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Trichoadenoma of the upper lip

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SUMMARY

Background. Trichoadenoma of Nikolowski, who describe the first cases in 1958, is a rare and benign tumor of the hair follicle. It is well-differentiated and slowly-growing. The clinical appearance of Trichoadenoma (TA) can be similar to basal cell carcinoma or epidermal cyst.

Results. We describe a 44-year-old male who was referred for nodular lesion on the upper lip and a TA was diagnosed. Oral examination showed exophytic yellow mass located between mucous membrane of the upper lip and vestibular gingiva, 1.2 per 0.8 cm. Anamnestic data was non-contributory. An excisional biopsy of the lesion was performed. Microscopically, the lesion consisted of multiple keratinous cysts lined with stratified squamous epithelium and intermingled with solid islands of basaloid cells lying within sclerotic stroma. The pathological diagnosis was TA. The surgical wound healed uneventfully.

Conclusion. Because the lesion is unique, it is uncertain how aggressive or indolent the tumor might be. Therefore, the microscopical analysis is mandatory. At the best of our knowledge, this is the second case of trichoadenoma of the lip.

Keywords: lip lesion, oral nodule, lip tumor, oral tumor, oral follicular hamartoma.

INTRODUCTION

Trichoadenoma (TA) is a rare benign tumor of the hair follicle, which was first described in 1958 by Nikolowsky as "organoid follicular hamartoma" (1). It is usually solitary, nodular, slowly growing, and commonly occurring on the face and/or buttocks of adults, with equal male-female ratio (2). The clinical appearance of TA can be misleading, suggesting a diagnosis of basal cell carcinoma, seborrheic keratosis or epidermal cyst (3). Microscopically, the TA presents unique features, as it less mature than trichofolliculoma and is more differentiated than trichoepithelioma (4). It is often apparent a differentiation towards the infundibular portion of the pilosebaceous canal (4).

We report a case of TA of the upper lip in a 44-year-old male and the differential diagnosis is discussed.

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CASE REPORT

In December 2010, a 44-year-old Indian male was referred to our oral pathology department for evaluation of a slow growing nodular lesion on the junction of the membrane mucosae of the upper lip and the vestibular gingiva. The patient reported a six months history of exophytic expanding mass, painless, without trauma episodes, dental infections, or granulomatous diseases. The patient was not smoker and/or drinker. He was no taking any drugs for known diseases. He denied previous allergic events. Its medical and surgical history was not contributory as well as extra-oral palpation of neck lymph nodes. Oral examination had revealed a 1.2×0.8 cm yellow nodule that appeared hard-elastic and fairly fixed at the deep tissues (Figure 1). Radiological examination showed no apical and/or periodontal infection of the upper anterior teeth. The lesion was entirely excised and sent for histopathological analysis (Figures 2, 3).

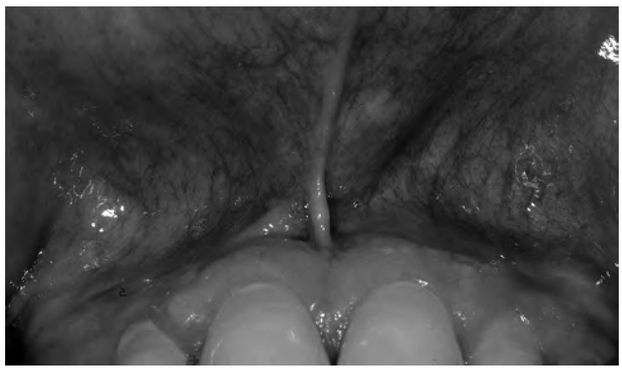

Fig. 1. Oral mass lesion of the left lateral surface of the inner upper lip. The size was 1.2 cm.

Fig. 2. The intraoperative picture of nerve trichoadenoma during the surgical procedure

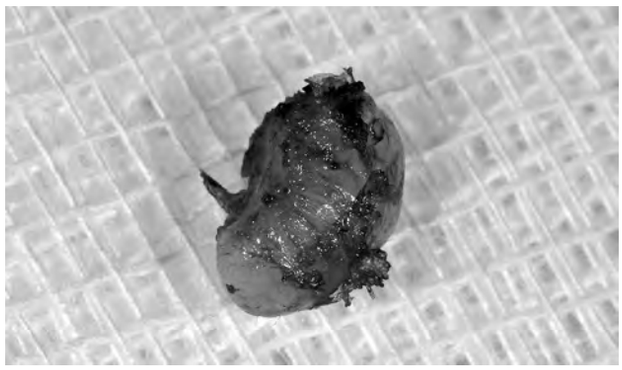

Fig. 3. The gross specimen showing a round mass, 1.2×0.8 cm of size

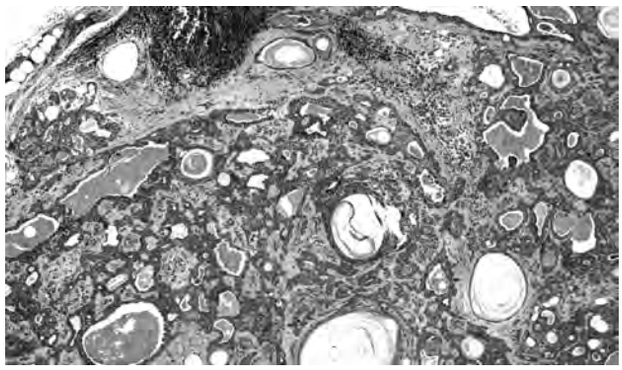

Fig. 4. Photomicrograph showing well defined tumor composed of islands of cells with eosinophilic squamoid to tricholemmal appearance, lacking cytologic atypia, surrounding numerous cystic cavities containing keratinous material (Hematoxylin and Eosin, ×20)

Microscopic examination showed evidence of multiple keratinous cysts lined with stratified squamous epithelium and intermingled with keratinizing islands (Figure 4). The areas were delimited by eosinophilic cells with attempted glandular formation, and surrounding myxoid stroma. These islands were similar to trichoepithelium, without any evidence of hair follicle formation. At fifteen months from the surgical resection there was no clinical evidence of relapses.

DISCUSSION

Trichoadenoma is believed to represent an abortive differentiation of hair follicle (4). Its topographical prevalence is reported to be of 57.4% on the face and of 24.2% on buttocks, though occasionally the neck, upper arm, thigh, shoulder and shaft of penis may also be affected (5). Although TA is characterized by a (particular) prominent cyst formation, at the microscopic examination, TA resembles some features of desmoplastic trichoepithelioma (DTE). Both the lesions retain Merkel cells as an element of differential diagnosis (6), with respect to basal cell carcinoma. Within the spectrum of follicular neoplasms, trichofolliculoma is characterized by numerous "secondary" hair follicles radiating from "primary" central dilated hair follicles and a well-organized fibrovascular stroma (7). Immunohistochemical studies have shown cytokeratin 20-positive in both TA and DTE as a specific marker for epithelial neuroendocrine cells of Merkel (6). Several studies have revealed the presence of the androgen receptor (AR) in basal cell carcinoma (8-10). Shimanovich et al. (11) demonstrated that TA and DTE appear to be universally AR-negative, confirming the entirely benign nature of these follicular tumors (6). In contrast, Ber-EP4 was expressed by most DTEs, whereas its expression was prevalently absent in TAs (6). Misago et al. stated that the infiltrative variant of the infundibulocystic squamous cell carcinoma can be considered as a counterpart of trichoadenoma on the basis of histologic pattern of cellular architecture described by Shimanovich (11). Occasionally, TA was reported as clinically mimicking a sebaceous cell carcinoma of the eyelid (12). Others case reports shown TA associated with five distinct neoplasms arising in a nevus sebaceous, and its simultaneous occurrence with intradermal melanocytic nevus (13).

CONCLUSIONS

Currently, TA still remains as histologically enigmatic tumor, although some immunohisto-

chemical evidence suggest its morphological differentiation as a distinct follicular tumor (6, 14). At the best of our knowledge, this is the second reported case of TA of the upper lip, after the first described in 1993 by Sieron (15). The excisional biopsy is recommended, especially to rule out a basal cell carcinoma.

STATEMENT OF CONFLICTS OF INTEREST

Authors state no conflicts of interest connected with this work.

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