Clinical Characteristics of Angioleiomyoma of the Hard Palate: Report of a Case and an Analysis of the Reported Cases

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Angioleiomyoma is a rare, benign tumor often found in the uterine myometrium, gastrointestinal tract, and skin and seldom observed in the oral and maxillofacial region. The most common site of occurrence in the oral cavity is the lip, followed by the palate, buccal mucosa, and tongue. The number of reports associated with angioleiomyoma arising from the hard palate is very small. The tumor is histologically characterized by the proliferation of mature smooth muscle cells and numerous blood vessels. When the diagnosis is difficult, specific immunohistochemistry is used. This report describes a case of angioleiomyoma in which there was a chronically increasing lesion for 5 years on the left hard palate and the means for making a definitive diagnosis was based on previous reports on angioleiomyoma of the palate.

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Angioleiomyoma is a rare, benign tumor often found in the uterine myometrium, gastrointestinal tract, and skin and seldom observed in the oral and maxillofacial region.[1,2] The most common site of occurrence in the oral cavity is the lip, followed by the palate, buccal mucosa, and tongue.[3] To the best of the authors' knowledge, the number of reports associated with angioleiomyoma arising from the hard palate is very small. To make the diagnosis, histologic examination is typically performed. The tumor is histologically characterized by the proliferation of mature smooth muscle cells and numerous blood vessels. When the diagnosis is difficult, specific immunohistochemistry is used. This report describes a case of angioleiomyoma in which there was a chronically increasing lesion for 5 years on the left hard palate and the means for making a definitive diagnosis was investigated based on previous reports on angioleiomyoma of the palate.

Report of Case

A 79-year-old man was referred to the authors' hospital in October 2009 with a chief complaint of an enlarging asymptomatic mass in the hard palate during the previous 5 years. His medical history showed that he had undergone surgery for anal carcinoma in April 2004, followed by a period of no evidence of local recurrence or metastatic spread at repeated imaging. There was no other medical history of note. On intraoral examination, a palpable firm mass, measuring 15 × 15 mm, was found on the left hard palate (Fig 1). The overlying mucosa of the round, painless mass was normal. To exclude the possibility of malignant salivary gland neoplasms, fine-needle aspiration biopsy was performed and was negative for any malignant findings and for any liquid or viscous contents. A panoramic radiograph showed no obvious odontogenic source of infection. Computed tomographic

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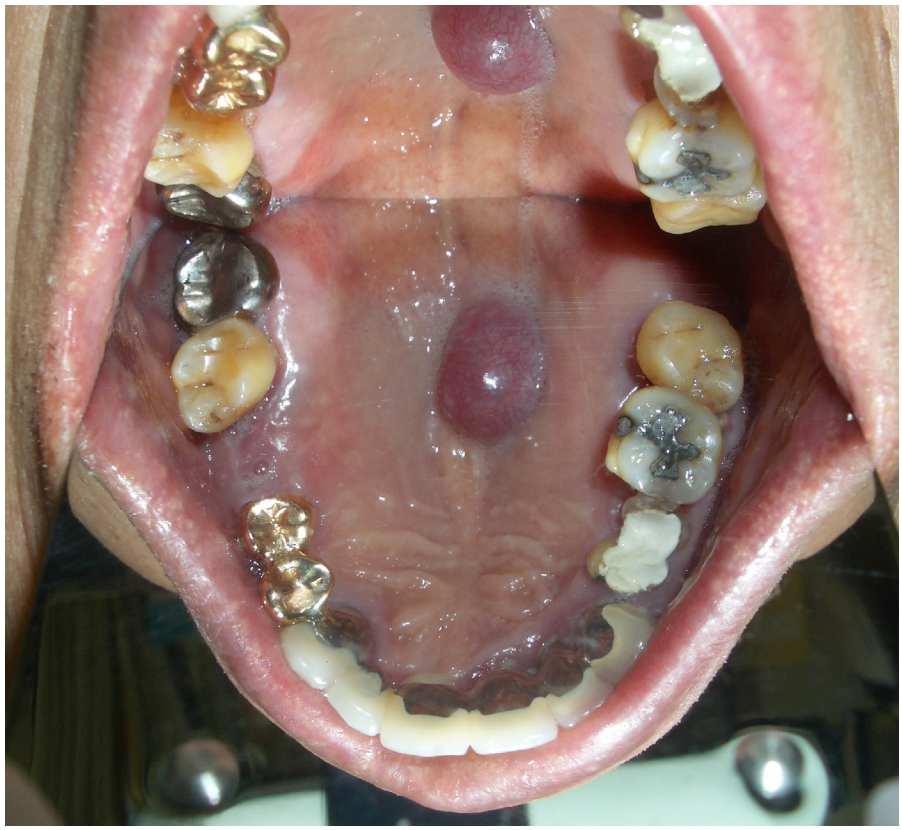

FIGURE 1. Mirror photographic image at first visit.

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(CT) images showed a 16 × 12 × 9 mm well-circumscribed mass in the left hard palate without any evident surrounding bony resorption (Fig 2).

A soft tissue lesion was suspected and surgical excision was planned. The procedure was performed in October 2009 under local anesthesia. The well-defined mass and certain peripheral tissues were excised in subperiosteal fashion, followed by curettage of the bottom of the palate owing to slight osseous erosion that was too small to be detected on CT images. The specimen was a solid and encapsulated mass that included vessels (Fig 3). The cut surface of the excised specimen was white and smooth (Fig 4). The surgical site was irrigated with sterile saline, sutured with an artificial collagen coating, and covered with a protection plate. Hematoxylin and eosin (H-E) staining of the section showed that the well-circumscribed lesion was surrounded by a capsule containing numerous vessels. Smooth muscle and collagen fibers were present between the vessels (Fig 5). Thick bands of smooth muscle cells were surrounded by vascular spaces (Fig 6). Furthermore, the section was subjected to testing for the smooth muscle immunohistochemical markers desmin and α-smooth muscle actin (SMA). The cells were positive for desmin and SMA, consistent with angioleiomyoma (Fig 7). During the 6-month follow-up period, excellent healing was observed with no recurrence.

Discussion

Angioleiomyoma is a benign soft tissue tumor that is microscopically characterized by the proliferation of smooth muscle cells intermingled with abundant vascular channels.[1] It also is described as arising from the tunica media of the blood vessel wall.[1] The incidence of this tumor in the oral cavity is rare and has been estimated to be approximately 0.065%.[4] Oral angioleiomyoma is found mostly in the lips, palate, buccal mucosa, and tongue.[3] As far as the authors have been able to determine from the literature, including the present case, 18 cases of angioleiomyoma of the hard palate have been reported.[2,3,5-18]

The authors' examination of the reports on this tumor in the hard palate found that the age ranged from 28 to 79 years (average age, 53.9 yr), and there was a slightly increased incidence in men (male-to-female ratio, 10:7). The typical clinical presentation of the lesion is a small, solitary, slowly developing mass.[1] Clinical symptoms most commonly consist of an asymptomatic mass, followed by pain[10,17] (cases 7 and 17), ulceration[3] (case 9), and difficulty in chewing and