Two Cases of Unusual Clinical Features of Keratoacanthoma

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Abstract
The Iraqi and the Yemeni dental literature lack documentation of keratoacanthoma (KA). It uncommonly appears at the mucocutaneous junction and very rarely reported to arise in the mucous membrane. Nevertheless, it has such a chance; therefore, it is necessary to bring the attention of dental practitioners to this lesion. It is a benign epithelial tumor that frequently affects the face of old people. It usually, appears as an exophytic nodule with a central keratin plug that grows rapidly but with several poses. Clinically and microscopically KA is similar to squamous cell carcinoma (SCC) and can be occasionally misdiagnosed therefore it is important to be biopsied and carefully interpreted. This report presents two clinically unusual cases of KA.

Key words
Keratoacanthoma, epithelial tumors, horn-like projection, pseudo carcinoma.

Introduction:
Keratoacanthoma (KA) is also termed as Molluscum pseudo carcinomatous is basically a rapidly growing epithelial tumor that is believed to be derived from hair follicle cells. It was first known in 1917 as verrucome but described and named in 1950 (1). For some authors, it was considered as a variant of SCC because of the potential metastases and local tissue destruction (2). However, others had considered KA as an aborted malignancy that only rarely progresses into an invasive squamous cell carcinoma (3, 4). Several factors were implicated in the etiology of KA like actinic rays, HPV, trauma, genetic factors, chemical carcinogens and immunocompromised status (5). In Egypt KA of the oral cavity and lips was attributed to occur in smokers of "Goza" and "Shisha" which contains a mixture of crude tobacco fermented with molasses and fruits (6). Typically, KA presents as a 1 to 2 cm dome-shaped or crater-like nodule with central hyperkeratosis. It gains that dimension within weeks, followed by a slow involution period lasting up to 1 year or more, finally it regresses leaving a scar (7). Usually, lesions are solitary and establish as round, firm, skin-colored or reddish papules, with a smooth shiny surface and a central volcano like-ulceration or keratin plug that may project like a horn. The unaffected skin retains its normal appearance. Most KAs affect the elderly and the middle-aged people and occur on sun-exposed areas like the face, neck, and dorsum of the upper extremities which are the common sites. They are more frequent in light-skinned individuals and predominantly affecting males (8, 9). Literally, surgical treatment is required to remove KA, because malignant transformation and metastasis was reported in some cases, and to minimize a non-acceptable scar formation if spontaneous healing goes over (10, 11). Lesions up to approximately 1.0 cm diameter could rather easily be excised by a wedge excision followed by primary closure with the advantage of obtaining the entire lesion, thus permitting thorough histological examination and increasing
the possibility of a more accurate final histological diagnosis \(^{(12)}\). Here we reported two clinically unusual cases of solitary KA that presented with a horn on the central lower lip of a 35-year-old female in the first case, and on cheek skin nearby the angle of the mouth of 20 years duration in a 51-year-old male in the second case.

**Clinical Presentation and Managements**

**The first case**

A 35-year-old healthy female was referred to oral surgery theater in the faculty of dentistry at Ibb University, the Republic of Yemen complaining of a painless horn like structure rising on her lower lip midline. The lesion; as she claimed; had started as a small red papule seven months ago. It has been elongated within four weeks and covered with a dark colored conical scale; which she removed it by her finger leaving the lesion without any bleeding. Then the lesion regained its horn again within three weeks and again she removed it. The patient used to apply cheap lipsticks for more than 10 years. On examination, an exophytic, firm but mobile dome shaped sessile nodule resides at the center of the lower lip vermillion. It was pinkish-red in color, 0.5 cm diameter and rose to 0.4 cm at an acute angle, covered with a central circular yellowish horny material (Figure 1A and B). The tissues around the borders of the lesion appeared with normal color. Depending on clinical examination and history, the provisional diagnosis was KA with a horn; therefore, excisional biopsy was performed. A week later healing continued without any complications (Figure 1C) and four months follow-up showed no recurrence.

**The second case**

A fifty-one years old male attended to maxillofacial surgery department at Sulaimani teaching hospital complaining of an asymptomatic (except a feeling of itching in spring) facial skin lesion near the angle of the mouth. He gave a negative medical history and claimed that the lesion was there for twenty years and without changes. On clinical examination, a papular yellowish brown skin lesion characterized by an irregular surface and well-demarcated borders about 1.5 cm in diameter was affecting left cheek skin near the angle of the mouth (Figure 2A and B). The surrounding skin appeared to be normal; no bleeding and no regional lymphadenopathy were observed. The provisional diagnosis was basal cell carcinoma (BCC) and nevus. The decision was made to excise the lesion (Figure 2C) with a 2-3 mm border. All the information was submitted to the department of oral and maxillofacial pathology for histological examination.

**Histopathological findings:**

Careful examination of the hematoxylin and eosin (H&E) stained sections of both cases under a light microscope showed hyperkeratotic, hyperplastic stratified squamous epithelium arranged into finger-like projections with connective tissue cores that are infiltrated with chronic inflammatory cells. The epithelial layer is sharply demarcated from the lamina properia and the rete ridges appear long with broad bases. The submucosa appears normal with their adipose tissue (Figure 3A). Under high magnification, several abrupt keratin-filled epithelial islands are seen in the downward growing epithelium, however, basal cell hyperchromatism and spinous layer acanthosis were evident (Figure 3B and C). Depending on these histological features the definite diagnosis was keratoacanthoma.

**Discussion**

KA is not an uncommon tumor, but on careful searching, we did not find reported cases in Iraqi or Yemeni dental literature. It can be seen on the facial skin, intraoral, perioral or on the vermilion border of the lips of white skinned elderly people, with the pick incidence of 45 to 69 years old. The types of KA are solitary, multiple or associated with syndromes \(^{(13-15)}\). Solitary KA is considered as a self-limiting benign tumor because, it presents as a small nodule that grows rapidly within 4-5 weeks to 1-2 cm, and then remains static for another 4-8 weeks before undergoing
spontaneous involution with the expulsion of keratin. The complete resolution terminates in the next 6-8 weeks (16). Involvement of the lip had been reported by Hardman (1971) as rare and comprising 12% of all KAs (5), but it is now more widely recognized (9, 11, 17, 18). Chauhan and his colleagues (2011) had mentioned that clinical diagnosis in typical cases can easily be made depending on the history, the rapid growth, the gross characteristics, the clinical course and the keratin plug in the center of the lesion (19). Following these criteria, KA was compatible with our first case, but the presence of a horn in young age was unusual. Only one similar condition was reported by Meghanand, et al (2011) on the lower lip center in of a 40-year-old female which was described as unique and attributed to sunlight exposure or possible viral pathology (11). Nevertheless, the existence of cutaneous horn has been noticed on top of KA and many other clinical conditions like actinic keratosis, wart, molluscum contagiosum, seborrheic keratosis, sebaceous cyst, BCC, SCC (20, 21). Meanwhile, it is worthy to say that the retrieval; after the twice manual removal; of the horn indicated an innocent behavior. In regard to the 2nd case, the surgeon suspicions went to BCC and nevus. Nevertheless, KA was not on their list because, of the perioral location, nodular appearance and 20 years duration. This unusual persistence and non-regression behavior were previously reported by Al-Hoqail and Bhatt (2009) (22). Their case was unusual as the relevant one but located in the forehead region. Although KA is a spontaneously regressing lesion, possible consequences of missed malignancy remain. Therefore, almost all researchers advised for the complete surgical removal as the best treatment for suspected lesions (16, 23, 24). Histopathological interpretations of the biopsies of both cases were fitted with the pattern of KA discussed fussily in the literature (3). This good news was a booster for our patients since they responded very well to the treatment and no further procedures were needed. Moreover, no recurrence was observed when followed over four months. However, we emphasize like other authors that KA looks like SCC or BCC; clinically and microscopically; therefore it was so important to warrant dental professionals to be familiar with this disease (25, 26). The diversity in affecting younger ages, manifesting different morphological features and lasting longer periods are interesting, unusual clinical presentations that assimilate the same histological picture for one disease ‘the Keratoacanthoma’.

Conclusions

- KA is a worthwhile lesion that should be recognized by dental professionals.
- It should be included in the differential diagnosis of exophytic lip lesions.
- Every suspected case should be biopsied.
- As heightened in our report, the lesion can occur in unusual presentations.

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Fig. (1): Clinical photographs of the first case:
A: shows a lateral view of an exophytic dome shaped sessile nodule. B: front view shows the nodule on the center of the lower lip vermilion. C: healing a week after surgical removal.

Fig. 2: Clinical photographs of the second case:
A: shows the skin lesion near the angle of the mouth. B: the papular yellow-brown lesion with an irregular surface and well-demarcated borders. C: the specimen after fixation.

Fig. 3: Photomicrographs of the H&E stained sections of the lesion:
A: shows hyperkeratotic, hyperplastic stratified squamous epithelium arranged into finger-like projections, X40. B: shows the downward growing epithelium and basal cell hyperchromatism (arrows), X100.
C: shows acanthosis and keratin-filled epithelial islands (star), X400.
References:


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