nodule. In addition, the tumor demonstrated a multilobulated appearance with nodules of neoplasm branching off the main tumor. Final histopathology confirmed the diagnosis of basal cell adenoma, membranous-type, with noted complete excision and two reactive lymph nodes (Figures 3 & 4).

On February 5, 2024 (three-days post-resection), the patient was seen in clinic for drain removal. He reported the expected peri-incisional numbness and some numbness of the left ear lobule. Facial nerve function was normal. On February 9, 2024, he re-

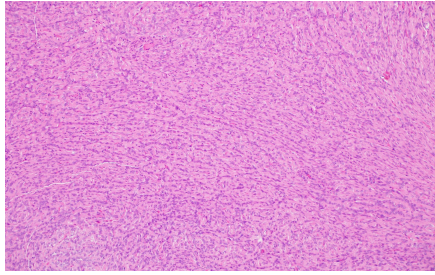


Figure 3: Histological section showing membranous basal cell adenoma. (H&E stain x10).

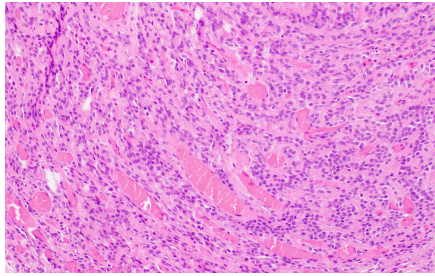


Figure 4: Histological section showing membranous basal cell adenoma. (H&E stain x20).

ported persistent swelling and mild pain, but cheek numbness was improving and there was no facial weakness. It was decided that he would follow up annually for tumor recurrence surveillance. The patient returned for one-year follow up on February 5, 2025, and had no evidence of tumor recurrence.

3 Clinical Recommendations

Although membranous-type BCA is often treated with total parotidectomy due to its high recurrence rate [12], our patient underwent superficial parotidectomy because the preoperative biopsy was suspicious for a neoplastic process without overt malignant features. The diagnosis of BCA was only made on the post-surgical pathology, underscoring the importance of adjusting management, which in this case required long term monitoring for recurrence. No formal guidelines or recommendations exist for postoperative surveillance in BCA. Prior reports of BCA have described annual follow-up as a reasonable approach to monitor for recurrence [18,19].

4 Discussion

The patient's final diagnosis was membranous-type BCA, characterized by a multilobulated appearance and a high rate of multicentricity. While BCAs predominantly affect women over the age of 50, there are a few documented cases of certain types of BCAs in younger males. However, our review

of the literature did not yield any other cases of membranous-type basal cell adenoma in a patient of similar demographics [7]. We found three reports of tubulotrabeular-type basal cell adenomas in adolescent and young adult males: one in a 14-year-old male, one in a 22-year-old male and one in a 25-year-old male [1,7,12]. Our search also yielded a case of a 25-year-old male with a BCA of the right parotid gland that was determined to be of a mixed membranous-tubulotrabeular pattern [8]. However, to our knowledge, this is the first description of a purely membranous-type BCA of the parotid gland in an adolescent male. Given our patient's age and rarity of diagnosis, we believe this case is a notable contribution to the existing literature.

Our case highlights the importance of considering BCA in the differential diagnosis of salivary gland neoplasms in pediatric patients, despite the uncommon occurrence of these tumors in this demographic. Given the rarity of BCAs in male adolescents, awareness of this diagnosis may aid clinicians when encountering cases with atypical features or unexpected age groups. Furthermore, the management of BCAs in this population warrants careful consideration because this subtype has the highest rate of recurrence and as previously discussed, some cases of membranous-type BCAs have demonstrated malignant features or potential for malignant transformation into basal cell adenocarcinoma [20,21].

5 Conclusion

Limited pediatric and adolescent data are available in the literature, both in terms of BCAs, and SGTs more broadly, emphasizing the need for further research to elucidate the epidemiology, underlying pathophysiology, optimal diagnostic strategies, and treatment approaches tailored to this group. We encountered limited datasets from single centers or health networks, but there is a need for additional data collection and analysis, particularly with respect to large-scale data collection in pediatric SGTs or meta-analyses of small-scale datasets and studies.

Continued surveillance and long-term follow-up of patients diagnosed with membranous-type BCAs are imperative to monitor for recurrence. Further incorporating males and pediatric patients in discussions regarding salivary gland neoplasms can both improve and ensure timely diagnosis, appropriate management, and superior patient outcomes.

Declarations

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Ethics Approval and Consent to Participate

Not applicable.

Consent for Publication

The described patient and his parent provided verbal consent to the description and publication of his case to Dr. Jeffrey Wilcox.

Data Availability

Not applicable.

Conflicts of Interest

The authors declare that they have no competing interests.

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Author Contributions

All authors contributed to the history gathering and written summary of the patient case. Woodring, Yang, and Christensen contributed to the literature review. Wilcox provided the necessary review, editing, and oversight to ensure accurate description of the case.

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