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CASE REPORT

Rhabdomyomatous (mesenchymal) hamartoma of the tongue: report of a case

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A 12-year-old girl presented with three yellowish polypoid lesions in the lateral border of tongue since birth. Histological examination showed oral mucosa covered by squamous epithelium and an underlying vascularized fibrous stroma, containing small and short bundles of striated muscle, corresponding to a rhabdomyomatous hamartoma.
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Case report

A 12-year-old white female was seen at the outpatient clinic for a plantar wart. On physical examination, a group of three small (4–5 mm diameter), smooth, yellowish polypoid lesions on the lingual right lateral border was also observed (Fig. 1). They were neither painful nor pruritic. The rest of the oral mucosa was otherwise normal. The patient's father referred that these lesions were present since birth, and a picture at age 6 months was available (Fig. 2).

One lesion was excised under local anaesthesia. Its histological examination showed oral mucosa covered by squamous epithelium, a vascularized fibrous stroma, with teleangiectatic vessels, and interwoven small and short bundles of striated muscle (Fig. 3), corresponding to a rhabdomiomatous hamartoma.

After a 1-year clinical follow-up, no recurrence has been observed.

Comments

The term hamartoma defines a tumour-like lesion composed of a mixture of mature tissues, haphazardly arranged. Hamartomas located in the lingual region are very rare, with only a few cases reported in the literature. Rhabdomyomatous hamartoma is a rare skin congenital lesion occurring particularly in the face and

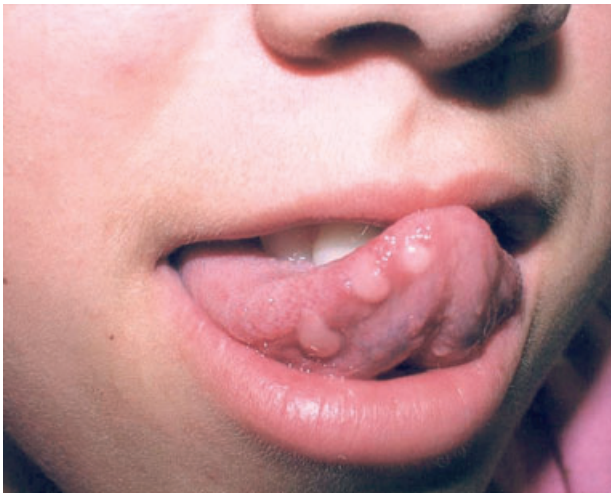

Figure 1 Patient at 12 years of age showing grouped nodules on the lateral border of the tongue.

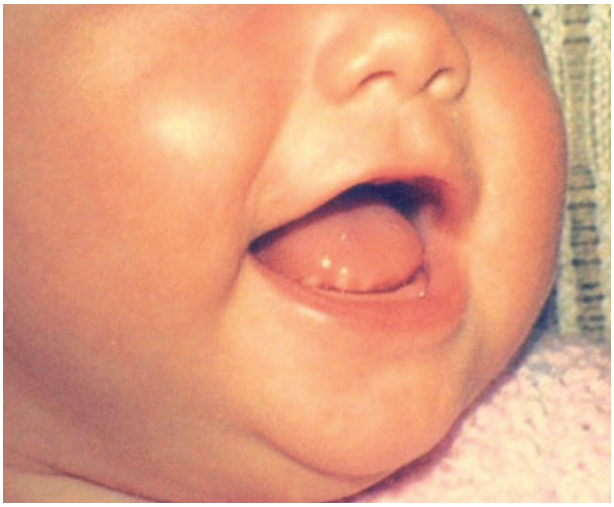

Figure 2 Patient at 6 months of age showing lesions that have been present since birth.

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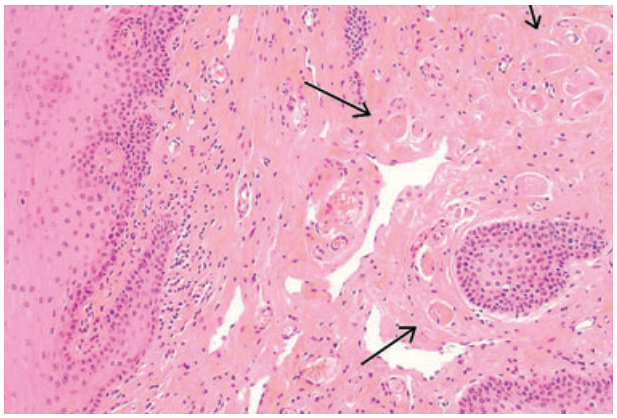

Figure 3 Microphotograph showing vascularized fibrous stroma, some telangiectasia, and interwoven small and short bundles of striated muscle (arrows) (haematoxylin and eosin, 200×).

neck, and usually having a midline location (eyelids, chin, anterior neck, nose and upper lip). These lesions are congenital, sometimes multiple and can exhibit voluntary movement (1, 2). The aetiology of rhabdomyomatous hamartoma is still unclear (2). One case has been published presenting as an intra-oral striated muscle hamartoma that, as the current case, was located on the lateral border of the tongue. However, unlike our patient, the lesion was acquired at the age of 11 years, suggesting that the lesion could represent an herniation of the lingual striated muscle through a weakened point of the overlying mucosa and not an actual hamartoma (3). There is another report of two cases of rhabdomyomatous hamartoma arising as polypoid masses of oral cavity, one of them localized in the tongue, as in our case. This is an unusual site for such a lesion, as most cases of rhabdomyomatous hamartoma reported in the literature have typically been located in the skin (2).

We report this case because of its very low incidence, and to include the striated muscle hamartoma in the differential diagnosis of lesions affecting the lateral border of tongue, namely haemangiomas, lymphatic malformations, dermoid cysts, thyroglossal duct cysts, lingual thyroid, granular cell tumour, heterotopic gastric mucosa cyst and other hamartomas (4).

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