

# *Acta Medica Okayama*

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*Volume 38, Issue 1*

1984

*Article 11*

FEBRUARY 1984

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## Abstract

We report a case of a right buccal cavernous hemangioma with two phleboliths in a 23-year-old man. Consideration of the literature on the cases of hemangioma with phlebolithiasis in the maxillofacial and oral regions in Japan is given in this report.

**KEYWORDS:** hemangioma, phlebolithiasis, maxillofacial and oral regions

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\*PMID: 6702488 [PubMed - indexed for MEDLINE]

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Acta Med. Okayama 38, (1), 79-87 (1984)

## HEMANGIOMA OF THE BUCCAL PAD WITH PHLEBOLITHIASIS : REPORT OF A CASE

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*Received September 10, 1983*

*Abstract.* We report a case of a right buccal cavernous hemangioma with two phleboliths in a 23-year-old man. Consideration of the literature on the cases of hemangioma with phlebolithiasis in the maxillofacial and oral regions in Japan is given in this report.

*Key words :* hemangioma, phlebolithiasis, maxillofacial and oral regions.

Hemangiomas include dilatation, proliferation, degeneration, and malformations of the blood vessels, and generalized malformations and deformities. The lesions occur in various parts of the maxillofacial and oral regions, most frequently in the lips and tongue followed by the gingiva, mandible, palate, bucca, mouth floor and parotid gland. However, there are relatively few reports on hemangioma with phlebolithiasis in these regions. In this report we present one case of a right buccal cavernous hemangioma with two phleboliths, and discuss the literature in Japan on hemangioma with phlebolithiasis in the maxillofacial and oral regions.

### CASE REPORT

A 23-year-old man visited our department on January 30, 1980 complaining of a painless swelling on the right bucca. About 2 years earlier he received medical treatment for nephrolithiasis. He had been well until about 2 weeks prior to his visit, when he noticed a right buccal tumor, which he ignored at first. Because the tumor continued to grow gradually, the patient consulted a dentist one week prior to his visit.

He appeared physically normal and well-nourished. Laboratory examinations were all normal. The right side of the face was slightly swollen, with normal skin color and with no spontaneous pain, tenderness or trismus. An elastic-soft and pea-sized lymph node without tenderness or any adhesion was felt in the bilateral submandibular regions. Outflow of saliva from the right parotid gland and submandibular gland was normal. Neither dryness in the oral cavity nor pain in the right parotid gland while eating was reported. A mild, diffuse swelling was recognized in the anteroinferior part to the orifice of the right parotid gland, and the surface of the swelling was partly bluish purple (Fig. 1). There was a

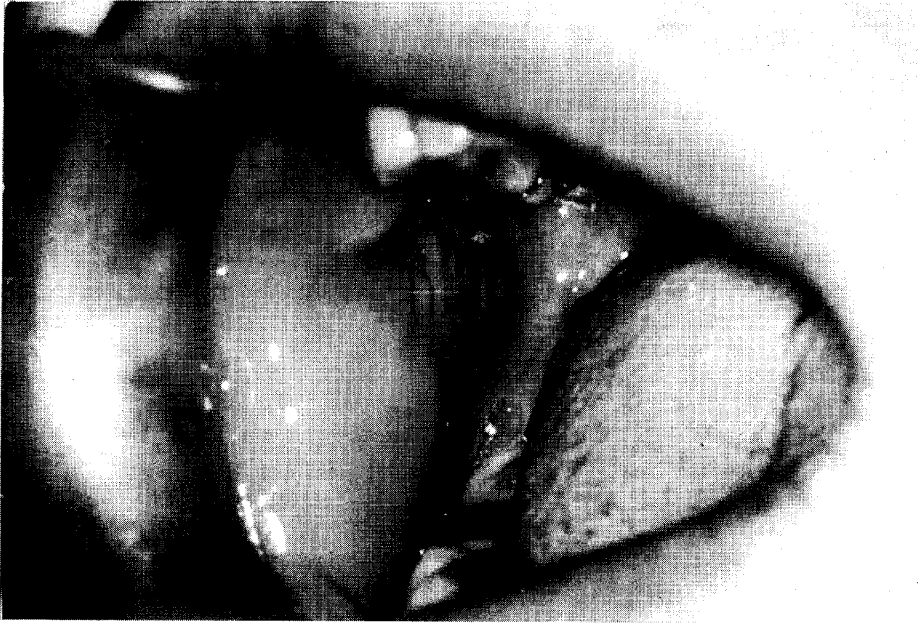


Fig. 1. Patient's oral cavity.

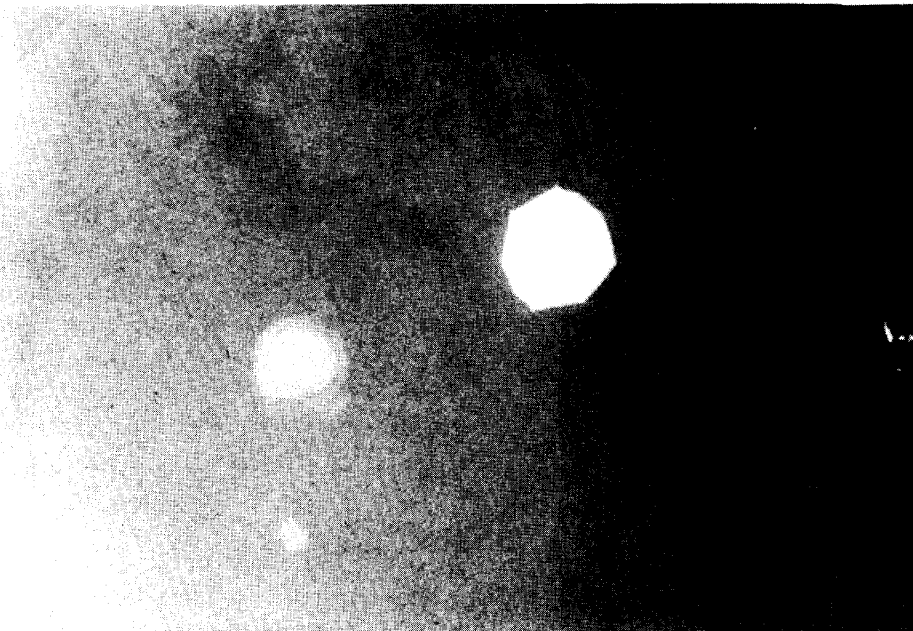


Fig. 2. X-ray film of phlebolithiasis.

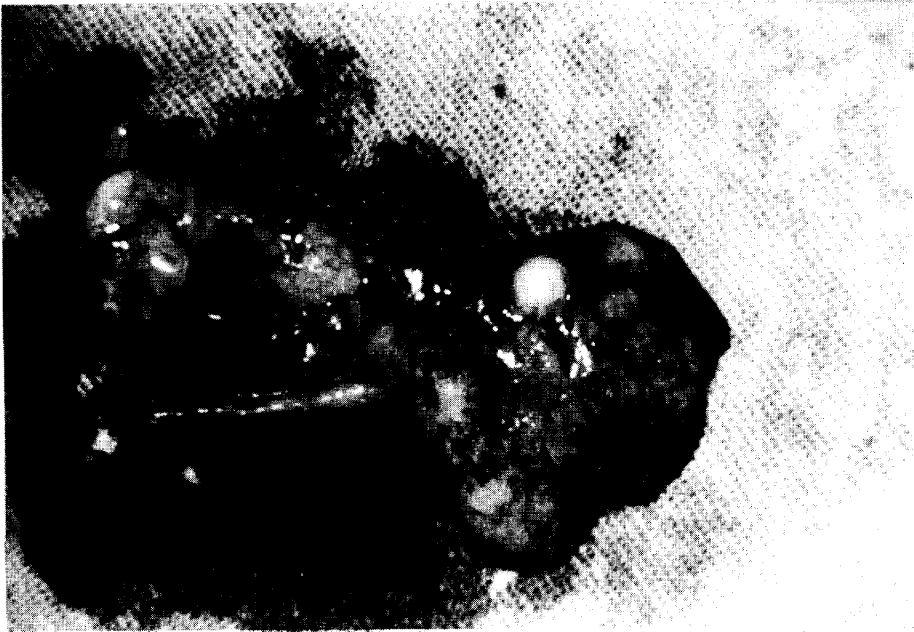


Fig. 3. The enucleated tumor.

well-defined, thumb-sized, elastic-soft tumor felt on the inner and outer sides of the oral cavity. A rice-grain-sized, mobile, hard, solid area was felt within the tumor. X-ray film with a gutta-percha plate on the orifice of the right parotid gland revealed two radiopaque images in the buccal tissue anteroinferior to the orifice of the right parotid gland; the larger one had a concentrically circular laminated structure, though indistinct (Fig. 2). An x-ray film of the right parotid gland showed no abnormalities. The case was diagnosed as hemangioma with concretions.

After local puncture to aspirate about 3 cc of the hemorrhage contents, the swelling disappeared, although several hours later the swelling recurred. After admission, on February 22, the tumor was enucleated. About one week later, the patient was discharged with no occurrence.

The enucleated tumor, measuring  $30 \times 25 \times 15$  mm, had an irregular surface and was elastic-soft and dark red, though grayish-white focally where concretions existed (Fig. 3). Soft x-ray film of the tumor revealed two radiopaque images 2 mm and 1 mm in diameter; the smaller one of which was irregularly shaped, and the larger one of which was oval with partially low radio-opacity. Microscopically, the tumor consisted of variously sized blood vessels with very irregular lumina surrounded by endothelial cells. There was a slight infiltration of round cells into the lumina (Fig. 4). The tumor was thus shown to be a cavernous hemangioma with concretions.



Fig. 4. Microscopic findings of the tumor, HE stain,  $\times 100$ .

#### DISCUSSION

Hemangiomas may be seen anywhere in the body, though usually in the head and face regions. Histologically most hemangiomas are cavernous. Hemangiomas are considered to be an embryogenic malformation, *i.e.*, a kind of hemartoma. Since Kirmisson (1) reported a case of hemangioma with phlebolithiasis in 1905 in Germany, 46 cases of hemangioma with phlebolithiasis in the maxillofacial and oral regions have been reported in Japan including our case (Table 1).

The patient's ages ranged from 5 to 72 ; with 15 patients (32.6 %) in their teens and 14 (30.4 %) in their twenties. Twenty-four of the patients were male, and 22 were female, with no difference in the incidence of hemangioma between sexes. The bucca was affected in 22 cases (47.8 %), the tongue in 7 (15.2 %),

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TABLE 1. HEMANGIOMAS WITH PHLEBOLITHIASIS IN THE MAXILLOFACIAL AND ORAL REGIONS REPORTED IN JAPAN

Case no.	Reporters (Refs.)	Age	Sex	Location	Clinical diagnosis	Number of concretions
1	Asahinata (2)	19	M	Lower eyelid	Hemangioma	Several
2	Toyama (3)	7	M	Bucca	〃	〃
3	Sawano (4)	27	F	〃	—	6
4	Chang (5)	20	M	Tongue	—	4
5	Ozasa <i>et al.</i> (6)	23	F	Bucca	—	Several
6	〃	16	M	Upper lip	—	〃
7	Tada <i>et al.</i> (7)	21	M	Hypoglottis & mouth floor	Sialolithiasis	12
8	Nagao <i>et al.</i> (8)	36		Lower jaw	Hemangioma	10 odd
9	Takahashi <i>et al.</i> (9)	16	F	Tongue	〃	3
10	Matsuda <i>et al.</i> (10)	9	M	Hypoglottis & mouth floor	Concretions in sublingual gland	10 odd
11	Imai (11)	44	M	Submentum	—	Several
12	Nagashima <i>et al.</i> (12)	13	M	Mouth floor & bucca	—	2
13	〃	21	M	Bucca and lips	—	Several
14	Shimizu <i>et al.</i> (13)	38	M	Bucca	Hemangioma	2
15	Nishijima <i>et al.</i> (14)	26	M	〃	〃	6
16	Kimura <i>et al.</i> (15)	53	F	Tongue	Extraneous foreign body in the edge of the tongue	1
17	Tsuge <i>et al.</i> (16)	27	M	Bucca	Hemangioma	Many
18	Masuyama <i>et al.</i> (17)	23	F	Face & bucca	〃	15
19	〃	12	M	face	〃	11
20	〃	36	F	Hypoglottis & mouth floor	〃	4
21	〃	19	M	Bucca	〃	22
22	〃	16	M	Upper lip	〃	1
23	〃	16	F	Bucca	〃	2
24	〃	17	M	〃	〃	2
25	Hamada <i>et al.</i> (18)	20	M	〃	〃	9
26	Kitamura <i>et al.</i> (19)	15	F	〃	〃	Several
27	Hisano <i>et al.</i> (20)	19	F	Hypoglottis & mouth floor	〃	8
28	Higashi <i>et al.</i> (21)	52	F	〃	〃	1

Table 1. continued.

29	Terai <i>et al.</i> (22)	14	M	Bucca	〃	4
30	Masuda <i>et al.</i> (23)	39	F	Submentum	〃	13
31	Honda <i>et al.</i> (24)	19	F	Bucca	〃	6
32	Ueno <i>et al.</i> (25)	12	F	〃	〃	40 odd
33	Inawashiro <i>et al.</i> (26)	29	F	Tongue	〃	3-4
34	Okui <i>et al.</i> (27)	26	F	Bucca	〃	5
35	Mori <i>et al.</i> (28)	9	M	Bucca	—	Many
36	Hashimoto <i>et al.</i> (29)	49	M	Tongue	Benign tumor of the tongue	1
37	Morinaga <i>et al.</i> (30)	21	F	Transitional part between gingiva & bucca	Hemangioma	1
38	〃	33	F	Tongue	〃	1
39	Shigematsu <i>et al.</i> (31)	72	F	〃	〃	2
40	Kanbe <i>et al.</i> (32)	56	F	Hypoglottis & mouth floor	〃	3
41	〃	22	F	Bucca	〃	3
42	〃	36	M	〃	〃	1
43	〃	5	F	〃	〃	1
44	〃	10	F	〃	〃	2
45	Hibino <i>et al.</i> (33)	9	F	〃	〃	11
46	Present case	23	M	〃	〃	2

the hypoglottis and mouth floor in 6 (13.0 %), the lips in 3 (6.5 %), the submental part in 2 (4.3 %), and the lower lid, submandibular part, mouth floor and bucca, bucca and lips, face and bucca and the part between the gingiva and bucca in one case each (2.2 %). Hemangioma of the lips or tongue occurs relatively rarely, as reported previously by Nishijima *et al.* (14) and Okui *et al.* (27). There was a large difference in the number of concretions : from 1 to about 40.

All cases were clinically diagnosed as hemangioma except one case each of sialolithiasis, concretion in the sublingual gland, extraneous foreign body in the edge of the tongue and benign tumor of the tongue. Differential diagnosis, which requires much attention in cases of deeply situated tumors with concretion, was thus relatively easy. Roentgenologically, sialolithiasis can be differentiated by its radiopaque image. However, as to the characteristics of phlebolithiasis, Lerche (34) reports scattered, round radiopaque patterns with lamination. Fabian (35)



reports mostly round, welldefined, irregularly scattered phleboliths of various sizes, mostly soybean to walnut-sized. Kirmisson (1) reports that a concentric circular stratum with shadow is often seen in the center of a phlebolith. These characteristics were also recognized in our case. Most of the cases of phleboliths we studied had a round or elliptical shape and lamination. Masuda *et al.* (23) report that concretion is found in the blood vessels by angyography, and can be differentiated from at least sialotithiasis, lymphangioma and concretion of the sublingual gland.

Ribbert's opinion (36) that phleboliths form by calcification on a thrombus which has been organized with collagen fibers is generally supported. In scanning electron micrographs, Kanbe *et al.* (32) showed that phleboliths had an acicular, cylindrical or cubic crystal structure and adhered to a substance with a blood cellular structure, and that the image suggested calcification on the thrombi which started the adhesion of crystals to the blood cells. Okui *et al.* (37) reported that in the center of the concretion there was a nucleoid substance with a convolute putamen, but the structure was irregular, there were no crystals, and ringed epibolies were built up around the nucleus. Fibrous epibolies forming matrices of organic components and those with inorganic components filling the slit were laminated alternately. Okui *et al.* (37) also reported that the principal ingredients of the nucleus-like substance was identified by x-ray analysis to be close to hydroxyapatite, that the principal elements were shown to be calcium and phosphorus by x-ray microanalysis and that  $PO_4$ ,  $CO_3$ , lipids and proteins were recognized in infrared absorption spectrographs.

Treatments of hemangioma include the package method (10, 13), radiotherapy (12, 38), injection of a tissue-sclerosing agent (18, 32) and surgical treatment (6, 14-29). Combined treatments have been reported, *e.g.*, the injection of a tissue-sclerosing agent and surgical treatment (17, 32), radiotherapy and surgical treatment (25), and the package method and surgical treatment (33). Surgery was employed in our case because the hemangioma was relatively well circumscribed.

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