Cellular hemangioma in an adult

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Abstract: This report describes an adult case of cellular hemangioma arising in the lower lip. A 39-year-old healthy woman presented with a polypoid mass of 4 months duration. The tumor imparted little color to the overlying mucosa and was misdiagnosed as a mucous granuloma preoperatively. The lobular proliferation of plump endothelial cells with inconspicuous vascular spaces was a cardinal morphologic feature of the present tumor. (*J. Oral Sci.* 42, 177-180, 2000)

Key words: capillary hemangioma; cellular hemangioma; hemangioendothelioma; juvenile hemangioma; lip.

Introduction

Cellular hemangioma (CH) is an immature form of capillary hemangioma whose increased cellularity and brisk mitotic activity may provide an erroneous impression of angiosarcoma (1-5). Virtually always, CH occurs during the first year of life and the various terms such as juvenile hemangioma, and hypertrophic or infantile hemangioendothelioma have traditionally been used as a designation (1-5). Since the term hemangioendothelioma came to be applied to hemangiomas of intermediate malignancy recently (6,7), the term CH seems most preferable for this type of hemangiomas.

In the head and neck, CH has a predilection for the parotid gland (8,9), but may also occur internally in the oral mucosa in infancy and early childhood (10-12). To our knowledge, the occurrence of CH in an adult is exceptional

(13,14). This rarity prompted us to report the present case.

Case Report

A 39-year-old woman was admitted with a painless, slow-growing mass on the lip. About 4 months before she had bitten her lower lip several times and subsequently a swelling appeared. On admission, a hard, grayish white polypoid nodule of 1.0 × 0.5 cm was seen on the mucosal surface of the lower lip (Fig. 1). Under the diagnosis of sclerosed mucous granuloma, an excision was performed. About three years later no recurrence was observed.

Microscopically, the tumor was less circumscribed and lobulated (Figs 2,3). Within each lobule, the plump endothelial cells with hyperchromatic nuclei were solidly packed and they often surrounded narrow vascular lumina (Fig. 4). Atypia and mitoses were present but slight. In focal areas, entrapment of skeletal muscle fibers was noted, resulting in a pseudoinfiltrative appearance. The stroma showed mild sclerosis. The ulcerated surface area was

Fig. 1 The polypoid tumor on the lower lip.

Fig. 2 Panoramic view of the lesion. HE, original magnification $\times 4$.

Fig. 3 Lobular proliferation of densely packed endothelial cells with diminutive vascular spaces. HE, original magnification $\times 100$.

Fig. 4 A solid hypercellular area, likely to be confused with a non-vascular mesenchymal tumor. HE, original magnification $\times 200$.

Fig. 5 Plump endothelial cells show strong immunoreactivity for CD34. ABC method, original magnification $\times 200$.

Fig. 6 Many endothelial cells stain strongly positive for factor VIII-related antigen. ABC method, original magnification $\times 200$.

accompanied by an inflammatory infiltrate that comprised neutrophils and lymphocytes. Erythrocyte extravasation and hemosiderin pigments were absent.

The plump endothelial cells expressed factor VIII-related antigen (Dakopatts) and CD34 (Q Bend-10, Serotec) (Figs. 5 and 6). Many perithelial cells were highlighted by positive immunoreactions for α -smooth muscle actin (1A4, Sigma).

Discussion

Capillary hemangioma is one of the most common soft tissue tumors in the oral mucosa (15,16). Although primarily infants and children are affected, it is also observed in middle-aged and older individuals. Such lesions are called senile hemangiomas (17). As a rule, the senile form is less cellular than the infantile form and often demonstrates progressive luminal ectasia of the vascular element. On the other hand, the present adult CH showed lobules with diminute vascular spaces and more cellular foci. Similar oral mucosal CH of adults have infrequently been reported in the English literature (13,14).

The present case must be differentiated from several vascular tumors, such as lobular capillary hemangioma (pyogenic granuloma), epithelioid hemangioma (angiolymphoid hyperplasia with eosinophilia) and well-differentiated angiosarcoma. Whereas the former two benign lesions show the striking predilection for the lip, the latter does not (18-20). Usually their histopathologic appearances are sufficiently distinctive to allow differentiation (21).

The predilection of several types of benign vascular lesions for the lip may be related to the fact that relatively large vessels are nearer to the surface at this location than in most others areas, and that the lip is often subject to traumatic insult (18-20, 22).

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