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LOCALIZED GINGIVAL ENLARGEMENT A RARE ENTITY OF SARCOMATOID CARCINOMA- A CASE REPORT

Mangesh B. Phadnaik., Ripunjay K. Tripathi*., Meghna Nigam., Ankit J. Solanki., Samit N. Javiya and Sharad Agrawal

Department of Periodontology, Government Dental College & Hospital, Nagpur

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ABSTRACT

Introduction: Gingiva is a very common site of oral tumors, which may be benign or malignant with or without metastasis. Common to rare malignant neoplasms of gingiva have been reported in the literature. Spindle cell carcinoma is a rare and peculiar biphasic malignant neoplasm that occurs mainly in the upper aero digestive tract. It consists of sarcomatoid proliferation of pleomorphic spindle cells in squamous cell carcinoma. It is considered potentially aggressive in its biological nature with a high incidence of metastases

Case presentation: A 51-year-old male patient reported with a chief complaint of swelling in the Lower left tooth region. The patient gave history of painless slow growing swelling associated with tooth number 33, 34 since 8 months with no history of treatment in the past. The excised tissue was fixed in 10% formalin solution and submitted for histopathological examination.

Results: The postoperative healing was uneventful at 2 week time interval at which time the histopathological report was also received and the lesion to our surprise turned out to be malignant i.e. sarcomatoid carcinoma.

Conclusion: The seemingly non threatening gingival enlargement may sometimes be a deceptive and rare entity challenging to the health science fraternity because of its unusual appearance and behaviour. Hence, thorough clinical and histopathogical investigation is a must.

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INTRODUCTION

Spindle cell carcinomas are rare and distinct malignant neoplasms. They originate from epithelium and show spindle cell appearance. As they are a rare entity their diagnosis is challenging for the pathologists. According to Ellis, the Spindle cell carcinomas are most commonly seen in lower lip, tongue and alveolar ridge but it accounts for less than 1% of the malignant tumours in the oral cavity. Thus gingival involvement seems to be a rare site. The state of the malignant tumours in the oral cavity.

It may be reactive or neoplastic, mesenchymal or epithelial in nature. ⁵ However, the WHO classification of tumors of the oral cavity and oropharynx has placed this disease entity under malignant epithelial tumors of squamous cell carcinoma. ⁶

In this report, we will describe a case of spindle cell carcinoma involving the mandible with an unusual clinical presentation.

*Corresponding author: Ripunjay K. Tripathi Department of Periodontology, Government Dental College & Hospital, Nagpur

Clinical Presentation

A 51-year-old male patient reported to the Department of Periodontology, Government Dental College & Hospital, Nagpur, India, in August 2017, with a chief complaint of swelling in the lower left tooth region. The patient gave history of painless slow growing swelling associated with tooth number 33,34 since 8 months with no history of treatment in the past. Patient had habit of placing the tobacco quid in the left vestibular area 3-4 times a day since 12 years.

The medical, dental and family history was non-contributory and he was in good health. The extra-oral examination revealed no facial asymmetry, no lymphadenopathy and no abnormality of the TMJ. There was no history of weight loss, coughing, persistent mouth-sore, difficulty in swallowing, numbness of the tongue (or any other area of mouth) or voice change.

Oral hygiene status of the patient was poor. On intra-oral clinical examination, well defined, solitary, reddish pink, non-ulcerative, oval pedunculated swelling with smooth surface involving gingiva of 33,34 was seen. The swelling measured approximately $3 \text{ cm} \times 1.5 \text{ cm}$ (Fig. 1). Upon palpation, the lesion was non-tender and firm in consistency around the

borders and soft at the center of the lesion. The lesion adhered to gingiva but was not fixed to the underlying bone. There was pus discharge and bleeding on probing associated with the lesion. The swelling was non-fluctuant, non-compressible and non-pulsatile. Grade 1mobility was seen with 34.



Fig 1 Pre-operative view of the Lesion in #33 #34 region

Differential Diagnosis

On the basis of clinical and radiological findings, differential diagnosis of peripheral fibroma, peripheral giant cell granuloma, pyogenic granuloma, or neoplastic lesion was given.

Investigation

Intra-oral periapical radiograph, ortho-pantomograph and CBCT were taken. Radiographs did not show any significant alteration like bony erosion or resorption of bone in the left mandibular posterior region of the jaws (Fig 2, Fig 3 and Fig 4).



Fig 2 Intraoral Periapical radiograph with#33 #34 region



Fig 3 OPG of #33 #34 region showing generalized horizontal bone loss



Fig 4 CBCT of lesion of #33 #34 region

Complete haemogram was done and all the blood parameters were within normal range.

Case Management

Patient was motivated regarding the cessation of the tobacco quid habit. Phase 1 therapy i.e. scaling and root planing was performed. Since the lesion showed as more of a benign entity, wide local surgical excision was performed under local anesthesia (Fig. 5 and Fig.6). The patient was prescribed medications (antibiotic amoxicillin 500 mg three times daily for 5 days, and analgesic Aceclofenac and paracetamol, twice daily for 3 days). He was also instructed to rinse with 10ml of chlorhexidine (0.2%) twice daily for 1 minute for 2 weeks. The excised tissue was fixed in 10% formalin solution and submitted for histopathological examination.



Fig 5 Surgical Excision of the lesion in #33 #34 region



Fig 6 Surgically Excised tissues with measurement

The postoperative healing was uneventful at 2 week (fig 7) time interval at which the histopathological report was also received and to our surprise the lesion turned out to be malignant i.e. sarcomatoid carcinoma.



Fig 7 2 week post operative healing of the site

Histopathological Report (Fig- 8,9,10)

Lesional tissue comprised of dysplastic-stratified squamous epithelium and other part shows ulcerated area showing invasion of malignant epithelial cells in underlying connective tissue in form of islands. These malignant cells show features like altered nuclear-cytoplasmic ratio, cellular nuclear pleomorphism, and vesicular nuclei with prominent nucleoli,

keratin pearl formation, and tutor giant cells formation. However these malignant cells have acquired predominantly spindle shaped morphology showing bizarre mitotic figures. Intervening connective stroma tissue shows collagen fibres interspersed with fibroblasts, endothelial lined blood vessels and moderate chronic inflammatory infiltrate in the form of lymphocytes and plasma cells.

Histopathological Report

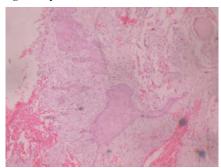
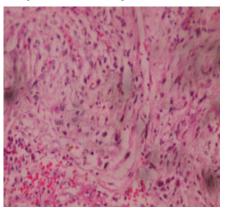


Fig 8 4x Neoplastic cells infiltrating the Connective tissue stroma



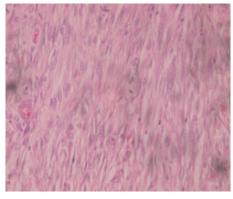


Fig 9 10x Malignant epithelial cells showing spindle cell differentiation

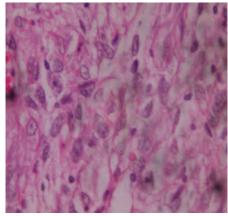


Fig 10 40x showing atypical spindle cells

The patient was informed about prognosis and treatment protocol and referred to oncology department for further treatment.

The patient reported for follow up after 3 months. On Intraoral examination there was no increase in tissue size at 3 month (fig.11). The periodontal tissue was in health. On enquiring about the treatment in oncology department, patient unfolded the fact that he did not go to meet the oncologist as he found the healing to be satisfactory and uneventful.



Fig 11 Postoperative healing after 3 month

The patient at was also persuaded to see the oncologist at that time also but he faltered. The recurrence of the lesion after 6 months was alarming for the patient who made him anxious and thus he reported to the department. On intra-oral examination there was massive gingival overgrowth (Fig. 12).



Fig 12 Post operative after 6 month showing reoccurrence of gingival overgrowth in #33 #34 region

The patient was made aware about his life threatening condition and he was then referred to the oncology department for further treatment (Fig 13).



Fig 13 Left segmental mandibulectomy and stabilization with reconstruction plate

DISCUSSION

In the current case, the growth was reddish pink in colour, non-tender, firm in consistency around the border and soft in the Centre. Peripheral fibroma was excluded from provisional diagnosis since it generally is pink in colour and firm in consistency all over.

Peripheral giant cell granuloma on the other hand is purplishred colour as seen in the current case but it generally presents itself in younger population and anterior region of oral cavity along with pathological migration of adjacent teeth and radiographic sign like erosion of adjacent bone.⁷ Such feature were not noted in our current case excluding it to be the lesion present.

Pyogenic granuloma generally presents as reddish or bluish smooth surfaced mass, characteristically ulcerated, which grows from beneath the gingival margin and so displaces it apically and typically bleeds readily. Similar features were seen in the present case. So the provisional diagnosis of pyogenic granuloma was clinico-radiographically established and was treated by wide local surgical excision. Overall the features were suggestive of benign lesion.

However histopathological diagnosis turned out to be malignant as sarcomatiod carcinoma which is a rare variant of squamous cell carcinoma.

Men are affected more than women, mostly in the middle or later decades of life. Clinical presentation of oral Sarcomatoid carcinoma can vary from an exophytic mass with an ulcerated surface to a frankly infiltrative ulcer, occurring mainly in the alveolar ridge. Among the various risk factors that influence the occurrence of Sarcomatoid carcinoma, predominant factors are alcohol abuse, poor oral health, previous irradiation to the area of the tumor and the most important risk factor being tobacco in various forms such as cigarette, cigar, pipe and smokeless tobacco.

In the present case the lesion is reported in a 51-year-old tobacco chewing male patient. The placement of the tobacco quid in that region of 33 and 34 acted as a constant source of gingival irritation thereby stimulating the activity of the carcinogens which increase the generation of deoxyribonucleic acid adducts. Sarcomatoid carcinoma presented clinically as a gingival overgrowth involving an unusual location, i.e., the mandibular gingiva in left posterior region of 33 and 34. Spindle cell or metaplastic carcinoma, is a rare malignant tumour that consists both malignant epithelial and mesenchymal components, with or without the presence of heterologous elements. ⁹⁻¹¹

The main features of sarcomatoid carcinoma are: (a) the tumor is of epithelial origin; and (b) both the epithelial and mesenchymal cell differentiations exist morphologically. Among a large number of spindle cells, malignant epithelial components are seen in transitional cell carcinomas, squamous cell carcinomas, adenocarcinomas or anaplastic carcinomas. Ultra-structurally, a transitional zone exists between the carcinomatous and sarcomatous components, and sarcomatous component generally accounts for > 50% of the lesion. 12-14

CONCLUSION

The seemingly non threatening gingival enlargement may sometimes be a deceptive entity and challenging to the health sciences fraternity because of its unusual appearance and behavior. Therefore, thorough clinical and importantly histopathogical investigation is a must.

Summary

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	This case report is unique as it presents and discusses the Spindle cell carcinoma of gingiva, which has a very rare occurrence.
Why is this case new information?	It also highlights that, even benign appearing gingival lesions should be looked upon with suspicion. Histopathology should always be used as an definitive adjunct to provide timely treatment to the patient.
What are the primary limitations to success in this case?	The most sensitive and reliable epithelial marker cytokeratin should have been done for demonstrating of the epithelial phenotype.

Conflict of Interest

There is no conflict of interest.

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