Completely Isolated Arteriovenous Hemangioma in the Tongue

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We report a rare case of a completely isolated hemangioma with the histological findings of an arteriovenous hemangioma which occurred in the tongue of a 42-year-old female. This lesion was completely encapsulated by fibrous connective tissue and had focally proliferated with extensive vascular development. The tumor showed a lobular arrangement of vessels of generally uniform wall thickness. Vessel walls were stained by EVG and were immuno-histochemically stained with SMA. Mast cells reactive for toluidine blue staining were frequently identified in the stroma and in the surrounding vascular area.

Key words: Hemangioma, arteriovenous formation, tongue, focal proliferation, encapsulation

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Introduction

Most hemangiomas are known as malformations which are unencapsulated and are reported as angiodysplasia (1) or as angiohyperplasia (2). The principal features of a benign neoplasm are encapsulation by fibrous connective tissue and focal proliferation. On the other hand, arteriovenous hemangiomas in the oral cavity have been reported by Barrett *et al.* (3) and by Koutlas *et al.* (4) who characterized their clinicopathological features. We report a rare case of a completely isolated arteriovenous hemangioma in the tongue.

Case report

A 42-year-old female patient noticed a painless swelling on the left side of the tongue in 2000 and was admitted to the Mitsuwadai General Hospital on September 28th, 2001. T2 weighted magnetic resonance imaging revealed that the swelling was isolated and hyperintense on the frontal plane of the tongue (Fig. 1A). The lesion was surgically removed under the clinical diagnosis of a benign tumor, and macroscopic examination revealed a well-defined, smooth, firm, round nodule measuring 1.5 × 1.3 × 1.3 cm (Fig. 1B). The specimen was sent to the Department of Clinical Pathophysiology of the Tokyo Dental College for pathological diagnosis. The tumor was elastically hard, and the cut surface was white. The pa-

tient had never experienced any history of trauma in that region.

Histopathological findings

After the specimen was fixed with 10% formalin, paraffin sections were prepared for light microscopy and were stained with Hematoxylin and Eosin using routine methods. The consistent histopathological feature of the lesion was an encapsulated mass composed of aggregations of blood vessels, located in the muscular region (Fig. 2A). There were many small vessels with smooth muscle walls and red blood cells. Mast cells stained by toluidine blue were identified in the stroma (Fig. 2B,C). Furthermore, Elastica van Gieson staining showed some expanded vessels with many elastic fibers but small vessels with only a few elastic fibers (Fig. 2D). Immuno-histochemically, smooth muscle actin (SMA, 1:15, Thermo Shandon, UK) was reactive in the smooth muscle fibers of the vascular walls (Fig. 2E). Many endothelial cells were positive for proliferating cell nuclear antigen (PCNA, 1:100, Dako, Denmark), and approximately 67% of the endothelial cells were PCNA positive (Fig. 2F).

Discussion

Hemangiomas are one of the most common types of benign tumor in the oral region, and are generally called

Fig.1: Clinical features. A: T2 weighted magnetic resonance imaging, B: macroscopic appearance during the operation.

Fig. 2: Histological findings. A: Loupe findings of Hematoxylin and Eosin staining; note that the tumor is isolated and is partitioned by fibrous connective tissue. B: Hematoxylin and Eosin staining ($\times 30$); note that vessels have a generally uniform wall thickness. C: Toluidine blue staining ($\times 75$); note that mast cells are stained (arrow head). D: Elastica van Gieson staining ($\times 75$); note that arteriovenous vascular walls reacted. E ($\times 75$): Immunohistochemical staining for SMA, and F: for PCNA.

malformations. We report a patient with a completely isolated hemangioma, which is rare in the tongue. Although a few cases of isolated hemangiomas have been reported (5-8), only Galletti reported an isolated cavernous hemangioma in the tongue (9). The present case was also isolated and partitioned from the surrounding tissue by fibrous connective tissue. Furthermore, this case was histologically determined to be an arteriovenous hemangioma, which is a collection of thick-walled blood vessels with the combined structural characteristics of arteries and of veins. Arteriovenous hemangiomas have been reported as infrequently encountered lesions, but most commonly arise in the head and neck, particularly in the lip, perioral skin, nose, and eyelids (4, 10, 11). The pathogenesis of this lesion is unknown, but Girard *et al.* suggested it is due to the hamartomatous proliferation of the subpapillary vascular plexus (12). The first report of an intraoral arteriovenous hemangioma was by Hassard in 1985 (13), but no histological findings were illustrated. Histological reports of intraoral arteriovenous hemangiomas have subsequently been documented (4), with the consistent histopathologic feature being an unencapsulated mass. This case report shows histopathologic findings with Toluidine blue staining, EVG, and immunohistochemical staining using SMA and PCNA. Toluidine blue staining is valuable to identify mast cells, which are increased in number in this vascular lesion. EVG and SMA are stains to identify vessel walls. Four cases of arteriovenous hemangioma in the tongue have been reported, while Barrett *et al.* reported 15 arteriovenous hemangiomas with lobular patterns in the oral cavity that had arisen at the tip of the tongue; they noted that the pathogenesis of arteriovenous hemangioma may have characteristics of hemangioma and of vascular malformation (3). Furthermore, Barrett *et al.* suggested that the pathogenesis of the arteriovenous hemangiomas of their cases may have entities of hemangioma and of vascular malformation. Oral superficial arteriovenous hemangiomas may be reactive lesions, but some could be regarded as hamartomatous, and the female dominance suggests the possibility of a hormonal influence (3). One interesting finding was the increased number of mast cells identified in the stroma. The report by Koutlas *et al.* about an increased number of mast cells in arteriovenous hemangioma is in agreement with our findings (4). On the other hand, hemangiomas as well as arteriovenous hemangiomas are normally unencapsulated masses of blood vessels (3). Although there has been no report about the proliferative activity of arteriovenous hemangiomas, this case had a high proliferative activity and many PCNA positive cells. Thus, our case is a rare case of a completely isolated hemangioma in the tongue which was histopathologically diagnosed as an arteriovenous hemangioma.

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References

1. Gasparini G, Perugini M, Vetrano S, *et al.* Angiodysplasia with osteohypertrophy affecting the oromaxillofacial area: clinical findings. *J Craniofac Surg* 2001; **12**: 485-9.
2. Mariatos G, Gorgoulis VG, Laskaris G, *et al.* Epithelioid hemangioma (angiolymphoid hyperplasia with eosinophilia) in the oral mucosa. A case report and review of the literature. *Oral Oncol* 1999; **35**: 435-8.
3. Barrett AW and Speight PM. Superficial arteriovenous hemangioma of the oral cavity. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2000; **90**: 731-8.
4. Koutlas IG and Jessurun J. Arteriovenous hemangioma: a clinicopathological and immunohistochemical study. *J Cutan Pathol* 1994; **21**: 343-9.
5. Akel SR, Rassi A, Tawil A, *et al.* Isolated renal haemangioma in children: presentation and management. *BJU Int* 2002; **90**: 758-60.
6. Lee CH. Intestinal obstruction caused by infarcted splenic hemangioma with renal vein thrombosis in a newborn: a case report. *Int Surg* 2002; **87**: 157-9.
7. Hecht DA, Jackson CG and Grundfast KM. Management of middle ear hemangiomas. *Am J Otolaryngol* 2001; **22**: 362-6.
8. Kiratli H, Bilgic S, Caglar M, *et al.* Intramuscular hemangiomas of extraocular muscles. *Ophthalmology* 2003; **110**: 564-8.
9. Galletti C. Cavernous hemangioma confined to the tongue. *Arch Stomatol* 1988; **29**: 1371-81.
10. Connelly MG and Winkelmann RK. Acral arteriovenous tumor. A clinicopathological review. *Am J Surg Pathol* 1985; **9**: 15-21.
11. Neumann RA, Knobler RM, Schuller-Petrovic S, *et al.* Giant arteriovenous hemangioma (cirsoid aneurysm) of the nose. *J Dermatol Surg Oncol* 1989; **15**: 739-42.
12. Girard C, Grahame JH and Johnson WC. Arteriovenous hemangioma (arteriovenous shunt): A clinicopathological and histochemical study. *J Cutan Pathol* 1974; **1**: 73-87.
13. Hassard AD and Byrne BD. Arteriovenous malformations and vascular anatomy of the upper lip and soft palate. *Laryngoscope* 1985; **95**: 829-32.

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