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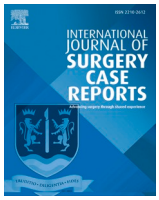

Case report

Surgical management of basal cell adenoma of the parotid gland: A case report and review of the literature

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ABSTRACT

Introduction and importance: Basal cell adenoma (BCA) is a rare, benign tumor of the salivary gland. It has distinct histologic features that are different from mixed tumors of the salivary gland. Often times, it occurs in the parotid gland. Usually it appears as a hard, slowly growing, asymptomatic lump that may exist for years prior to treatment. Pathologically, BCA tumors demonstrate a monomorphic basaloid cell population devoid of myxochondroid components. This rather uncommon tumor's clinical appearance, pathological characteristics, differential diagnosis, and available treatments have been briefly described.

Case presentation: A 52-year-old female patient presented with a longstanding, gradual facial asymmetry that resulted from a mass of parotid region. She had no other symptoms. Radiology and cytology test results were suggestive of a benign salivary gland tumor of the parotid gland. A superficial parotidectomy was successfully done with a preservation of the facial nerve. Histopathology reported concluded the diagnosis of BCA trabecular subtype. During the 12-month follow-up period, the tumor did not recur, and there was no postoperative impairment of the facial nerve.

Clinical discussion: Imaging studies, including CT scans and MRI, are helpful in determining the tumor's size, location, and relationship to adjacent structures. The definitive diagnosis is achieved through histopathology. This clinically asymptomatic mass requires early recognition, a prompt therapeutic intervention and long-term follow-up.

Conclusion: As clinicians, it is critical that we have a thorough understanding of clinicopathology in terms of clinical presentation, pathology, diagnosis, treatment, and prognosis of this rare tumor.

1. Introduction

A salivary gland tumor is an abnormal growth that occurs in one of the salivary glands, which are responsible for producing saliva. These neoplasms can be benign or malignant, and they include a group of various histopathological disease entities that affect different sites within the region of the head and neck [1]. Basal cell adenoma (BCA) is an extremely uncommon benign salivary gland tumor constituting approximately 1 % to 3 % of all salivary gland epithelial tumors [2]. BCA is a distinctive entity recognized in the classification of tumors of the head and neck by the World Health Organization [1]. Herein, we present a case of a 52-year-old female who attended our facility with an asymptomatic parotid mass that was histopathologically identified as BCA. We also briefly reviewed the pertinent literature and discussed the diagnostic methods and treatment approaches of this uncommon salivary gland tumor. This work has been reported in line with the SCARE 2023 criteria [3].

2. Case report

A 52-year-old female, medically free and with no identifiable allergies, presented to the oral and maxillofacial clinic of our facility with a painless mass in her right parotid region that had been present for a 6-year period. On further examination, the patient disclosed having no associated symptoms besides the noticeable facial swelling on her cheek. There was no history of dyspnea or dysphagia. The patient denied any

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history of pain, skin color changes, numbness, facial weakness, xerostomia, odynophagia, or other obstructive symptoms.

On physical examination, the patient was vitally stable. An isolated single, firm, non-tender mass on her right parotid region was evident (Fig. 1A). The mass was mobile and not fixed to the underlying structures. There was no cervical lymphadenopathy. The facial nerve proved intact, and the rest of the examination pertaining to the head and neck was essentially unremarkable. Her blood workup test results were within the normal ranges. A computed tomography (CT) scan of the head and neck result described a unilateral mass of soft tissue density in the right parotid gland arising from the superficial lobe (Fig. 1B). Magnetic resonance imaging (MRI) demonstrated a well-circumscribed tumor, which showed a cystic and solid. The mass had a diameter of 4.5 cm in its greatest dimension. A fine needle aspiration (FNA) biopsy showed monomorphic basaloid cell population with minimal nuclear atypia, mitosis, or any tumor necrosis. The diagnosis of a benign cellular basal cell neoplasm was considered. The patient was scheduled for surgery after securing her authorization.

2.1. Surgical procedure

The treatment of choice was the right superficial partial parotidectomy under general anesthesia. The patient received 1 g of Cefazolin IV as prophylaxis 24 h prior to surgery in order to reduce the risk of postoperative wound infection. The operation was done by an experienced team of oral and maxillofacial surgeons. The endotracheal tube was fixed to the contralateral side. The head was turned away from the side being operated on, and the neck was somewhat stretched. A modified Blair incision that begins at a preauricular skin crease around the lobule posteriorly and extends inferiorly into a cervical skin crease was made. After incising the dermis and platysma muscle, an anterior skin flap was raised to expose the tumor that was fixed to the superficial capsule of the parotid gland (Fig. 2A–B). All the branches of the facial nerve were identified and visualized. Care was taken to avoid the damage to branches of the facial nerve. The tumor was removed en block. A drain was kept in situ at the end of the procedure. In our setting, intraoperative diagnostic services, including frozen sections, are not available. The surgery went smooth and uneventful, and the surgical specimen (Fig. 3A) was sent for histopathological examination. She was later kept on Paracetamol to control pain.

Histopathology report described well-circumscribed, encapsulated solid and cystic tumor components (Fig. 3B). The individual tumor cells were basaloid with eosinophilic cytoplasm, with indistinct cell borders and round to oval nuclei, distributed in a trabecular pattern (Fig. 4). The pathologist confirmed the definitive diagnosis of BCA trabecular type of the parotid gland. The patient had a quick recovery post-operatively,

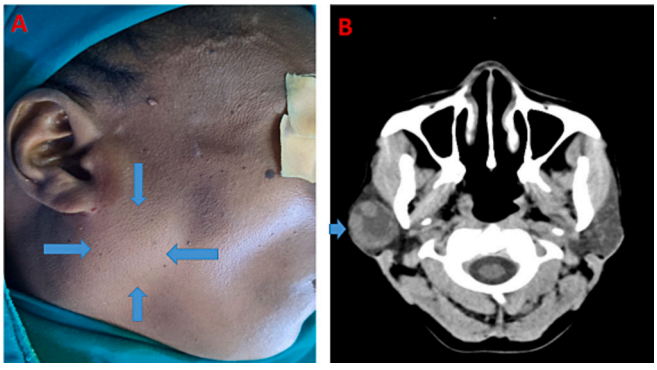

Fig. 1. Clinical examination of the patient highlighting a right parotid gland mass (blue arrows) (A); an axial CT scan image of the patient demonstrating the right parotid gland mass (blue arrow) arising from the superficial lobe (B). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

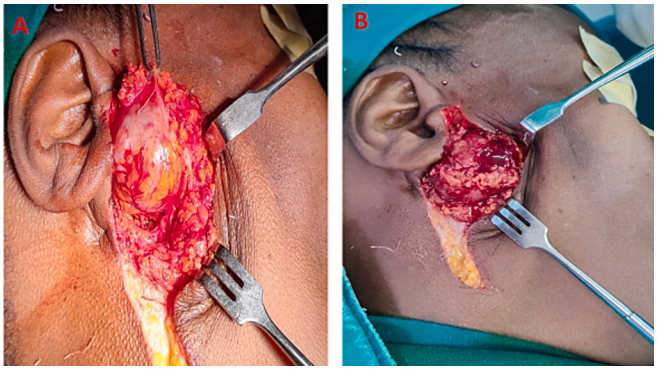

Fig. 2. An incision on the dermis and platysma muscle exposing the tumor on the superficial capsule of the parotid (A); precautions were taken to avoid the damage to branches of the facial nerve (B).

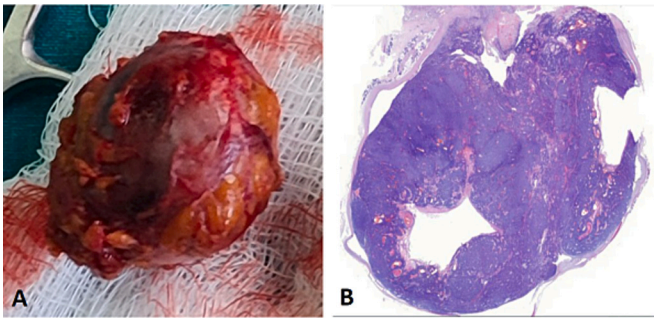

Fig. 3. A gross appearance of the excised tumor that was submitted for histopathology evaluation (A); histopathology of BCA demonstrating an intact encapsulated benign tumor with solid and cystic components, H&E staining at a low power magnification (B).

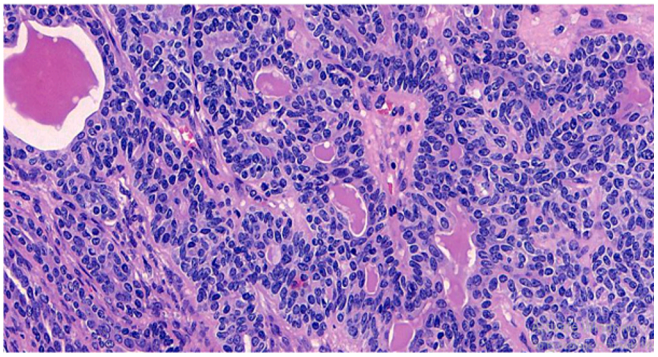

Fig. 4. A high-power magnification of BCA consisting of basaloid cells with eosinophilic scant cytoplasm, indistinct cell borders, and round to oval nuclei distributed in trabecular pattern. The tumor demonstrates peripheral palisading of cuboidal to columnar cells with occasional variable sized cystic spaces.

and she reported no new complaints in the subsequent follow-up clinics. The patient confirmed to have resumed optimal functional and cosmetic effects. To date, at least 12 months have passed, and the patient remains disease-free. The plan is to keep her on long-term monitoring given the rare but possible recurrence or malignant transformation of BCA.

3. Discussion

Our report describes an uncommon case of BCA of the parotid gland in a 52-year old female patient that was successfully managed at our

institution. This tumor, for the first time, was described by Kleinasser et al. [4]. Due to the rarity of this entity, the precise epidemiological data are lacking. It is estimated to account for about 1–3.7 % of all tumors of the salivary gland [1]. The tumors mostly affect adults in their 5th to 7th decade of life, with a 2:1 female preponderance. This is similar to our patient, who was female, 52 years old.

As was mirrored in our patient, the majority of BCA tumors develop from the parotid gland (75 %), followed by the submandibular gland (5 %). Rarely, it does occur in the minor salivary glands [5,6]. Typically, BCA tumors are encapsulated, well circumscribed, solitary, and mobile. The lesions are usually firm, but at times they can be cystic, as observed in our patient. Unfortunately, the cause as well as the risk factors for BCA is poorly understood. However, genetic aberration has been described in some cases of BCA. For instance Trisomy 8 and chromosomal 13 alterations have been reported in some cases [1], while the 7;13 translocation and/or inv. were present in another case [7]. Cytogenetic alterations have been only rarely characterized in non-membranous basal cell adenomas [7].

Radiological tests such as MRI and CT scans are essential in determining the tumor's size, location, and relationship to adjacent structures. On MRI, parotid gland BCA usually demonstrates a well-defined mass lesion that can be at the medial portion of the deep lobe of parotid gland. The mass can be a cystic and solid tumor with a thin fibrous capsule, composed of central cystic and peripheral soft-tissue components [8]. In our patient, both the CT scan and an MRI of the head and neck highlighted a right parotid gland mass arising from the superficial lobe. The radiological findings for BCA in the parotid gland in our patient corroborate with other studies described in the literature [7,8]. However, some authors reported that these lesions do not have specific features [9]. Because of prognostic implications, the potential differential diagnoses such as basal cell adenocarcinoma, basaloid squamous cell carcinoma, and adenoid cystic carcinoma should always be considered and adequately excluded before establishing the diagnosis of BCA of the salivary gland in the oral and maxillofacial region [2].

The definitive diagnosis of BCA is established by the histological study, with the biopsy being the gold standard for rendering the most accurate diagnosis. However, some scholars have documented the utility of cytology in diagnosing these rare tumors [10]. In our case, FNA was initially applied, whose results were suggestive of a benign salivary gland lesion. The definitive diagnosis was later confirmed by an excisional biopsy histopathology evaluation. Morphologically, BCA neoplasms are characterized by the basaloid cell population and absence of the myxochondroid stromal component that is demonstrated in mixed tumors such as pleomorphic adenoma [1]. BCA has four histological growth patterns, which are trabecular, solid, membranous, and tubular patterns [1]. The ductal cells stain positive with keratin, CEA, and CD117 immunohistochemical markers, while the basaloid cells express immunopositivity with S100, actin, vimentin, and p63 [11].

As is the case in other benign salivary gland tumors, surgical removal with a cuff of normal salivary tissue is the recommended treatment approach for these tumors [12,13]. This could be either suprafacial or total parotidectomy depending on the location, size, and histological subtype of the lesion. For instance, total parotidectomy rather than suprafacial parotidectomy has been proposed in the membranous type of BCA. In our case, superficial parotidectomy along with the tumor excision was done without injury to the facial nerve. The capsule of the tumor remained intact (Fig. 3B). It is important not to disrupt the capsule in order to minimize the risk of recurrence, which is observed on rare occasions. Despite this benign behavior, it is imperative that the patient is kept on a long-term follow-up in order to detect recurrences in a prompt time. These lesions usually do not recur after surgical removal, as was seen in our patient during the12-month follow-up. However, it is important to keep the patients on long-term monitoring given the rare but possible recurrence or malignant transformation in certain subtypes of BCA in which up to 25 % recurrence rates have been reported [2]. Malignant transformation is more common in the membranous type (4 %) than in the other types; thus, total parotidectomy rather than superficial parotidectomy is proposed in the membranous type of BCA [14].

4. Conclusion

BCA of the parotid gland is a rare tumor with a wide list of differential diagnoses. Its uncertain clinical picture as an asymptomatic mass requires early recognition, histopathological diagnosis, and therapeutic intervention. A long-term follow-up is critical given the unlikely but possible recurrence or malignant transformation in certain subtypes of BCA. As clinicians, it is critical that we have a thorough understanding of clinicopathology in terms of clinical presentation, pathology, diagnosis, treatment, and prognosis of this rare tumor.

Author contribution

All authors made substantial contributions to this work.

Kanankira A. Nnko[1,2], Raphael T. Pima[1] and Alex Mremi[2,3,4]

Kanankira A. Nnko: Conceptualization, study design, data curation, patient management, and prepared initial manuscript version.

Raphael T. Pima: Involved in the patient management, collected the data, and reviewed, and approved the final manuscript draft.

Alex Mremi: Conceptualization, and also performed histopathological analysis and prepared the final manuscript draft.

Guarantor

Alex Mremi.

Research registration number

Not applicable.

Informed consent

Written informed consent was obtained from the patient to publish this case report and accompanying images. On request, a copy of the written consent is available for review by the Editor-in-Chief of this journal.

Ethical approval

The ethical approval in this study was exempted on August 10, 2024 by the Research Ethics Committee, Kilimanjaro Christian Medical University College, Box 2210 Moshi, Kilimanjaro, Tanzania, email: info@kcmuco.ac.tz; web: www.kcmuco.ac.tz; Tel: +255272753616); because individual case reports do not require ethical approval.

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Conflict of interest statement

All authors have declared that no competing interests exist.

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