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Solitary Neurofibroma of the Gingiva: An Unusual Case Report

Aravinda Basireddy[1*], Santha Kumari Prathypaty[2], Durga Bai Yendluri[3], Santi Priya Potharaju[4]

[1]Assistant Professor, Department of Periodontics, Sri Sai College of Dental Surgery, Vikarabad, Telangana India
[2]Professor and HOD, Department of Periodontics, Government Dental college and Hospital, Hyderabad, Telangana, India
[3]Associate Professor, Department of Periodontics, Government Dental college and Hospital, Hyderabad, Telangana, India
[4]Assistant Professor, Department of Periodontics, Government Dental college and Hospital, Hyderabad, Telangana, India

***Corresponding author:** Aravinda Basireddy
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Abstract | **Case Report**

Neurofibromas are benign soft tissue tumours derived from nerve sheath. This article reports a case of solitary neurofibroma of gingiva in a 35yr old male patient. The lesion was round, well circumscribed, sessile, measuring 1x1 cm in size, colour matching the adjacent mucosa, involving the attached and interdental gingiva in relation to upper left lateral incisor and canine. No radiographic changes were noted. Excisional biopsy was performed. On histopathological examination, bundles of spindle shaped cells were observed along with nerve bundles, blood vessels and fibroblasts. A diagnosis of Neurofibroma was given. The patient was followed up for a year, with no recurrence. Clinicians should include neurofibroma in the differential diagnosis of discrete gingival overgrowths.
Keywords: Solitary neurofibroma, gingival enlargement, oral soft tissue tumour, benign nerve sheath tumour.

INTRODUCTION

Neurofibroma is a benign peripheral nerve sheath tumour, derived from Schwann cells or perineural fibroblasts. Neurofibromas are generally associated with Neurofibromatosis type I (NFI) (also known as Von Recklinghausen disease of the skin), a syndrome with high prevalence of 1 in 2000 to 4000 births[1]. Neurofibromas of the head and neck account for 25% of all neurofibromas. 6.5% of neurofibromas are seen in oral cavity, which include the solitary type and multiple neurofibromas [2]. Both the forms of neurofibroma are identical histopathologically, the only difference being the other syndromic manifestations of NFI. Solitary neurofibromas (SNF) usually present as a painless, slow growing superficial lesion [1]. The present case is a rare finding of a gingival solitary neurofibroma, omitted in differential diagnosis, revealed on histopathologic examination.

CASE REPORT

A 35yr old male patient reported to the Outpatient department of Periodontics, Government Dental College and Hospital, Hyderabad with a chief complaint of a swelling in the upper front tooth region since one month (Figure 1). Patient noticed a small swelling one month before, which had enlarged to attain the present size. There was no history pain or tenderness in the region.

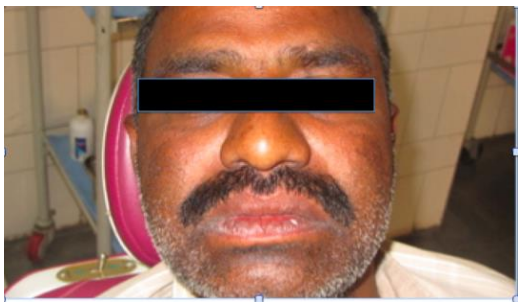

Fig-1: 35yr old male systemically healthy patient showing no other signs of Neurofibromatosis type 1

On intraoral examination, a gingival growth approximately round in shape, measuring 1 x 1 cm, sessile, well circumscribed, was observed in relation to upper left lateral incisor and canine involving the attached gingiva and interdental papilla (Figure 2). The colour of the gingival lesion matched the adjacent

mucosa and did not show any signs of redness or inflammation. On palpation, the lesion was firm in consistency, non-tender and non-reducible. The upper left lateral incisor and canine were periodontally healthy and vitality tests were positive. Extraoral examination of the patient revealed no abnormality. Medical and family history was non-contributory.

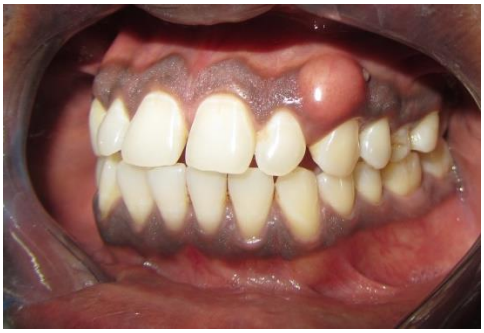

Fig-2: Intraoral photograph showing the soft tissue lesion involving attached gingiva and interdental papilla of 22 and 23

On radiographic examination, the intraoral periapical radiograph showed no changes in the teeth or surrounding tissues. (Figure 3) Based on the clinical and radiographic examination, a differential diagnosis of fibroma, lipoma, gingival cyst of adult, peripheral ossifying fibroma were established.

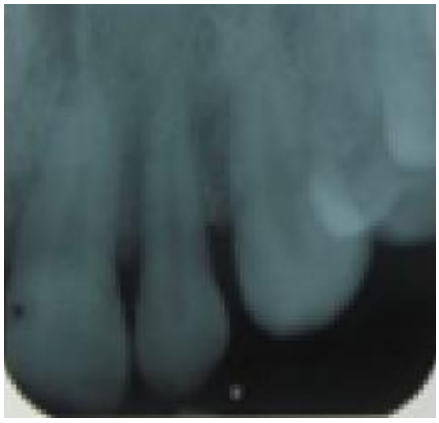

Fig-3: Intraoral Periapical Radiograph showing no signs of abnormality i.r.t 22 and 23

An excisional biopsy of the lesion was performed under local anesthesia. After adequate infiltration with 2% Lignocaine solution containing 1:80,000 adrenaline, the lesion was completely excised along with a margin of normal tissue using no. 15 Bard and Parker blade (Figure 4,5). The tissue was immediately washed with saline and immersed in 10% formalin for histopathological examination. Hemorrhage was controlled in the site and periodontal pack was placed (Figure 6). Patient was prescribed pain killers and recalled after 2 weeks for evaluation of healing.

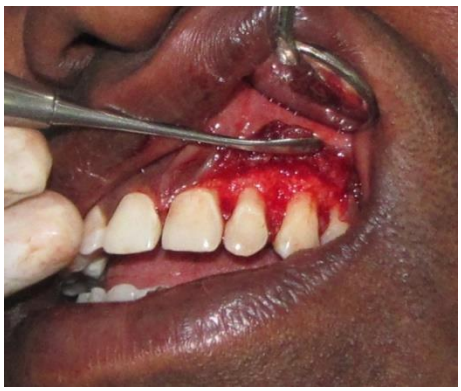

Fig-4: Intraoperative image showing the site after excision of the lesion

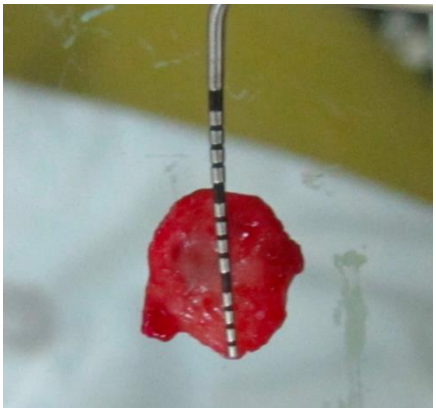

Fig-5: Image showing the excised tissue measuring 1 cm

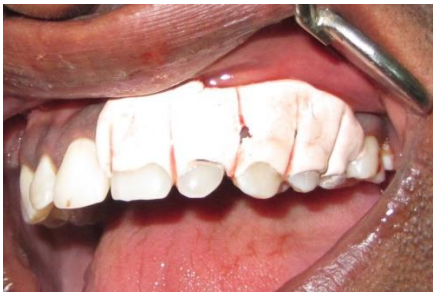

Fig-6: Postoperative image showing periodontal pack placement

On histopathological examination, the Hematoxylin and Eosin stained sections showed multiple, elongated spindle shaped cells with wavy, normochromatic nuclei arranged in fascicles. Presence of numerous nerve bundles was noted. The stroma consisted of blood vessels, fibroblasts and collagen fibrils. (Figure 7) The histopathologic diagnosis was given as Neurofibroma.

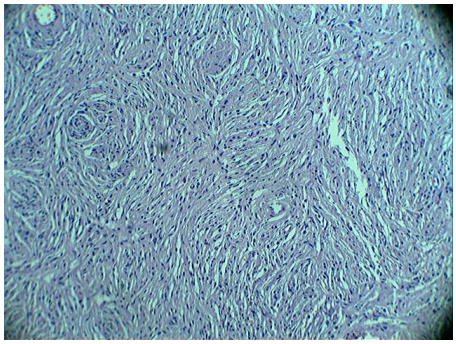

Fig-7: Hematoxylin and Eosin stained section showing Spindle shaped cells, nerve bundles, blood vessels and fibroblasts

The postoperative healing was uneventful (Figure 8). The patient was followed up for one year with recall every 3 months. No recurrence was observed during the period. (Figure 9)

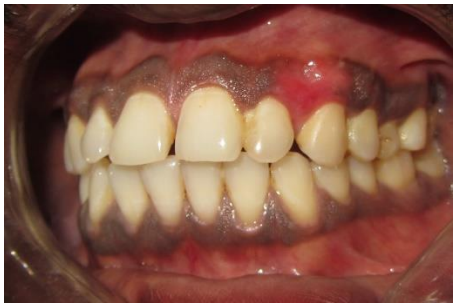

Fig-8: Image showing healing after 2 weeks

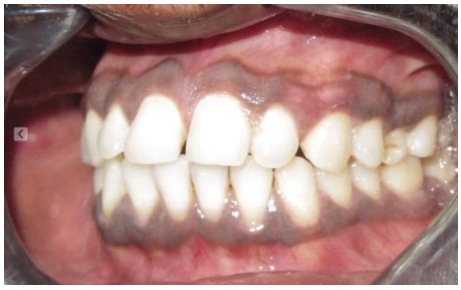

Fig-9: Image showing healing after 6 months

DISCUSSION

A solitary neurofibroma of the gingiva is a rare finding. Oral neurofibromas are common in NFI cases and are accompanied by other systemic manifestations as listed in table 1 [3]. The present case did not show any of the systemic manifestations of NFI. The patient was referred to the Dermatology and Opthalmology departments of Osmania Medical College to rule out these syndromic manifestations.

Table-1: Two or more of the following criteria should be present for diagnosis of Neurofibromatosis type I

Six or more *café-au-lait* macules over 5 mm in prepubertal persons and over 15 mm in greatest diameter in postpubertal persons
Two or more neurofibromas of any type or one plexiform neurofibroma
Freckling in the axillary or inguinal regions
Two or more Lisch nodules or optic glioma
A distinctive osseous lesion such as sphenoid dysplasia or thinning of the long bone cortex with or without pseudoarthrosis
A first-degree relative with NF1.

Only 1.7% of all the oral lesions assessed account for benign nerve sheath tumours, which include Schwannomas, sporadic and multiple neurofibromas [4]. The predominant site of occurrence of SNF in the oral cavity is unknown. SNF has been reported to occur on tongue, palate, buccal mucosa, lip and gingiva. Intraosseous lesions also have been reported in maxilla and mandible [2-10]. They usually present as a painless, slow growing and well-circumscribed lesion.

The World Health Organisation has classified Neurofibromas as – dermal and plexiform [2]. Dermal neurofibromas usually affect a single peripheral nerve while plexiform affects multiple nerve bundles. The present lesion appears to have involved a peripheral nerve of the anterior superior alveolar branch of the maxillary nerve. The NFI lesions also show predilection for cranial nerves V, IX and X [11].

SNFs are considered to be hamartomatous hyperplastic lesions in nature [4]. The etiology of SNF is unknown. The NFI occurs due to a mutation in the 17q11.2 gene which codes for Neurofibromin, a tumour suppressor molecule. Though NFI is inherited through Autosomal Dominant pattern, 50% of the cases occur due to spontaneous mutation. A somatic inactivation of NF1 gene in Schwann cells through chromosomal translocations has been proposed as the probable etiology of SNF by Storlazzi *et al*. [12].

SNF is not usually included in the differential diagnosis of gingival growths. Unlike NFI, they cannot be diagnosed based only on clinical findings. The present case was diagnosed only after histopathological examination. If left untreated, SNF may lead to disfigurement, Paresthesia, difficulty in chewing, pain and tenderness. Clinicians should include SNF in the differential diagnosis of discrete gingival overgrowths.

Recurrence after complete excision in SNF cases has not been reported. However, recurrence with malignant transformation has been reported in 2-6% of multiple neurofibroma cases. The plexiform neurofibromas are more prone for malignant transformation [2]. Thus complete excision with follow up is warranted for these lesions.

CONCLUSION

Clinician should consider SNF in the differential diagnosis of gingival overgrowths. SNFs are rare but do warrant a thorough general systemic examination to rule out syndromes. Once detected, prompt treatment should be given to avoid complications.

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