

Ackerman's tumor of the oral cavity: A study of four cases with its conglomerate appearance

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ABSTRACT

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Oral verrucous carcinoma was first recognized by Ackerman in 1948 as a distinct entity. Although it occurs at various anatomic sites, most intraoral cases involve buccal mucosa, alveolar mucosa and gingiva. The purpose of this article is to describe four cases of verrucous hyperplasia and carcinoma with special emphasis on various clinical presentation and the most predilection sites.

Verrucous hyperplasia and verrucous carcinoma may not be distinguished clinically or may coexist, resulting in diagnostic difficulties. It should be born in mind that leukoplakic lesions may transform into verrucous carcinoma or squamous cell carcinoma, so all such pre-cancerous lesions should be scrutinized conscientiously. Thus, it is amenability of both the oral physicians and histopathologists to be scrupulous about warty and exophytic lesions.

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INTRODUCTION

Oral verrucous carcinoma is a special form of well-differentiated squamous cell carcinoma with specific clinical and histological features. It is a rare tumor first described by Lauren V. Ackerman in 1948.¹ Various names are used in the literature to describe this entity, including verrucous carcinoma (upper aerodigestive tract), Ackerman's tumor, Buschke-Loewenstein tumor, florid oral papillomatosis, on the genitalia (condyloma acuminatum), or on extremities (carcinoma cuniculatum).² The oral cavity is the most common site of occurrence. The most common sites of oral mucosal involvement include the buccal mucosa, followed by the mandibular alveolar crest, gingiva, and tongue. In addition, it is known to occur in the larynx, pyriform sinus, esophagus, nasal cavity and paranasal sinuses, external auditory meatus, lacrimal duct, skin, scrotum, penis, vulva, vagina, uterine cervix, perineum, and the leg.³ The tumor grows slowly, locally invasive and nonmetastasizing behavior. It appears as a painless, thick white plaque resembling exophytic cauliflower like growth with keratin plugging. Shear and Pindborg⁴ first described verrucous hyperplasia, a potentially malignant disorder presenting as a verrucous or exophytic growth characterized by keratosis and/or varying grades of dysplasia and lack of invasive growth. Verrucous hyperplasia is a histopathological entity with clinical features that may be indistinguishable from a verrucous carcinoma. It has been considered an antecedent stage or early form of verrucous carcinoma and is believed to have the same biological potential. The purpose of this article is to describe four cases of verrucous hyperplasia and carcinoma with special emphasis on various clinical

presentations and the most common predilection sites.

CASE SERIES

CASE 1: A 65 year old male patient presented with an exophytic growth on lower alveolar ridge, since 2 years. History revealed that the patient was a chronic smoker with the habit of smoking 10 bidis/day since 38 years. On clinical examination, there was an exophytic finger like growth present on lower alveolar ridge i.r.t edentulous region 31,32,41,42. The growth was measuring around 1.5X1 cm. in its greatest dimension. The growth was light pink in color and had finger like blunt projections. On palpation, the growth was non-friable, non-tender, with well-defined raised margins and no infiltrative induration. Submental lymph nodes were not palpable. In the immediate vicinity, non-scrapable white plaque like lesion was present suggestive of homogenous leukoplakia transforming into verrucous hyperplasia.(Fig. 1)

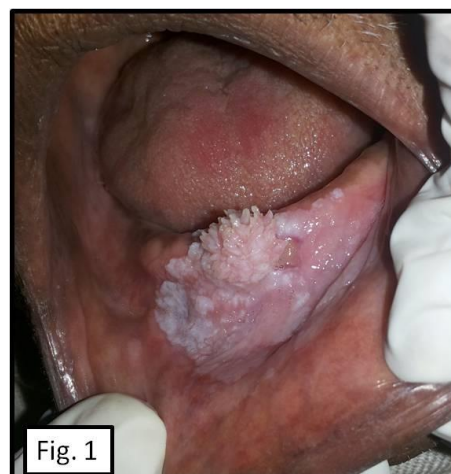


Fig. 1

Fig. 1: Intraoral aspect showing exophytic mass involving lower anterior alveolar ridge.

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CASE 2: A 54 year old male patient reported to the department of Oral Medicine and Radiology with the complaint of growth on left anterior buccal mucosa since 6 months. History revealed that the patient was a chronic bidi smoker with the frequency of 20 bidis/ day since 25 years. Intra-oral examination showed a well-defined growth present on left retrocommisure measuring around 1x1 cm in its greatest dimension and 2-3 mm raised above the mucosal surface. On palpation, the growth had small blunt finger like projections, was non-tender with well-defined margins and no induration could be appreciated. On the posterior extent of the growth, non-scrapable white plaque like lesion was present giving the corrugated appearance of the surrounding mucosa along with diffuse hypermelanin pigmentation. The features were consistent with concomitant occurrence of homogenous leukoplakia and verrucous hyperplasia.(Fig. 2)

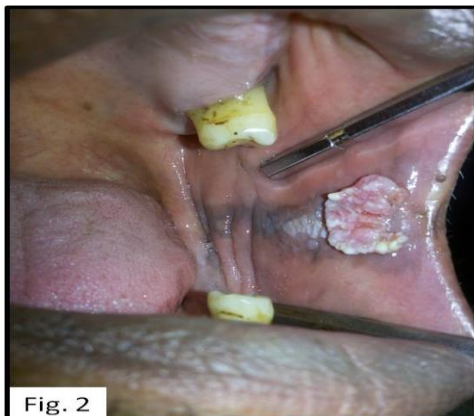


Fig. 2

Fig. 2: Intraoral aspect showing exophytic growth with concomitant occurrence of homogenous leukoplakia in right retrocommisure.

CASE 3: A 58 year old female patient complained of the large growth on right anterior buccal mucosa since 10 months. She was a chronic smoker with the frequency of 40 bidis/day since 32 years. Intraoral examination revealed pebbly growth covering entire right buccal mucosa. It measured around 4x3.5 cms in its longest diameter and was 4-5 mm raised above the mucosal surface. The surface of the growth had pebbly appearance along with small blunt finger like projections extending outward. It was light pink to white in color. The antero-posterior extent of the growth was from right retrocommisure to the posterior buccal mucosa. On palpation, the growth had ill-defined margins, rough texture, slightly tender and induration. The anterior extent of the growth was projecting 1 cm outward from the anterior buccal mucosa and was tan brown to black in color. These features were consistent with the diagnosis of verrucous carcinoma.(Fig. 3a,3b)

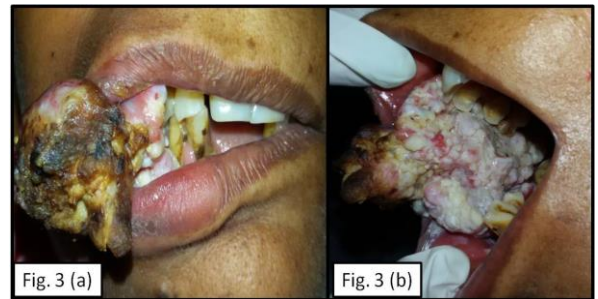


Fig. 3 (a)

Fig. 3 (b)

Fig. 3(a): Extraoral aspect of the anterior extent of exophytic growth projecting outwardly from anterior buccal mucosa with tan brown to black in color

Fig. 3(b): Intraoral examination showing pebbly appearance along with small blunt finger like projections extending outwardly entire right buccal mucosa.

CASE 4: A 50 year old female patient with a low socio economic status residing in a remote rural area and working on a daily wage, reported with chief complaint of growth on palate since 3 months. Patient noticed a tea burn like ulcer in the anterior palate region 18 months back for which she got treated by various doctors without any healing. 12 months before she had undergone biopsy from the palate, reports were not conclusive. Since then it has gradually grown to attain the present dimension. She is a chronic tobacco user 3-4 times/day since last 20 years. She got extraction of upper right lateral incisor due to mobility, 3 months back after which the ulcero-proliferative growth in anterior palate started growing rapidly. On intraoral examination the exophytic growth was covering entire palate till junction of hard and soft palate, laterally extending between the molars and appearing on the buccal gingiva.(Fig. 4a,4b)

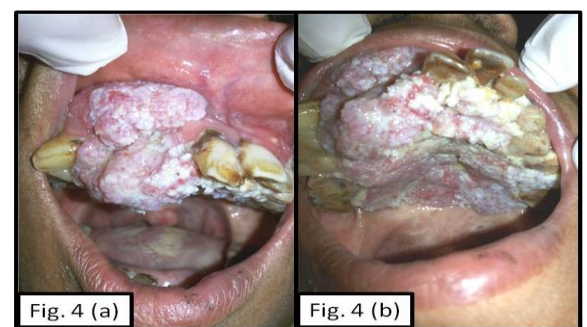


Fig. 4 (a)

Fig. 4 (b)

Fig. 4 (a): Intra oral examination showing the exophytic growth covering anterior alveolus.

Fig. 4 (b): Intra oral examination showing the exophytic growth covering entire palate.

The larger mass was present in anterior alveolus measuring about 3x4cm with grade III mobility of anteriors. There was no sign of sinusitis or pharyngeal bleeding, but she had frequent history of bleeding from anterior mass. Neurosensory deficit for nasopalatine and greater palatine nerve could not be assessed. Radiographs and CT

Table 1: Summary of 4 patients with exophytic lesions

S. no	Sex	Age	Site	Habit	Presentation
1.	Male	65	Lower alveolar ridge	Bidi Smoker	Exophytic growth
2.	Male	54	Left anterior buccal mucosa	Bidi smoker	Exophytic growth
3.	Female	58	Right anterior buccal mucosa	Bidi smoker	Pebbly surface with exophytic growth
4.	Female	50	Anterior marginal gingiva, Palate	Tobacco user	Extensive exophytic growth

examination revealed extensive soft tissue thickening in entire palate with generalized alveolar bone resorption and evidence of extension into left nasal cavity and maxillary sinus. There was left submandibular lymphadenopathy measuring 1cm in diameter. These features were indicative of extensive verrucous hyperplasia with possible malignant transformation.

RESULTS

All the patients were diagnosed with verrucous carcinoma following excisional biopsy. One patient was diagnosed with verrucous hyperplasia and another with verrucous keratosis in their initial histological findings. Mandibular, posterior alveolar crest, and retromolar trigone were the most affected sites (41.6%), followed by the buccal mucosa (16.6%), the palate (16.6%), the floor of the mouth (16.6%), and the lip (8.3%). No patients had evidence of recurrence after treatment.

DISCUSSION

Verrucous carcinoma was considered as slow-growing, exophytic, well-demarcated, hyperkeratotic lesions.^{5,6} They are typically present as extensive, white, warty lesions. In our case series, all the patients had a similar clinical presentation.

Oral verrucous carcinoma traditionally occurs more commonly in older males, above sixth decade. In our series of cases we observed similar demographics wherein the male patients were more preponderant and the mean age at presentation was between the fifth and sixth decade.⁷The reasons accredited could be primal attainment of the disparate habits, frequency and nature of the habits. Although there is a striking male preponderance to OVC, there are studies where equal sex distribution and female predominance has been demonstrated.⁸ This finding was consistent in our series of cases.

According to the study done by Shear and Pindborg⁴ the most common location was gingiva and alveolar ridge. These sites were in correlation with the site to quid placement. However, in the current literature buccal mucosa was the most affected site that was reported by Yeh⁹. In our case series, two patients had lesions on buccal mucosa, one patient had lesion on lower alveolar ridge, while another patient had lesion on palate and marginal gingiva. This was consistent with the previous literature.

Various etiologies of verrucous carcinoma have been postulated. Human papillomavirus (HPV) was considered as one of the causative factors.⁷ Smoking is also associated with development of verrucous carcinoma.¹⁰ In our case series, four patients (Case 1, 2, 3, 4) had the habit of bidi smoking and there was also occurrence of verrucous carcinoma. Tobacco chewing is also a significant factor in the development of this lesion.¹¹ In our study, there was one patient (Case 4) who had this lesion related to habit of tobacco usage. Verrucous hyperplasia and verrucous carcinoma are indistinguishable entity. Both lesions closely resemble each other clinically and pathologically. The most reliable way to separate both the entities, is that in verrucous hyperplasia there is exophytic growth pattern while in verrucous carcinoma there is combined exophytic and endophytic growth pattern recognized on routine haematoxylin-eosin stained tissue sections. Verrucous hyperplasia does not extend into deeper tissues i.e it is superficial to the normal epithelium while verrucous carcinoma extends more deeply.^{12,13} The association of these entities with leukoplakia is significant, as untreated leukoplakia progresses into verrucous hyperplasia/ verrucous carcinoma. Similarly in our series of cases two patients (Case 1 & Case 2) had verrucous hyperplasia accompanied by leukoplakia in the vicinity.

Regional lymph nodes are often tender and enlarged, because of inflammatory component, resembling malignant tumor.¹⁴ On the contrary, only one of the patient (Case 4) had enlarged lymph nodes while in rest of the patients lymph nodes were not affected. Surgery is considered the elementary mode of treatment for verrucous carcinoma. Radiation therapy alone or in conjunction with surgery is seldom being performed. Whenever surgery is contraindicated, other treatment modalities like cytostatic drugs are often preferred.

CONCLUSION

Verrucous hyperplasia, verrucous keratosis, and verrucous carcinoma may not be distinguished clinically or may coexist, resulting in diagnostic difficulties. It should be kept in mind that verrucous hyperplasia may also develop from leukoplakic lesions, and it may transform into verrucous carcinoma or squamous-cell carcinoma, acting as a potential precancerous lesion.

The purpose of this article is to describe four cases of verrucous hyperplasia and carcinoma with special emphasis on various clinical presentation and the most predilection sites. Verrucous hyperplasia and verrucous carcinoma may not be demarcated clinically. Leukoplakic lesions may transform into verrucous carcinoma or squamous cell carcinoma, so all such precancerous lesions should be scrutinized conscientiously. Thus, it is amenability of both the oral physicians and histopathologists to be scrupulous about warty and exophytic lesions.

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