

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.e-jds.com

Correspondence

Intravascular papillary endothelial hyperplasia in the lower labial mucosa mimicking a mucocele

**KEYWORDS**

Intravascular papillary endothelial hyperplasia;
Lower lip;
CD31;
CD34;
Immunohistochemistry

Intravascular papillary endothelial hyperplasia (IPEH) is a benign lesion in the subcutis. It is rarely found in the lower labial mucosa.¹ This report presented a case of IPEH in the lower labial mucosa.

This 45-year-old female patient came to the dental clinic of our hospital for treatment of a mass in the lower labial mucosa for 5 months. Oral examination showed a mass measuring 8 mm in diameter in the lower labial mucosa looking like a mucocele. Thus, the clinical diagnosis was a mucocele. The mass was totally excised under local anesthesia. Histopathological examination of the specimen showed two masses of proliferating endothelial cells with papillary surface each associated with a residual organizing thrombus in two dilated vascular spaces (Fig. 1A, B, C, and D). By immunohistochemical staining, the proliferating endothelial cells were positive for both CD31 (Fig. 1E and F) and CD34 (Fig. 1G and H) and negative for cytokeratin AE1/AE3 (not shown). These histological findings supported the diagnosis of an IPEH. The post-operative course of the patient was uneventful and no recurrence of the lesion was found 2 years after the operation.

IPEH is a benign subcutaneous lesion that is rarely discovered in the lower labial mucosa. Histologically, it can

be classified into 3 subgroups: a pure form that occurs within a dilated vascular space, a mixed form that appears as a focal change in a hemangioma, and a third form that belongs to neither of the first two.¹ In the pure form, the lesions are most frequently located in the subcutis of the fingers, of the head and neck, and in the region between the elbows and hands. For the mixed form, half of the lesions are found in accompany with an intramuscular hemangioma located in no particular predilective sites. Microscopically, the tuft-like or papillary proliferation of endothelial cells is nearly always intimately associated with a thrombus and seems to represent a peculiar variant of an organizing process.¹ Although the proliferating plump endothelial cells look like a hemangiosarcoma, three particular features, including the intraluminal location of the lesion, the absence of tissue necrosis, and the intimate association of the proliferated tuft-like structures with a thrombus, may aid in differential diagnosis from a hemangiosarcoma.¹

The immunohistochemical staining is very useful for identification of tumor cell origin or specific cell type.^{2–5} In the present case, the tumor cells were positive for both CD31 and CD34 (suggesting that the tumor cells are endothelial cells) and negative for cytokeratin AE1/AE3 (suggesting that the tumor cells are not epithelial cells). With the assistance of finding the aforementioned three particular histopathological features in this case, the hemangiosarcoma can be ruled out.¹

The initial clinical diagnosis of the present case was a mucocele. The mucocele is a common lesion of the lower labial mucosa resulting from a trauma-induced rupture of a minor salivary gland duct and spillage of mucin into the surrounding soft tissue. In the present case, we suggest that the similar trauma may cause a thrombus in a medium-sized blood vessel. The thrombus in the dilated vascular lumen undergoes organization and some of endothelial cells proliferate and further to form an IPEH.

<https://doi.org/10.1016/j.jds.2018.09.002>

1991-7902/© 2018 Association for Dental Sciences of the Republic of China. Publishing services by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

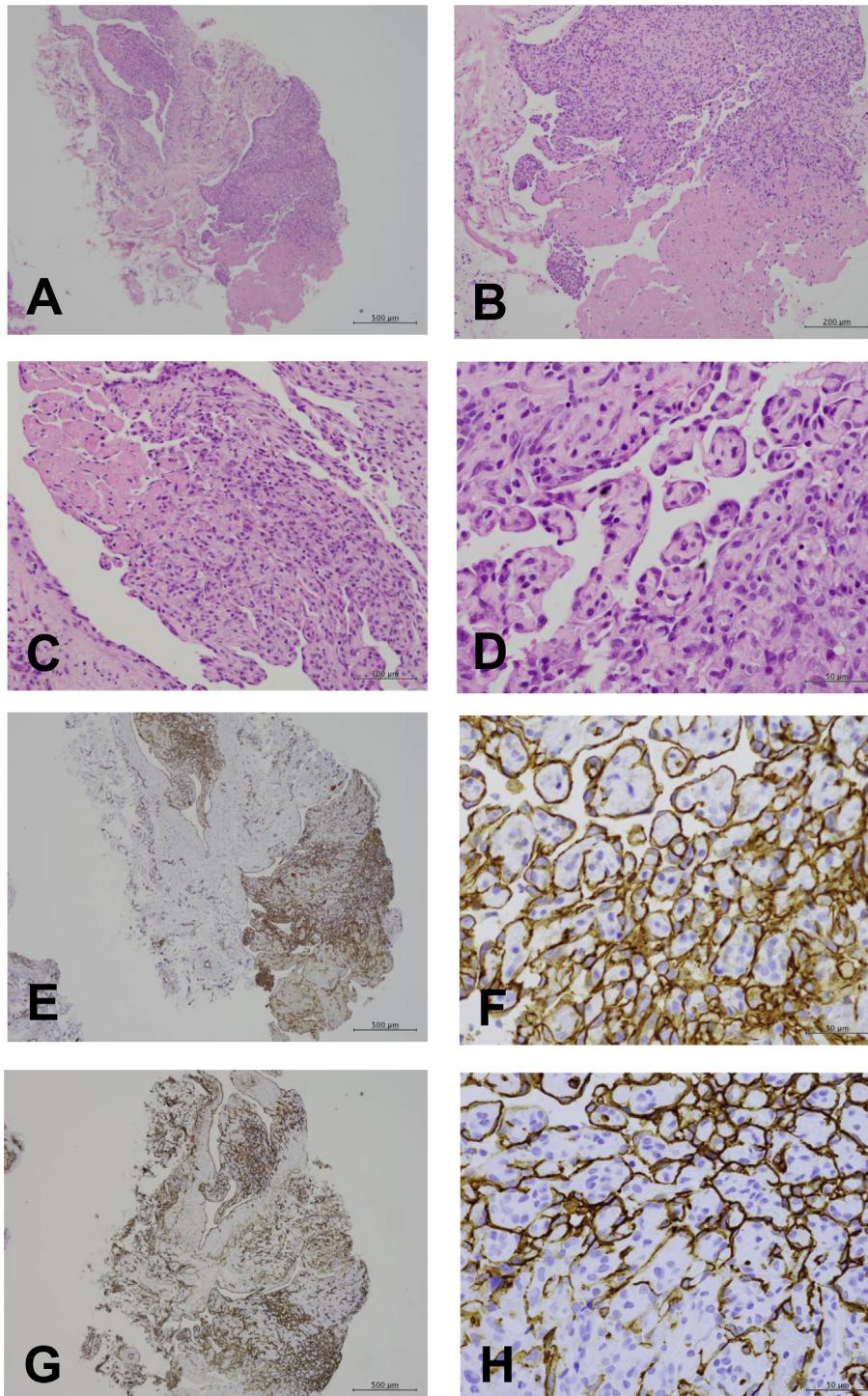


Figure. 1 Histological and immunostaining microphotographs of our case of intravascular papillary endothelial hyperplasia. (A) Low-power view showed two masses of proliferating endothelial cells with papillary surface each associated with a residual organizing thrombus in two dilated vascular spaces (H&E stain, original magnification, 2 \times). (B) Medium-power view revealed a mass of proliferating endothelial cells (upper part) associated with a residual organizing thrombus (lower part) (H&E stain, original magnification, 4 \times). (C) Medium-power view demonstrated a mass of proliferating endothelial cells (right 4/5) associated with a residual organizing thrombus (left upper 1/5) (H&E stain, original magnification, 10 \times). (D) High-power view exhibited proliferating endothelial cells with papillary projections (H&E stain, original magnification, 20 \times). The proliferating endothelial cells were positive for both CD31 (E and F, immunostaining, original magnification, 2 \times and 20 \times , respectively) and CD34 (G and H, immunostaining, original magnification, 2 \times and 20 \times , respectively).

Conflicts of interest

The authors declare no conflicts of interest relevant to this article.

References

1. Hashimoto H, Daimaru Y, Enjoji M. Intravascular papillary endothelial hyperplasia. A clinicopathologic study of 91 cases. *Am J Dermatopathol* 1983;5:539–46.
2. Wu YH, Chang JYF, Wang YP, Chiang CP. Langerhans cells in plexiform ameloblastoma. *J Dent Sci* 2017;12:195–7.
3. Wu YH, Wang YP, Sun A, Chang JYF. Oral plexiform neurofibroma. *J Dent Sci* 2016;11:468–9.
4. Kuo YS, Wu YH, Sun A, Chiang CP. Burkitt's lymphoma of the mandible. *J Dent Sci* 2017;12:421–3.
5. Kuo YS, Wu YH, Sun A, Chiang CP. Eosinophilic granuloma of the mandible mimicking a periapical lesion. *J Dent Sci* 2017;12:424–5.

Ming-Chih Chen

Division of Oral and Maxillofacial Surgery, Department of Stomatology, MacKay Memorial Hospital, Taipei, Taiwan

Chun-Pin Chiang

Department of Dentistry, National Taiwan University Hospital, College of Medicine, National Taiwan University, Taipei, Taiwan

Graduate Institute of Clinical Dentistry, School of Dentistry, National Taiwan University, Taipei, Taiwan

Graduate Institute of Oral Biology, School of Dentistry, National Taiwan University, Taipei, Taiwan

Department of Dentistry, Far Eastern Memorial Hospital, New Taipei City, Taiwan

Julia Yu-Fong Chang
Department of Dentistry, National Taiwan University Hospital, College of Medicine, National Taiwan University, Taipei, Taiwan

Graduate Institute of Clinical Dentistry, School of Dentistry, National Taiwan University, Taipei, Taiwan

Graduate Institute of Oral Biology, School of Dentistry, National Taiwan University, Taipei, Taiwan

Hung-Pin Lin*

Department of Nursing, Mackay Junior College of Medicine, Nursing and Management, Taipei, Taiwan

Department of Stomatology, MacKay Memorial Hospital, Taipei, Taiwan

Department of Dentistry, School of Dentistry, National Taiwan University, Taipei, Taiwan

School of Dentistry, Taipei Medical University, Taipei, Taiwan

School of Dentistry and Graduate Institute of Dental Sciences, China Medical University, Taichung, Taiwan

*Corresponding author. Department of Stomatology, MacKay Memorial Hospital, No. 92 Zhongshan North Road, section 2, Taipei, 10449, Taiwan.
E-mail address: hungpin@gmail.com

Received 12 September 2018
Available online 4 October 2018