〈Case Report〉

A case of angioleiomyoma in the hard palate with accompanying pain

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ABSTRACT Angioleiomyoma is a benign tumor of smooth muscle origin that commonly arises in the skin and subcutaneous tissue of the lower extremities and is rarely found in the oral cavity. We treated a case of angioleiomyoma in the hard palate of a 59-year-old male. He was referred to our hospital with the chief complaint of spontaneous pain in the palate. The lesion measured 11×8×4mm and was surgically removed and diagnosed as angioleiomyoma. Previous cases of oral angioleiomyoma in the hard palate reported in Japanese literature are also reviewed here. doi：10.11482/KMJ-E202046173 *(Accepted on November 6, 2020)*

Key words：Angioleiomyoma, Hard palate, Oral region

INTRODUCTION

Angioleiomyoma is a benign tumor that originates from the proliferation of the smooth muscle cells and commonly arises in the lower extremities of middle-aged to elderly women, but the occurrence in the oral cavity is rare[1)]. Stout first reported on angioleiomyoma in the oral cavity in 1938[2)]. Oral presentations tend to be on the lips[3)] or the tongue, with the hard palate being somewhat rare.

We report here on a case of angioleiomyoma that occurred in the back of the hard palate with some considerations.

CASE REPORT

Patient: a 59 year-old male

Initial visit: October 2019

Chief complaint: a tumor mass in the palate

Current medical history: The patient acknowledged that the tumor mass in his palate originated approximately 10 years prior to his initial visit, but he left it untreated because the tumor didn't cause any pain. He did not notice that the tumor mass had been increasing in size. Shortly before his initial visit, the tumor started to cause pain, so he consulted his dentist. He was referred to us at Kawasaki Medical School (Oral Department) to examine the lesion, which was suspected of being a mucinous cyst.

Medical history: hypertension treated with three medications, glaucoma

Medical history in the family: nothing outstanding

CURRENT CONDITION

Systemic findings: body mass was in the average range; nutritional status was normal; no other abnormal findings

Findings outside of the oral cavity: symmetric

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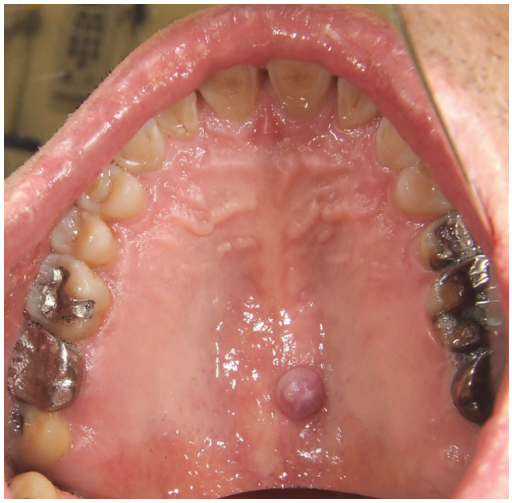

Fig. 1. Findings inside of the oral cavity

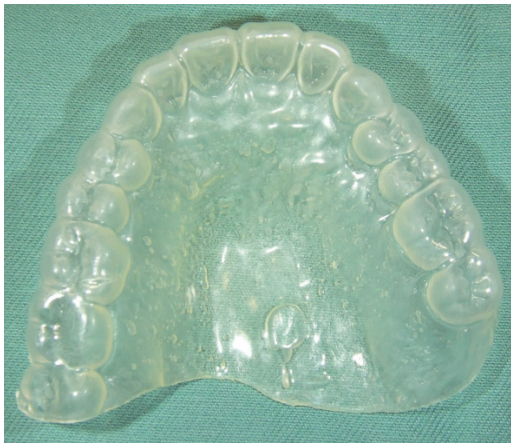

Fig. 2. A rubber surgical splint

facial features; no face and neck lymph node enlargement or tenderness

Findings inside of the oral cavity: A mass (11 × 8 × 4 mm) was found in the back of the hard palate, slightly left of the median. The pedunculated mass was elastic soft and hemispherical (fig. 1). The superficial membrane was dark purple and a partially denuded wound was found. There was no discoloring caused by compression.

X-ray findings: no bone resorption was found on the panoramic x-ray; no other abnormal findings

Clinical diagnosis: benign palatal tumor mass

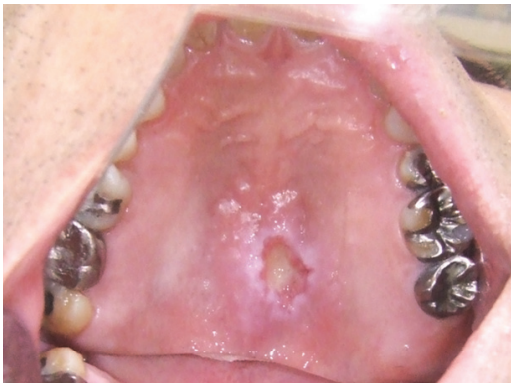

Fig. 3. 14 days after operation

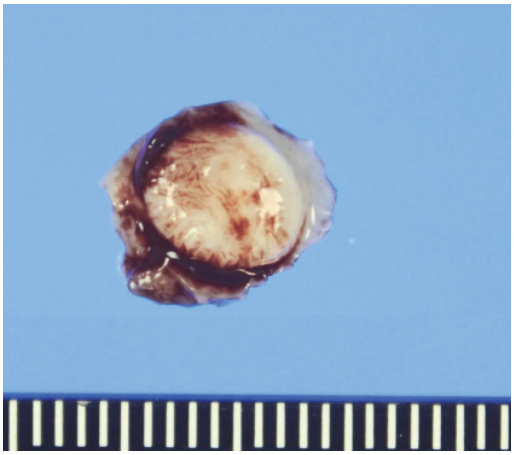

Fig. 4. extracted specimen

Treatment and follow up: Upon the patient's first visit, a surgical protective dental splint (fig. 2) was created. Under local anesthesia, the mass and 2 mm beyond the edges of the mass were resected, including the periosteum. Bleeding was seen under the tumor and the membrane was sutured to stanch the bleeding. The splint was reinserted at the end of the procedure. At a 2-week post-surgery follow up, the lesion was healing without complications (fig. 3). Currently, there is no recurrence or any infection.

Histopathological findings: The mass was an ovoid micronodule (6 × 4 mm) with a clear boundary under the epithelium. Acidophilic spindle cells proliferated as they tangled. Inside of them existed numerous small vessels and the spindle cells

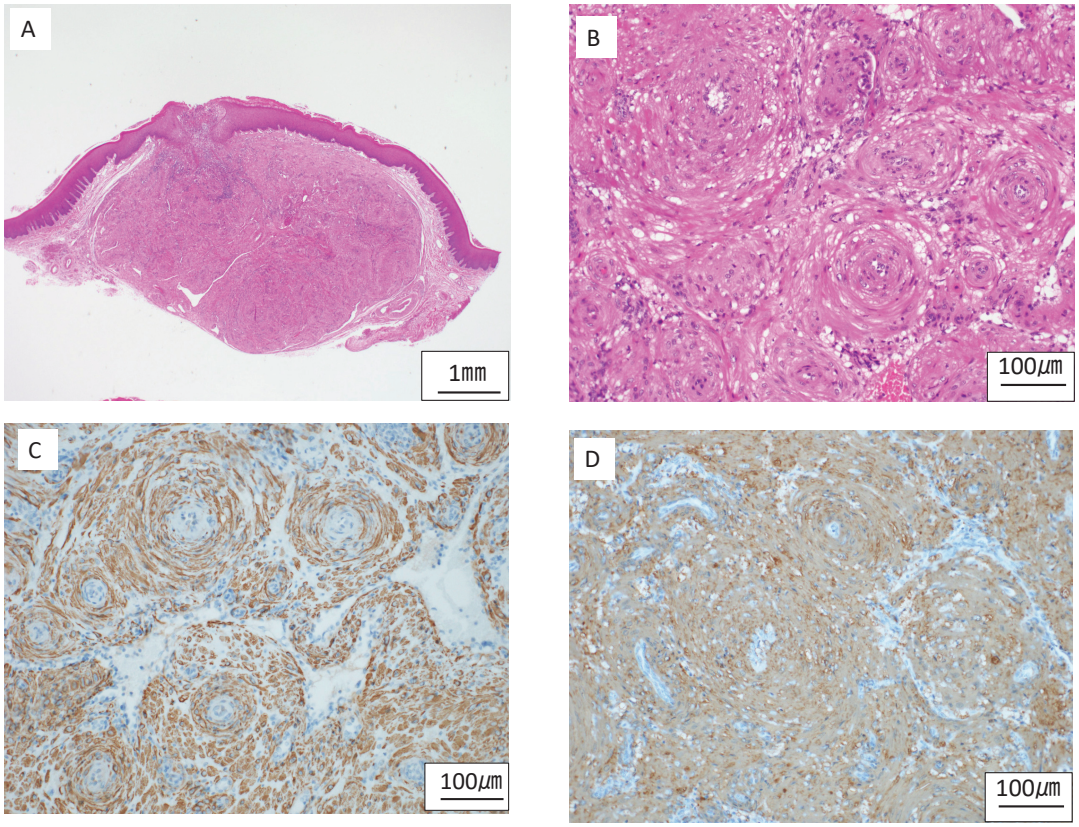

Fig. 5. Histopathological findings (A: H-E stain × 20, B: H-E stain × 200, desmin stain × 200, D: α-SMA stain × 200)

were surrounding those small vessels.

Immunostaining showed the spindle cells positive for α-SMA and also positive for desmin except the surrounding of the vessels. No malignant findings were detected, and the surgical margin was negative (figs. 4, 5A, B, C, D). The case was histopathologically diagnosed as angioleiomyoma.

DISCUSSION

Angioleiomyoma is a benign tumor that typically arises in the dermis or subcutis and is composed of well-differentiated perivascular smooth muscle cells arranged around numerous vascular channels[4]. Angioleiomyoma is classified the tumor into three histologic types: 1) capillary or solid, 2) cavernous, and 3) venous[5, 6]. A morphological continuum exists between angioleiomyoma and myopericytoma. Matsuyama[7] suggested that they are closely related tumor. Angioleiomyoma is said to most commonly occur subcutaneously in the lower limbs of middle-aged women[5], and it is rare for angioleiomyoma to occur in the oral region where there are not very many smooth muscle cells. In Japan, Shimada[8] reported the following oral region occurrences: 54.5% on the lips, 24.2% in the palate, 12.1% on the gingiva, and 4.6% on the tongue or mucous membrane of the cheeks. Occurrence in the hard palate is relatively rare.

In the 52 years from 1969 to 2020, there have only been 27 reported cases (including the one in this report) of angioleiomyoma in the hard palate in Japan[5, 9–29] (Table 1). There were decidedly more male patients, 24 male cases and just 3 female cases (11%). Reports[5] on the occurrence

Table 1. Angioleiomyoma that occurred of the hard palate in Japan

	year	Author	sex	age	reported durations	size (mm)	color	hardness	histological type	pain
1	1969	Terayama [9]	male	39	6 years	10	−	elastic hard	unknown	−
2	1973	Morimoto [5]	male	35	−	9	−	elastic hard	venous	unknown
3	1978	Kinoshita[10]	male	52	4 years	7×7×5	light yellow	elastic hard	unknown	−
4	1978	Kinoshita[10]	male	6	1 week	15×15×7	light pink	elastic hard	capillaries	−
5	1978	Nakamura[11]	male	54	12 years	10	dark purple	elastic soft	venous	−
6	1978	Nakamura[11]	male	39	3 years	15	−	elastic soft	venous	+
7	1983	Kajiyama[12]	male	66	10 years	11×9×10	light pink	elastic soft	venous	−
8	1984	Ishibashi[13]	female	42	−	9×9×5	dark purple	compressibility	venous	−
9	1991	Oda[14]	male	72	28 years	15×12×8	dark red purple	elastic soft	venous	−
10	1991	Edamatsu[15]	male	48	28 years	30×30×40	dark purple	elastic soft	venous	−
11	1993	Iwama[16]	male	69	−	12×8	dark purple	elastic soft	venous	−
12	1996	Kobayashi[17]	male	24	3 months	11×10	dark purple	elastic soft	venous	−
13	1996	Kobayashi[17]	male	64	−	10×10	red	elastic soft	venous	−
14	1997	Kamibayashi[18]	female	16	2 months	38×30×23	light pink	elastic hard	unknown	+
15	1998	Takagi[19]	male	61	8 months	10	−	−	venous	−
16	2000	Nishihara[20]	male	39	3 years	8×8	red	elastic soft	venous	−
17	2001	Kurokawa[21]	male	45	−	33×22×15	dark purple	elastic soft	venous	−
18	2001	Kurokawa[21]	male	32	−	10×10×7	dark purple	hard	venous	−
19	2001	Suzuki[22]	male	36	a few weeks	9×15×17	red	elastic soft	venous	−
20	2001	Ozawa[23]	male	40	1 week	13×10	red − white	elastic soft	unknown	−
21	2003	Tajima[24]	male	51	10 years	10×10	red	elastic hard	mixed	−
22	2003	Okamoto[25]	male	65	10 years	15×13×8	red	elastic soft	capillaries	−
23	2013	Tatehara[26]	male	55	7 years	10×7×6	red purple	elastic soft	mixed	−
24	2014	Tsuji[27]	male	79	5 years	15×15	dark purple	firm	unknown	−
25	2019	Higashikawa[28]	male	67	−	11×7	dark purple	elastic soft	mixed	−
26	2020	Nagai[29]	female	79	−	10×10	dark red	elastic soft	venous	−
27	2020	Yamamoto	male	59	10 years	11×8×4	dark purple	elastic soft	venous	+

of angioleiomyoma in the limbs show a 2:3 male: female ratio, which is markedly different than the reports on hard palate cases. The onset age of the 27cases was as young as 6 years old and as old as 79 years old, and with an average age of 49.4 years. Either way, the fact stated above basically matched with the case we experienced.

The reported durations of angioleiomyoma in the hard palate had an incredible range from as short as a week all the way to 28 years. The reason for the long duration is that many patients never experienced pain or discomfort and the angioleiomyoma went undetected for years in some cases. This included 7 cases discovered by dentists.

The average reported tumor size was 14.2 mm, with the largest being 40 mm in diameter. Most of them were spherical and some were ulcerous[11, 18], so differential diagnosis was required to determine malignancy. The tumor masses were often dark purple, and if not, then tended to be reddish or purplish. Also, almost all of the masses were elastic, with 17 soft-elastic cases and 6 hard-elastic.

Hachisuga [6] said the total of 562 cases of angioleiomyoma could be separated into 374 cases (66%) of the solid type, 61(11%) of the cavernous type, and 127(23%) of the venous type. The majority of cases occurring in the oral cavity are the venous type, as was the case we are reporting on here. Morimoto [5] also reported that 67% of the cases had pain in one or more limb. And Matsuyama [7] reported that 83% (54 cases) of the cases had pain in all anatomic sites. Among the 27 hard palate tumor masses, only 3 (11%) reported to have pain. Among those three cases with pain, 2 had an accompanying ulcer[11, 18] and the third was our case, so accompanying pain is very rare. Even the tumor

mass in our case had a denuded ulcerous region on the surface of the tumor mass that appeared as if it had been scrapped.

Morimoto[5] also reported on the relationship between pain and the specific tumor mass. In cases where the tumor mass occurred in a limb, the mass tended to be of a smaller diameter, and the capillary histological type tended to have pain. However, this tendency was not applicable to the cases that occurred in the oral region. The pain mechanism for limb tumors has a few different theories. One theory is that limb tumor masses have a considerable number of nerve fibers and pressing the nerve fibers down eventually causes pain[30]. Another theory is that the sympathetic nerve tightens the smooth muscles in the vessels, causing pain when the cavity in the blood vessels does not get enough blood[5]. The other theory is that nerve fibers around the tumor act as the stimulus receptor[31]. Hasegawa[32] said that the pain could be mediated by the nerve fibers especially located within the tumor parenchyma by the immunohistochemical study. However, the numbers of oral region tumors are still too limited to clarify the pain mechanism. We suggest that 3 cases of tumor mass with pain had a denuded ulcerous region on the surface that appeared as if it had been scrapped, so the nerve fibers are pressed eventually and cause pain like limb tumor.

In Japan, no recurrence has been reported in any of the cases. However, outside of Japan there was a report of recurrence 9 months after surgery[33] and another one 2 weeks[34] after surgery, so careful follow up observation is necessary.

Our case had an important consideration. The surgical lesion was close to the palatal arterial branch and suturing the palatal mucous membranes after removing the periosteum proved difficult. Therefore, we created a rubber surgical splint that could protect the wound. This kind of splint has often been used at our facility to stanch bleeding and avoid any external pressure onto the wound. The splint was very useful in this specific case as well, blocking any foreign substance from getting into the wound and thus helped the wound membrane recover more quickly.

CONCLUSION

We experienced a case of angioleiomyoma in the hard palate that accompanied pain, and we reported the case and its summary.

CONFLICT OF INTEREST

The authors declare no conflicts of interest.

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