Nasal Vestibular Huge Keratoacanthoma: An Unusual Site

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Abstract: Keratoacanthoma (KA) is a rapidly growing, low-grade neoplasm of pilo-sebaceous and hair follicle units which most often appears on the sun-exposed skin of the middle aged and older persons with multiple or localized occurrence. This tumor is dome-shaped nodule with a central keratinous plug. The etiology of this tumor is not obvious. Exposure to excessive sunlight is the most frequently noted responsible factor in the etiology of KA. About 80% of the tumors occur on the face. The histological features of the KA are often very similar to those of a cutaneous squamous cell carcinoma; however, the tumor structure usually provides a basis for their difference. There are many unusual cases of keratoacanthoma reported regarding site, size or other specifications. In this study, we excised a mass of nasal vestibule, a site far away sun-exposure. To our knowledge, this is the first case of nasal vestibular keratoacanthoma. For a clinician and a pathologist it is important to consider a benign lesion like Keratoacanthoma (KA) in the differential diagnosis of ulcerated nasal lesions and pay attention to differ it from Squamous Cell Carcinoma (SCC) which has a different and aggressive management.

Key words: Keratoacanthoma, nasal vestibule

INTRODUCTION

Keratoacanthoma (KA) is a rapidly growing, low-grade neoplasm of pilo-sebaceous and hair follicle units which most often appears on the sun-exposed skin of the middle aged and older persons with multiple or localized occurrence (Schwartz, 1994; Patterson, 1983). Sir Jonathan Hutchinson (1889) first described it as crateriform ulcer of the face (Godbolt et al., 2001). This tumor is most often a painless solitary pink or flesh-colored, dome-shaped nodule with a central keratinous plug that undergoes spontaneous involution over 6 to 12 months, resulting in a wrinkled scar (Schwartz, 1994; Patterson, 1983). The etiology of this tumor is not obvious. Viruses have been long suggested as a reason, although the deoxyribonucleic acid sequence of the human papilloma virus has been detected only in a few cases. Exposure to excessive sunlight is the most frequently noted responsible factor in the etiology of KA. Other factors include tar exposure, immunosuppressive states, burns, oncogenic chemicals, psoriatic lesions previously treated using psoralen and ultraviolet A therapy and other dermatoses (Schwartz, 1994; Patterson, 1983; Goodwin and Fisher, 1980). Trauma has long been considered as a possible cause of KA (Sean et al., 2003). In recent years, there have been an increasing number of reports of KA arising at sites of trauma (Sheyner and Osipov, 2006).

Eighty-five percent of older people in the tropics and subtropics with this type of tumor develop it on the extremities, whereas 70% of lesions develop on the head and neck of the people in temperate climates (Sullivan and Colditz, 1979). About 80% of the tumors occur on the face (Patterson, 1983). Although, the lesion is usually in the 10-20 mm range, it rarely becomes larger. A keratoacanthoma larger than 20-30 mm in size is called a giant keratoacanthoma (Moriyama et al., 2000; Chow et al., 2005).

The histological features of the KA are often very similar to those of a cutaneous squamous cell carcinoma; however, the tumor structure usually provides a basis for their difference (Patterson, 1983). There are many unusual cases of keratoacanthoma reported regarding site, size or other specifications, but none is reported in site far away sun-exposed areas, such as nasal vestibule. To our knowledge, this is the first case of nasal vestibular keratoacanthoma.

MATERIALS AND METHODS

A 76-year-old man was referred to Amir-AlAm Hospital in June 2007 with a 5 year history of an enlarging mass in his left nostril. Problem had begun as a brown papule in his left nasal vestibule. He did not pay any attention to it until nearly 4 months before its presentation as a mass fell

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out of his left nasal cavity. As it seemed until that time, it was growing in the nasal cavity and so he had no problem. There was no history of trauma or any special topical drug usage. The patient had past history of diabetes mellitus, hypertension, ischemic heart disease and mild Alzheimer.

After local anesthesia, excisional biopsy was performed, the lesion was excised in a full thickness manner with a fusiform incision and narrow margins of normal tissue (about 5 mm) were resected with the tumor. It had no extension to deeper tissue.

RESULTS

At the time of examination a 4 cm firm horn shaped mass with 1.5×1.5 cm base on the vestibular mucosa was present (Fig. 1a-d). As it is shown in the picture, a keratotic well-circumscribed hairy mass fell out of his left nostril. It seems to have an internal base. Medical history was negative for prolonged sun exposure, but he had many actinic lesions on the skin of his hands (Fig. 2) as the brown maculae with irregular margins in different size. We took care not to cut the lesion tangentially and it was

Fig. 1: A huge keratotic mass originating in the left nostril

Fig. 2: Actinic lesions of the hand skin
completely excised. Histological analysis showed elevated symmetric hyperkeratotic acanthotic lesion with epidermal hyperplasia without stromal invasion or atypical cells, demonstrating keratoacanthoma.

No recurrence of the disease was noted during a follow-up period of 6 months.

CONCLUSION

Keratoacanthoma is characterized by rapid growth. In a few weeks, it may become 10-20 mm in size. This is followed by spontaneous healing over a period of 2-6 months (Patterson, 1983). The tumor is rarely larger than 20 mm in size (Moriyama et al., 2000). Interestingly, our patient reported a long period about 5 years for its growth that may be due to invisible site of the tumor before bothering presentation and the size of the lesion in this patient was bizarre.

As we know sun exposure has a key role in pathophysiology of keratoacanthoma (Schwartz, 1994), so nasal vestibule is not a common place and medical history of the patient was negative for prolonged sun exposure. Keratoacanthoma occurs very rarely on the mucosal membrane of the oral cavity. Chen et al. (2003) has reported one of these intraoral keratoacanthomas in a 23 year old Chinese woman. He has mentioned that spontaneous regression of the intraoral one has not been seen because the lesions were completely excised in all of the cases reported. So may be we can categorize the lesion of the nasal vestibule in the mucosal group of the keratoacanthomas.

The most important differential diagnosis of KA is SCC. Many benign lesions may easily be confused with a malignant ulcer (Patterson, 1983) even after histologic examination, due to pseudo-epithelialization (Khorsandi-Ashtiani et al., 2009). Therefore, for a definite pathological diagnosis it is necessary that our biopsy be fully representative of the lesion (Chen et al., 2003). The margins of the KA have the most characteristic feature, which is the elevation of the normal adjacent mucous membrane toward the core of the ulcer with a sudden change in the normal epithelium at the hyperplastic, acanthotic border (Chen et al., 2003). With incisional biopsy, the central part of the lesion is sampled and the diagnosis may be impossible because the margin of the specimen, which is important for differential diagnosis, is not included (Chen et al., 2003). The treatment of a keratoacanthoma is usually surgical resection. A full thickness excision with narrow margins is recommended. Medical treatments include intralesional injection of 5-fluouracil, methotrexate, bleomycin, interferon and steroids. Cryosurgery, curettage and/or electrodessication and radiotherapy are the alternatives (Denkler and Kivett, 2006). In this case, as we were able to fully resect the tumor, no other topical method was used.

In conclusion, this is the first case of nasal vestibular keratoacanthoma and it seems KA should be included in the differential diagnosis of ulcerated nasal lesions with circumscribed, rolled margins where a keratinized bed is confirmed histopathologically (Chen et al., 2003). The treatment of SCC, the most important differential diagnosis of this benign lesion, is more aggressive. So always an initial good biopsy and having a list of possible diagnosis in the mind, guide us to a good practice.

REFERENCES