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Case Report

Adenoid (Acantholytic) squamous cell carcinoma of the alveolar ridge: A rare case report

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ABSTRACT

Adenoid squamous cell carcinoma (ASCC) is a one of the histological variants of oral squamous cell carcinoma with a rare occurrence of less than 4% of all the cases. The tumour has a slight male predilection with lower lip being the most affected site. ASCC is said to have a varied biologic behaviour which accounts for its ability to metastasize to distant sites and hence has poor prognosis. The histopathology shows numerous duct-like structures with central acantholytic cells. Distinct cytological atypia is seen which aids in diagnosis of SCC. Special stains help in differentiating the tumour from other variants of SCC. The presented case of a 70-year-old female emphasizes the importance of histopathogical examination of the unusual and rarely observed ASCC which can be missed due to similarities with other entities. Recurrence rates being very high, proper treatment starting with definitive diagnosis is imperative.

1. Introduction

Of all the cancers affecting the oral cavity, more than 90% are squamous cell carcinomas occurring in the mucous membranes of the mouth and oro-pharynx. As of 2006, oral squamous cell carcinoma (OSCC) accounted for 2,75,000 cases and 1,28,000 deaths annually and has set off as a global health concern.[1]

Conventional OSCC can present as diverse variants which constitute about 10-15% of all squamous cell carcinomas. These variants include verrucous carcinoma, basaloid SCC, adenoid SCC, adenosquamous carcinoma, spindle cell/sarcomatoid carcinoma, papillary SCC and undifferentiated SCC.[2] Among these, basaloid SCC and adenoid SCC guide the prognosis and biological behaviour, and hence it is of utmost importance to differentiate these histological variants from the conventional OSCCs.[3]

Adenoid (acantholytic) squamous cell carcinoma (ASCC) represents 2-4% of all SCCs which mainly affects the sun-exposed areas of the skin.[4] The first case of oral cavity-associated ASCC was reported by Goldman et al in the year 1977.[5]

Herein, we present a case of a 70-year-old female patient which was later diagnosed as ASCC. This case report intends to emphasize the occurrence of a rare histopathological variant of OSCC and its association with the biological behaviour of the tumour.

2. Case Report

A 70-year-old female patient reported with a complaint of painful swelling in the left maxillary posterior region which was present for the past 3 months. The teeth associated with the swelling that is tooth number 23,24,25,26 were extracted due to mobility one and a half months back, yet the swelling did not reduce. Patient's medical history was non-

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contributory. However, she gave a betel-nut chewing habit (5 times per day since 50 years).

On clinical examination, a solitary well defined swelling of size approximately 4.0x3.0cm was noted on the left palatal region extending antero-posteriorly from residual alveolar ridge from 23 to 27 area and medio-laterally extending from the mid-palatine raphe till residual alveolar ridge on the buccal aspect. The swelling appeared oedematous and inflamed with pus discharge associated with ulcer formation along with bleeding on the buccal aspect which was tender on palpation and soft in consistency (Figure 1 A and 1B). A provisional diagnosis of 'infected dento-alveolar abscess' was made. Clinical differential diagnosis of osteomyelitis and Carcinoma of residual alveolar ridge was given. An incisional biopsy was performed under local anaesthesia and the tissue was sent for pathological evaluation.

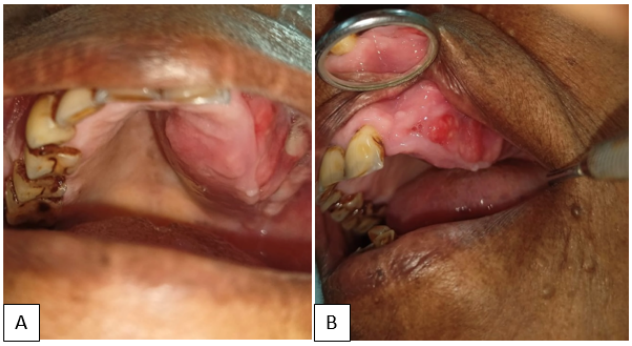

Fig. 1: A: A well-defined swelling noted on the palate; **B:** Ulceration noted on the buccal aspect of the alveolar ridge

On gross examination, the specimen received was whitish red in colour with largest tissue bit measuring 1.0x0.5x0.5cm (Figure 2).

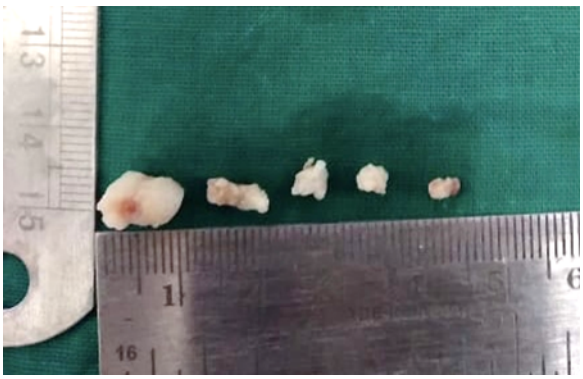

Fig. 2: Macroscopic gross picture

The microscopic examination of the Haematoxylin and Eosin stained section revealed invasion of the epithelial cells into the connective tissue. Multiple keratin pearls were noted in the sub-epithelial region (Figure 3 A). Multiple islands resembling duct like-structures were evident scattered all over the connective tissue (Figure 3 B). These islands showed features of dysplasia in the form of cellular and nuclear pleomorphism, increased nuclear/cytoplasmic ratio with prominent nucleoli and numerous mitotic figures. The duct-like or pseudo glandular structures were lined by a single layer of flat to polygonal cells with central acantholysis (Figure 3 C). Periodic acid-Schiff (PAS) and mucicarmine stains confirmed these structures to be non-glandular without any mucinous components (Figure 4 A and 4B). The surrounding connective tissue was fibro-cellular with chronic inflammatory cell infiltrates. No perivascular or perineural invasion was evident. Contemplating all the histological features, a final diagnosis of adenoid (acantholytic) squamous cell carcinoma was made. The patient was referred to a cancer centre because of the extensive nature of the lesion and age.

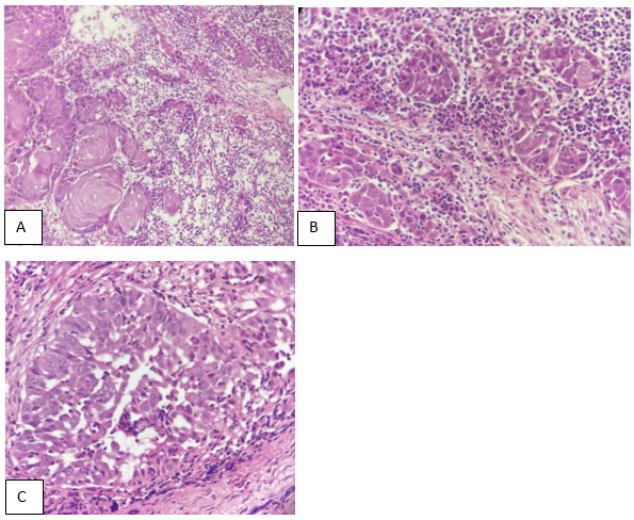

Fig. 3: A: Keratin pearls noted along with some epithelial islands (Hematoxylin and eosin stain, original magnification x200); **B:** Multiple duct-like structures noted separated by fibrous stroma (Hematoxylin and eosin stain, original magnification x200); **C:** Adenoid area with central acantholytic cells (Hematoxylin and eosin stain, original magnification x400)

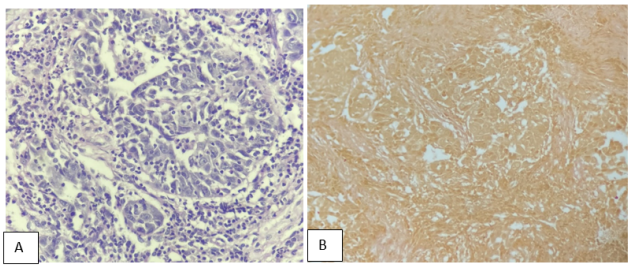

Fig. 4: A: Negative PAS reaction (Periodic Acid Schiff's stain, original magnification x400); **B:** Negative mucicarmine reaction (Mucicarmine stain, original magnification x200)

3. Discussion

Adenoid (acantholytic) squamous cell carcinoma (ASCC) was first described by Lever in 1947 and now it is a well-recognized histopathological variant of squamous cell carcinoma.[6] It is most commonly seen in the sun-exposed areas of the head and neck region where infra-red radiation is said to be the etiological factor.[7] The conventional OSCC has alcohol and tobacco usage as the prime etiological agents. However, due to less number of cases and studies on intra-oral ASCCs, no definitive etiology has been described yet.[8] The loss of cellular adhesion in ASCC is attributed to the desmosomal defects especially desmoglein 1,2 or desmoplakin or both are affected.[9]

Around 50 cases of ASCC affecting the oral cavity have been reported till now with lower lip being the most affected site and with more male predilection.[4,10] However in our case, the affected was a female with alveolar ridge being the site of occurrence.

ASCC is otherwise known as squamous cell carcinoma with gland-like (adenoid) structures, pseudoglandular squamous cell carcinoma.[11]It is also often known as pseudovascular adenoid squamous cell carcinoma or pseudoangiosarcomatous carcinoma because of their similar histological features. Formation of the anastomosing channels and spaces in ASCC closely ressembles that of angiosarcoma, hence the names.[12]

The histological differential diagnosis of ASCC include conventional SCC, adenosquamous cell carcinoma (ASC), adenocarcinoma of salivary gland and angiosarcoma.[10] The conventional SCC lacks the duct-like structures with central acantholysis, ASC on the other hand shows true glandular differentiation with positivity for mucin seen in special stains like PAS, mucicarmine and alcian blue. Although, salivary gland tumours like adenocarcinoma and mucoepidermoid carcinoma can be confused with ASCC but presence of mucinous material helps in differentiation. While angiosarcoma shares few similarities with ASCC histologically, the clinical and immunostaining profile aids to demarcate between the two lesions (CD31, CD34, Fli-1 protein are highly specific markers of angiosarcoma).[4,8] The present case had to be differentiated from ASC and the conventional SCC. Employing the special stains helped to rule out ASC and presence of acantholytic cells and duct-like areas rules out the latter.

ASCC of the oral cavity is said to have a poor prognosis. The detachment of the neoplastic cells due to process of acantholysis is considered to be the reason for distant metastases and subsequent aggressive behaviour and poor outcome.[9] The 30-month survival was 20.4% and 46-month survival was 0% in a study conducted which incorporated and followed 13 cases of ASCC of oral cavity.[6] Despite the less number of cases available, owing to the aggressive behaviour, radical surgery along with adjuvant therapies is the treatment of choice.[10]

4. Conclusion

The comprehension of histopathological variants of oral squamous cell carcinoma is very critical. Rare variants like ASCC could be missed due to lack of knowledge of the microscopic features. Accurate reporting of ASCC is paramount so as to devise a best possible treatment protocol in order to minimize the rate of metastasis and reoccurrence thereby improving the prognosis.

5. Source of Funding

None.

6. Conflict of Interest

None.

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