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CASE REPORT

Lingual striated muscle hamartoma or herniation?

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Three grouped, small polypoid lesions were removed from the right lateral border of tongue of a healthy male aged 12 years. They were composed of packed, mature striated muscle fibres covered by oral epithelium and thinned lamina propria. Hamartomatous growth of striated muscle, or herniation through underdeveloped lamina propria is postulated to explain the exceedingly rare clinicopathological features.
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A 12-year-old Caucasian male was referred to the Oral Medicine Department at Liverpool University Dental Hospital for multiple, small swellings on the side of his tongue. These had been present for approximately 12 months and had been gradually increasing in size. There was no history of trauma, discomfort, bleeding or ulceration from the affected site.

The patient was fit and well and had no relevant medical history or evidence of skin lesions.

On intra-oral examination, a group of three small (2–3 mm diameter), pedunculated, smooth, white, polypoid lesions were seen on the right postero-lateral border of his tongue (Fig. 1). There was no obvious trauma associated with these lesions. The rest of the oral mucosa appeared normal.

The lesions were excised under local anaesthesia and sent for histological examination in the Oral Pathology laboratory. Routinely prepared sections showed that the bulk of the polypoid lesions were composed of packed, mature, striated muscle fibres covered by parakeratinized squamous epithelium and thin lamina propria. Hyperaemia and a sprinkling of chronic inflammatory cells were seen subepithelially (Fig. 2).

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Comments

Benign polypoid lesions containing mature striated muscle are exceedingly rare, which allows only speculation about the nature and aetiology of the pathology described here.

Two hypotheses are proposed; the first suggests that the present lesions result from hamartomatous overgrowth of striated muscle. The multiple, albeit grouped, lesions, age of patient and the maturity of muscle fibres support the hamartomatous hypothesis. Twenty-four cases of striated muscle hamartomas, also known as rhabdomyomatous mesenchymal hamartomas, have been recently reviewed (1, 2). These polypoid tumours are usually found in the eyelids, chin, anterior neck and nose (1–3). A case from the upper lip has been reported (4), but none from an intra-oral site. However, rhabdomyomatous mesenchymal hamartomas occur in neonates, have a midline location and histologically the striated muscle are set in fibrofatty tissue with blood vessels (1). Although it is difficult to reconcile these features with our case, the hamartomatous hypothesis remains plausible. Further, support for this is given by the site, because overgrowth of other tissue elements (e.g. subgemmal nerve plexus) has been reported on the lateral border of the tongue (5).

The second hypothesis suggests that the lesions of our patient represent herniation of lingual striated muscle

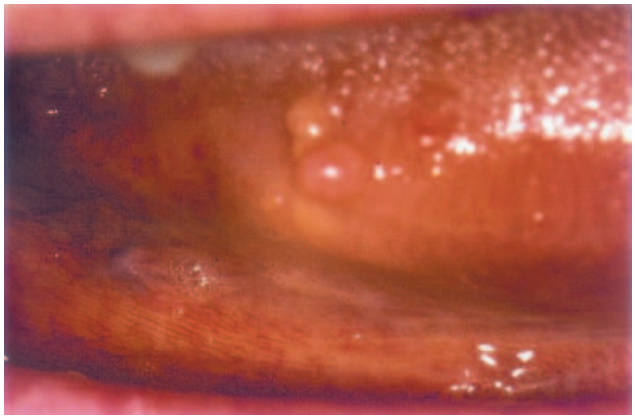

Figure 1 Close-set nodules along the margin of the tongue.

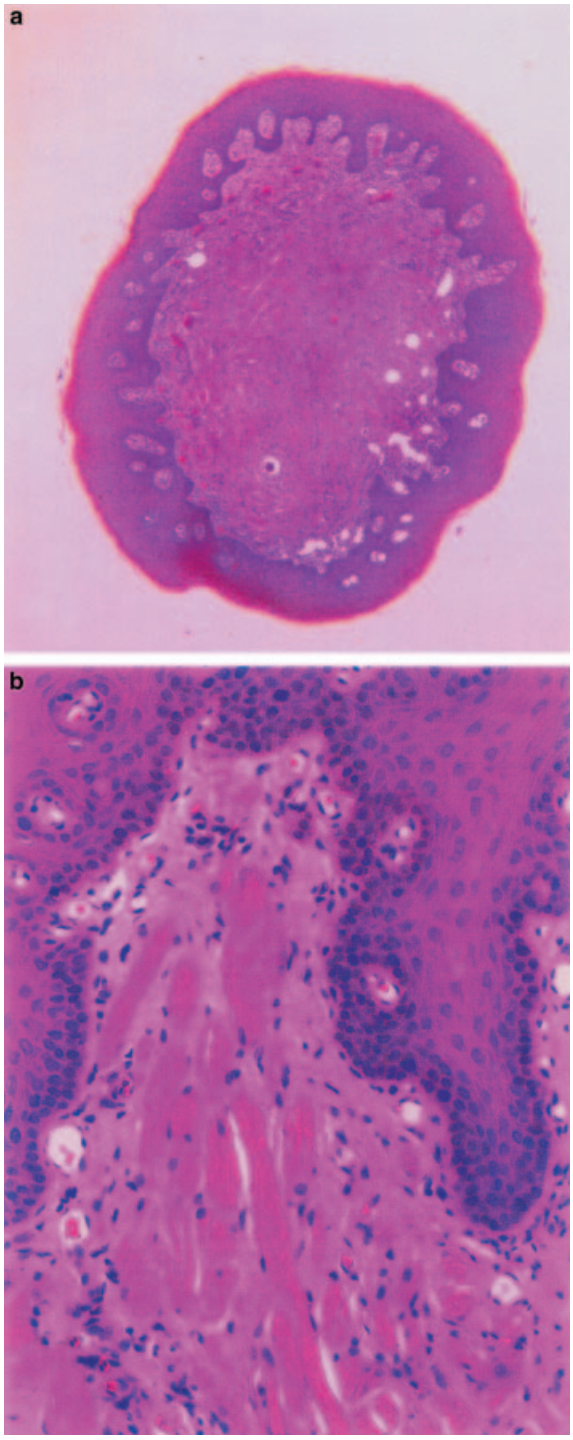

Figure 2 (a,b) The transverse section shows an intensely 'pink' nodule at scanning magnification. It is composed of normal-appearing skeletal muscle fibres oriented in various directions and of scant intervening stroma. There is no capsule and the fibres are separated from the epithelium by a thin vascular and inflamed zone, although some extend to its undersurface. That is seen better in (b), taken at higher magnification, wherein the characteristic arrangement of the nuclei is also discernible (haematoxylin and eosin).

through a weakened point of overlying mucosa or at a site of injury. Weakening of oral mucosa would be expected to occur in a congenital malformation affecting particularly the lamina propria and effecting its thinning. Our finding of striated muscle fibres running in a very superficial location beneath the lingual epithelium would be consistent with thinned and possibly underdeveloped lamina propria. Underdeveloped lamina propria and dermis are features of focal dermal hypoplasia (6), but our patient had no clinical evidence of this or any other syndrome. Nevertheless, focal underdevelopment/thinning of lamina propria is still a plausible explanation, although its aetiology is obscure. The clinical history of the patient does not give credence to the notion of lingual striated muscle protruding at a site of injury or minor trauma. The hyperaemic and mild inflammations found below the covering epithelium are secondary and more likely to be due to rubbing/inadvertent chewing of the lesions rather than factors of aetiological significance.

Although definite conclusions cannot be drawn concerning the aetiology of this rare clinical and histopathological presentation, the constellation of features are of interest. Lingual striated muscle hamartoma, or herniation, should be considered in the differential diagnosis of lesions affecting the lateral border of tongue.

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