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CASE REPORT

Congenital angiomyoma of the tongue: case report

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Angiomyomas of the oral cavity are rare benign vascular neoplasms. In particular, the congenital form has not been reported before in the English language literature. We present a congenital angiomyoma of the tongue that was found on the posterior middle of the tongue in an infant. On MRI, the mass showed an isointense signal to muscle on the T_1 weighted image and a slightly hyperintense signal on the T_2 weighted image. Immunohistochemically, tumour cells were positive to desmin and smooth muscle actin, but negative to vimentin and S100. The treatment was surgical excision and no recurrence was found during the 26 month follow-up period.
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Introduction

Angiomyoma, sometimes called angioleiomyoma, is a benign smooth muscle neoplasm that can be classified as three histological types on the basis of their cytoarchitecture: solid, venous and cavernous subtypes.[1] Most angiomyomas have been observed in the subcutaneous tissue of the lower extremities.[1] It is very rare in the oral cavity, especially in the tongue.[2] In a previous study, the incidence of oral angiomyoma was reported as 2.7%.[3] In a study of the head and neck, angiomyoma was not found in the tongue but was found in the lip in three cases.[4] Another study reports that out of 12 oral lesions only 1 was found on the tongue.[5]

Oral angiomyomas are usually found between the ages of 30 and 50 years and are rarely seen in children.[4,6] It is very hard to decide the proper time for surgical resection of the congenital lesion. In the case of congenital angiomyoma, it is very hard to find any information for treatment timing because of its rarity. If there is a congenital tongue mass in a neonate, radiographic findings are especially important because of the risk of general anaesthesia when performing the biopsy.[7] We experienced a case of congenital angiomyoma in the tongue and present it along with a literature review.

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Case report

A 2-month-old infant presented with a congenital soft tissue mass in the posterior tongue. The mass was sessile and covered by reddish mucosa and the appearance was not pathognomonic. As the condition of the patient did not allow an incisional biopsy under local or general anaesthesia, MRI was performed under sedation. MRI showed an elevated mass on the base of the tongue (Figure 1). It also showed an isointense signal to muscle on T_1 weighted images and a slightly hyperintense signal with a hyperintense rim on T_2 weighted images. The tentative diagnosis was a benign lesion of the tongue. The mass was excised under general anaesthesia when the patient was 6 months old.

The mass size was approximately 2.5×2.0 cm. Histopathologically, the lesion was partially circumscribed and composed of variably prominent and thickened vessel walls consisting of benign and mature smooth muscle cells (Figure 2a). The smooth muscle bundles were interspersed with collagen fibres and tumour cells were spindle shaped with elongated nuclei (Figure 2b). There were few mitotic figures. Masson trichrome (MT) staining showed that the muscle fibre of the lesion stained a purple colour, which indicated the presence of myofibrils in the tumour cells (Figure 2c). In the immunohistochemical studies, the tumour cells showed a positive reaction to desmin (Figure 2d) and smooth muscle actin (SMA) (Figure 2e), but the tumour

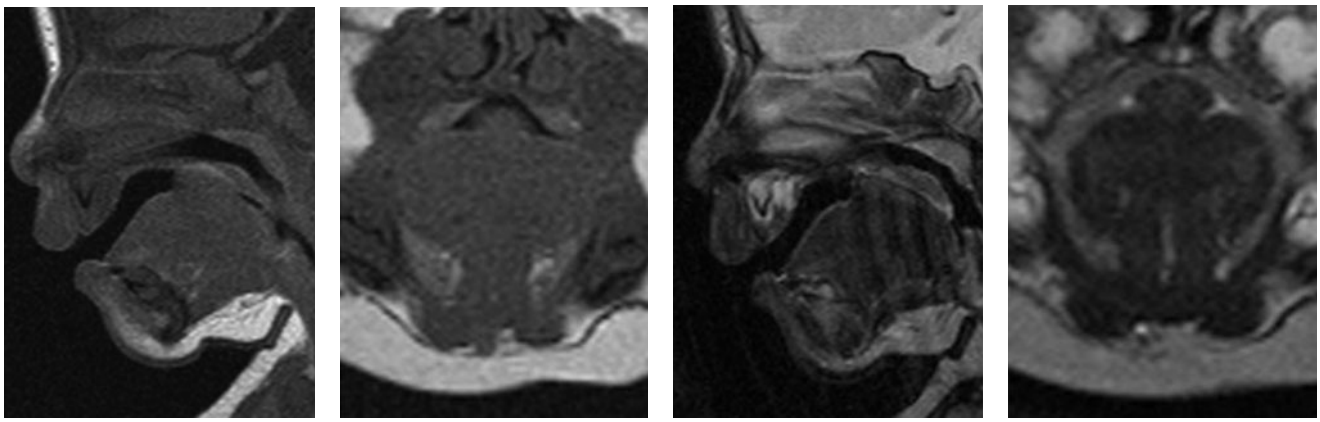

Figure 1 Pre-operative MRI. (a, b) T_1 weighted images show an isointense signal to muscle and a well-defined margin. (c, d) T_2 weighted images show a slightly hyperintense signal with a partially hyperintense rim

cells reacted negatively to vimentin (VMT) and S100 (Figure 2 f,g). The final diagnosis was angiomyoma.

The post-operative course was uneventful, and there was no evidence of recurrence 26 months post-operatively.

Discussion

Angiomyoma is usually found in the subcutaneous tissue of the extremities and is rarely found in the oral cavity.[8] In the case of lesions in the oral cavity, it is frequently reported on the lip.[5] The mean prevalent age for oral angiomyoma is the early fifth decade.[3,5] To the best of our knowledge, our case of congenital angiomyoma is the first case report in an English language journal.

Characteristically, angiomyoma on MRI can be described as a well-demarcated, strongly enhancing mass with an isointense to slightly hyperintense signal compared with muscle on T_1 weighted images and hyperintense signals on T_2 weighted images.[8] The MRI findings of our case were in accord with the findings in

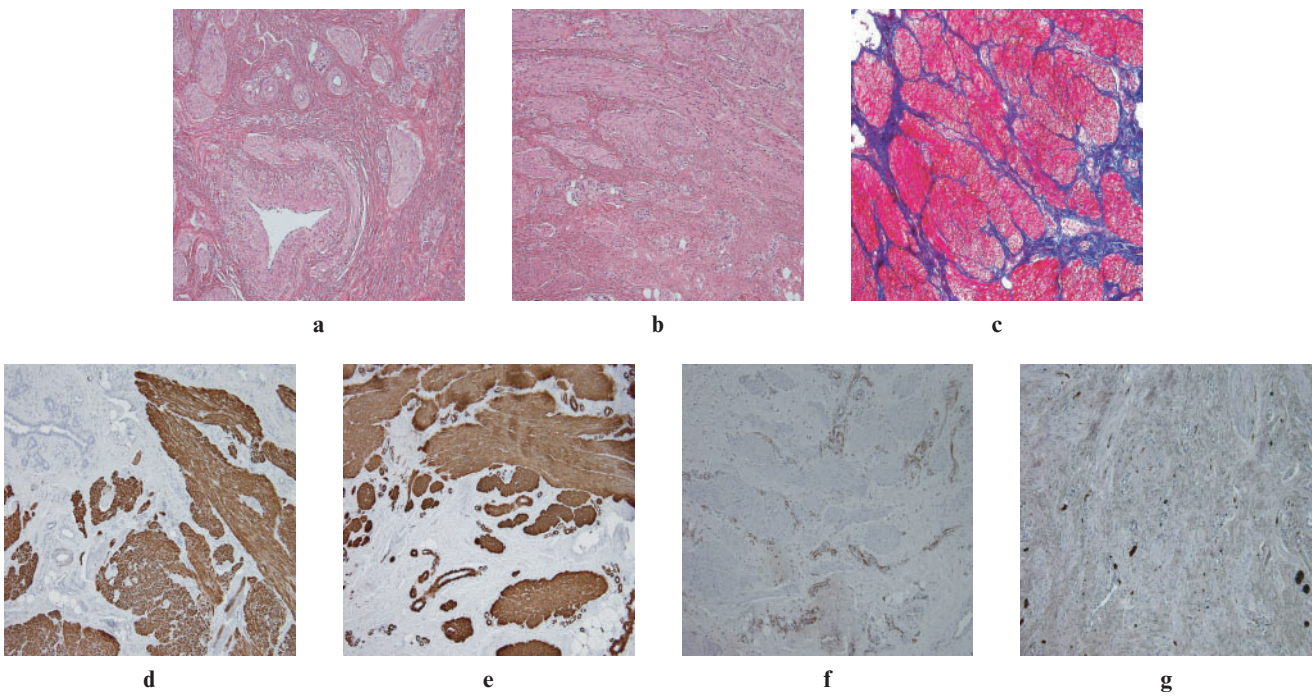

Figure 2 (a) The islands of tumour cells were scattered and the vessel walls were thickened (haematoxylin and eosin, original magnification ×100). (b) The smooth muscle bundles were interspersed with collagen fibres and tumour cells were spindle shaped with elongated nuclei (haematoxylin and eosin, original magnification ×200). (c) Masson trichrome staining revealed red in tumour cell nests (original magnification ×100). (d) The tumour cells were positive to desmin (original magnification ×100). (e) The tumour cells and vessel walls had a positive reaction to smooth muscle actin (original magnification ×100). (f) The tumour cells were negative to vimentin (original magnification ×100). (g) The tumour cells were negative to S100 (original magnification ×100)

the literature.[8] On the basis of MRI findings, a vascular lesion and benign tumour were suspected. As we had already ruled out malignant tumour, we delayed the biopsy until the risk of general anaesthesia was reduced. Therefore, the radiographic findings of congenital lesions are very important in neonates who cannot receive an incisional biopsy. All vascular lesions, except for lymphatic malformations, are isointense or hyperintense on T_1 weighted images and markedly so on T_2 weighted images.[9] However, the MRI findings of our case showed slightly hyperintense signals, unlike ordinary vascular lesions (Figure 1).

Histologically, the proliferation of smooth muscle cells may resemble other benign spindle-shaped lesions, such as haemangiopericytoma, neurofibroma, neurilemmoma, myofibroma and nodular fasciitis. Special stains like MT or phosphotungstic acid haematoxylin (PTAH) and immunohistochemical stains such as SMA, desmin and VMT can be helpful in the diagnosis. In our case, tumour cells showed positive reactivity to desmin and SMA, and the muscle fibres in the tumour were stained purple in the MT stain (Figure 2). These characteristics demonstrated that the tumour cells originated from primitive muscle cells. The final diagnosis was confirmed as angiomyoma.

Angiomyoma is a slow-growing, firm, elastic, asymptomatic tumour.[10] Differential diagnosis includes benign lesions such as fibroma, haemangioma, lymphangioma, teratoma, neurofibroma, granular cell tumour and minor salivary gland tumours. The radiographic study is highly valuable to differentiate the vascular lesion from the others. The vascular lesion from the lymphatic channel will show mixed intensity on MRI without flow voids.[11] Fast-flow lesions such as arteriovenous malformation will show many flow voids on MRI.[12] Our case did not show prominent flow voids but showed slightly hyperintense signals unlike ordinary vascular lesions on an MRI (Figure 1). The different findings of our case may be due to the immature vessel of the congenital lesion. As congenital angiomyoma has not been presented before, the radiographic and the immunohistochemical features would be valuable for future differential diagnosis.

The treatment for oral angiomyoma is usually surgical excision and recurrence is extremely rare, regardless of the pathological subtype.[3] Our case also showed a slow growth rate and showed no recurrence at 26 months' follow-up and there were no complaints. Therefore, congenital angiomyoma might have the same prognosis as oral angiomyoma and urgent resection might not be needed. As this case report was based on a single case, in order to determine the optimum time for resection of congenital angiomyoma and its prognosis, further case reports or case series with long-term follow-up should be encouraged.

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