doi: 10.1111/j.1600-0714.2007.00518.x

J Oral Pathol Med (2007) 36: 241–4

www.blackwellmunksgaard.com/jopm

CASE REPORT

Multiple leiomyomatous hamartoma in the oral cavity

Seiji Iida[1], Mitsunobu Kishino[2], Hitomi Senoo[1], Masaya Okura[1], Ichijiro Morisaki[3], Mikihiko Kogo[1]

[1]*The First Department of Oral and Maxillofacial Surgery, Osaka University Graduate School of Dentistry, Osaka;* [2]*The Department of Oral Pathology, Osaka University Graduate School of Dentistry, Osaka;* [3]*Division of Special Care Dentistry, Osaka University Graduate School of Dentistry, Osaka, Japan*

Leiomyomatous hamartoma (LH) is congenital lesion rarely seen in oral cavity. In English literature, all reported cases appeared as solitary lesion in alveolar ridge or the tongue, and there have never been a report showing a case of multiple occurrence of this lesion. A quite rare case of multiple LH occurred in a 2-year-old Japanese boy is presented. A polypoid lesion was presented at incisive papilla and two isolated lesions in the tongue dorsum, one appeared as a polypoid mass in the posterior dorsum and other as a small spindle-shaped agger in the anterior dorsum. All of them were histologically diagnosed as LH.
J Oral Pathol Med (2007) **36**: 241–4

Keywords: congenital epulis; leiomyoma; leiomyomatous hamartoma; polyp

Case report

A 2 years 7-month-old Japanese boy was referred to Osaka University Dental Hospital for evaluation and treatment of a polypoid mass on the palatal surface at the incisive papilla. He had delivered normally full-term and there were no abnormal events during pregnancy and history of any congenital anomalies in her family. According to his mother, the lesion was first clearly noticed at 2 years 5 months old and was slowly growing. Another polypoid mass was also presented in the dorsal surface of the posterior tongue, which had been pointed by his paediatric doctor at 4 months old.

Intra-oral examination revealed the presence of polypoid lesion with elastic softness, measuring $5 \times 3 \times 4$ mm at the incisive papilla (Figure 1). The left upper primary central incisor was slightly dislocated towards the labial side. In the midline of the tongue dorsum, two lesions were observed; one presented posterior part, which had been pointed out previously, and another in the anterior dorsum of the tongue (Fig. 2). Former lesion appeared as a polypoid lesion with a smooth surface and the size of the mass was $4 \times 4 \times 3$ mm. The latter was presented as a small spindle-shaped agger with rather smooth surface and the size was 2 mm in diameter. Any indurations in the tongue beneath both lesions were not palpable. The tightened lingual and upper labial frenulums were also seen. Radiographic examination of the maxillary alveolar ridge did not reveal the presence of any radiopaque or radiolucent lesions. Under the clinical diagnosis as multiple benign tumours, the excision was performed under general anaesthesia.

Tissues obtained by surgery were fixed in 10% buffered formalin followed by paraffin embedding. Histological sections from paraffin-embedded blocks were stained with haematoxylin and eosin (HE). Microscopically, the lesions were composed of proliferating mesenchymal components that contained mainly smooth muscles admixed with collagen fibres, nerve fibres and small vessels (Fig. 3a–c). Furthermore, mucous salivary glands were detected in the lesion at the incisive papilla (Fig. 3d). Immunohistochemical stain was performed on the serially prepared sections by streptavidin–peroxidase complex technique. The mesenchymal components were almost all positive for vimentin (V9, pre-diluted; Dako, Glostrup, Denmark), smooth muscles were positive for α-smooth muscle actin (1A4, 1:200; Dako) and desmin (D33, 1:100, Dako; Fig. 3e), and nerve fibres scattered throughout the lesions were positive for S100 protein (polyclonal, pre-diluted, Dako; Fig. 3f). From these findings, the lesions were diagnosed as leiomyomatous hamartoma (LH). Post-operative course was uneventful and no recurrence was observed.

Comments

Hamartoma is a tumour-like malformation composed of a focal overgrowth of mature normal cells located where they are normally found and the intra-oral hamartoma is a rare lesion (1). Hamartoma can be histopathologically classified according to main components, and LH is defined as the hamartoma composed only or predom-

Correspondence: Seiji Iida, The First Department of Oral and Maxillofacial Surgery, Osaka University Graduate School of Dentistry, 1-8 Yamadaoka, Suita, Osaka 565-0871, Japan.
Tel: +81 6 6879 2936, Fax: +81 6 6876 5298,
E-mail: iida@dent.osaka-u.ac.jp
Accepted for publication December 11, 2006

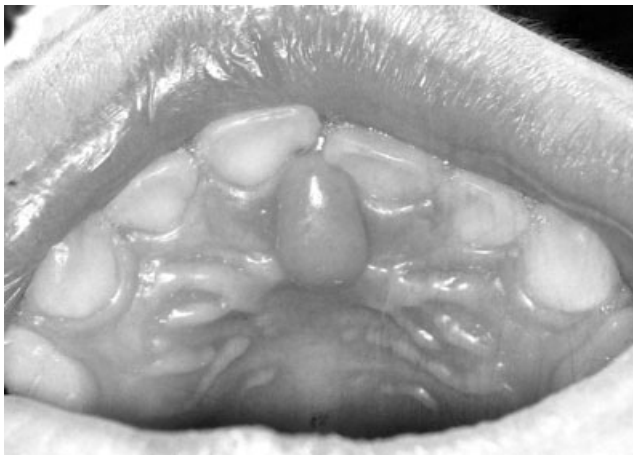

Figure 1 Clinical feature of the lesion located in the median maxillary alveolar mucosa. The lesion was located at the incisive papilla and the size of the lesion was $5 \times 3 \times 4$ mm.

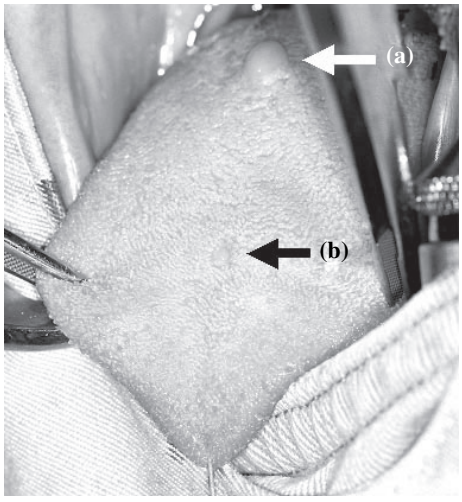

Figure 2 Clinical feature of the lesions in the midline of dorsum of the tongue. The larger lesion was located in the posterior dorsum as polypoid mass measuring $4 \times 4 \times 3$ mm (a). The smaller lesion was located at the anterior dorsum of the tongue, presented as a small spindle-shaped agger with the smooth surface and the size was 2 mm in diameter (b).

inantly of smooth muscle. LHs are often found in lung or kidney, but the case observed in the oral cavity is quite rare. In English-language literature, only nine cases have been reported; six cases in the maxilla (2–7) and three in the tongue (1, 8, 9; Table 1). All of them appeared as a solitary lesion and there have never been a report showing multiple occurrence of LH, except a case reported in Japanese literature (10).

There are some differences in the location of the lesion among past reported six cases observed in the maxilla. In four cases (2, 3, 5, 6), the lesion was located in the midline of the gingiva on the palatal side, but, in the case reported by Takeda et al. (4), the lesion was located in that on the labial side. In the case reported by Napier et al. (7), the lesion was located in hard palate beside the midline. On the other hand, among three reported cases of the lesion in the tongue, two were seen in posterior dorsum (8, 9) and one in the tongue tip (1). In our case, the larger polypoid lesion was presented in the posterior tongue dorsum and the smaller lesion was in the center of the tongue dorsum. In the case of multiple LH reported by Kanekawa (10), lesions were located in midline of the alveolar gingiva on the palatal side and the posterior tongue, and these clinical features are resembled to our case, except the multi-occurrence of the tongue lesion.

In clinical, any intra-oral lesions located in or near the midline of the tongue and the anterior alveolar ridge of the maxilla with a polypoid or pedunculated solid mass in infants require differential diagnosis from LH. Among intra-oral congenital lesions appeared as a solid mass, the choristoma is rather common lesion than hamartoma, though its incident is also low. This lesion is defined as a cohesive tumour-like mass consisting of normal cells in an abnormal location and shows tumour-like growth and, according to main component cell, this lesion is histopathologically classified into some variants and the osseous choristoma, cartilaginous choristoma and lingual thyroid choristoma are rather common lesion in oral cavity (11). Among them, the osseous choristoma predominantly occurs in the dorsum of the posterior third of the tongue, and often appears as a pedunculated mass (11). However, choristomas are seen in rather elderly group because they show the tumour-like growth (11), which is different from the clinical feature of hamartomas.

On the other hand, most common and well-known congenital intra-oral solid mass is 'congenital epulis'. This lesion is mainly found in the maxillary alveolar ridge and rarely occurs in the tongue (12). However, these cases were observed mainly in anterior ventral surface of the tongue (12). Fibrous lesions also appear as well circumscribed solid mass in younger children. These lesions are mainly caused by chronic inflammation or mechanical irritation, such as digit sucking and bottle feeding. Most of fibrous lesions are reported as epulides (13) but often as fibroepithelial polyp (13). The clinical features of the reported three cases of palatal fibro-epithelial polyp by Tomizawa et al. (13), were quite resembled to that of the present case.

Histopathologically, LH should be distinguished from some tumoral lesions composed predominantly of smooth muscle. Leiomyoma in the oral cavity can be divided into two categories, one is angioleiomyoma and another is solid leiomyoma. The former lesion is more common in oral cavity and is easy to distinguish microscopically, due to the histological appearance, a well-demarcated nodule composed a plethora of thick-walled muscular vascular channels and interspersed fibrous connective tissue (1, 7). To the contrary, solid leiomyoma consists of poorly defined mass of interlacing bundles of smooth muscle in a fibrous stroma, which is similar to LH. To distinguish them, the difference of clinical features of two lesions is important, because the most of solid leiomyomas are found in adult and the occurrence in infant is quite rare (2, 14). Some reports (1, 9) indicated that the presence of salivary gland tissue

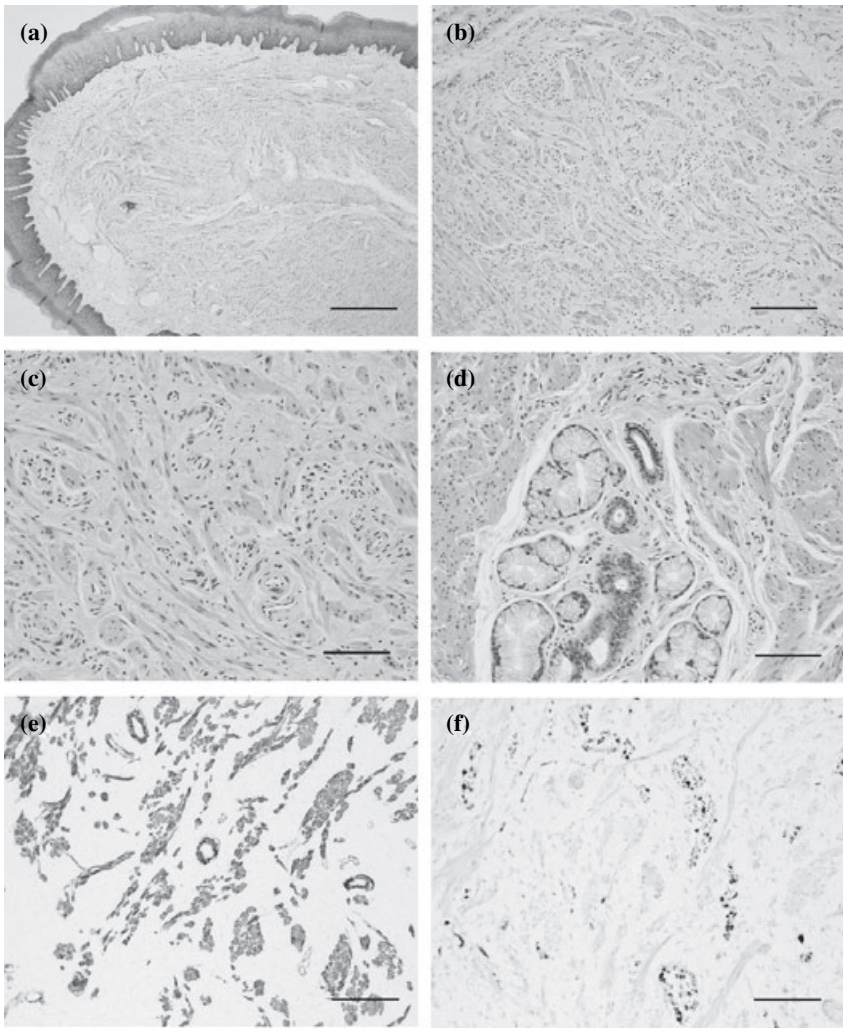

Figure 3 Histological findings of the lesions. All lesions are exophytic mesenchymal masses (a), mainly composed of smooth muscles admixed with collagen fibres, nerve fibres and small vessels (b and c). Mucous salivary glands are presented in the lesion at the incisive papilla (d). Immunohistochemically, the mesenchymal components are positive for α-smooth muscle actin (e) and staining for S100 protein reveals many peripheral nerve fibres intermingled with the proliferating smooth muscle cells (f; a–d, HE; a, bar 500 μm; b, bar 200 μm; c–f, bar 100 μm).

Table 1 Leiomyomatous hamartomas reported in the English-language literature

Authors	*Sex*	*Age*	*Location (number of lesions)*
1. Ng et al. (2)[a]	Female	3 months	Median maxillary alveolar (1)
2. Semba et al. (3)	Male	2 years	Median maxillary alveolar (1)
3. Takeda et al. (4)	Male	10 months	Median maxillary alveolar (1)
4. Correa et al. (5)	Female	6 years	Median maxillary alveolar (1)
5. Kujan et al. (6)	Female	11 months	Median maxillary alveolar (1)
6. Napier et al. (7)	Female	5 years	Left hard palate (1)
7. Goldsmith et al. (8)	Male	1 year	Posterior dorsum of the tongue (1)
8. Kobayashi et al. (9)	Male	3 months	Posterior dorsum of the tongue (1)
9. de la Rosa-Garcia and Mosqueda-Taylor (1)	Male	6 years	Tip of the tongue (1)
10. Present case	Male	2 years	Dorsum of the tongue (2) and median maxillary alveolar (1)

[a]The case was reported as leiomyoma in the original literature, but this can be represented as leiomyomatous hamartoma.

is an important microscopic feature in LH to distinguish from leiomyoma, though only three lesions (7, 8) including our case showed its presence. Because leiomyomas do not contain numerous nerve fibres (4), the microscopic feature of many peripheral nerve fibres intermingled with the proliferating smooth muscle cells, examined by immunochemical staining with antibody for S100, is now the important information (3, 14).

Incidentally, the case of leiomyoma in infant, reported by Ng et al. (2), have been suggested to be represented as LH (3, 5, 7). Leiomyosarcoma is also the lesions requiring the histological differential diagnosis. Leiomyosarcoma is microscopically more cellular, nuclear pleomorphism, mitotic acting and local invasion, and these histological features can be easily distinguished from LH as well as the clinical feature showing aggressive growth (7).

Various intra-oral lesions may be presented as pedunculated lesion in infant. When the clinical diagnosis as congenital disease is suspected for the lesion, detailed oral examinations for another location should also be performed, because of the possibility of multiple occurrence of such disease.

References

1. de la Rosa-Garcia E, Mosqueda-Taylor A. Leiomyomatous hamartoma of the anterior tongue: report of a case and review of the literature. *Int J Paediatr Dent* 1999; **9**: 129–32.
2. Ng KH, Siar CH, Abdul Latif H. Leiomyoma of the incisive papilla region: a case report. *Ann Dent* 1992; **51**: 29–31.
3. Semba I, Kitano M, Mimura T. Gingival leiomyomatous hamartoma: immunohistochemical and ultrastructural observations. *J Oral Pathol Med* 1993; **22**: 468–70.
4. Takeda Y, Satoh M, Nakamura S, Matsumoto D. Congenital leiomyomatous epulis: a case report with immunohistochemical study. *Pathol Int* 2000; **50**: 999–1002.
5. Correa L, Lotufo M, Martins MT, Sugaya N, de Sousa SC. Leiomyomatous hamartoma of the incisive papilla. *J Clin Pediatr Dent* 2001; **25**: 157–9.
6. Kujan O, Clark S, Sloan P. Leiomyomatous hamartoma presenting as a congenital epulis. *Br J Oral Maxillofac Surg* 2005; Sep 12; (Epub ahead of print).
7. Napier SS, Devine JC, Rennie JS, Lamey PJ. Unusual leiomyomatous hamartoma of the hard palate: a case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1996; **82**: 305–7.
8. Goldsmith P, Soames JV, Meikle D. Leiomyomatous hamartoma of the posterior tongue: a case report. *J Laryngol Otol* 1995; **109**: 1190–1.
9. Kobayashi A, Amagasa T, Okada N. Leiomyomatous hamartoma of the tongue: case report. *J Oral Maxillofac Surg* 2001; **59**: 337–40.
10. Kanekawa A. A case of hamartoma occurring in tongue and upper gingival. *Jpn J Oral Maxillofac Surg* 1990; **36**: 317–20.
11. Chou LS, Hansen LS, Daniels TE. Choristomas of the oral cavity: a review. *Oral Surg Oral Med Oral Pathol* 1991; **72**: 584–93.
12. Loyola AM, Gatti AF, Pinto DS Jr, Mesquita RA. Alveolar and extra-alveolar granular cell lesions of the newborn: report of case and review of literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1997; **84**: 668–71.
13. Tomizawa M, Kohno M, Noda T, Suzuki M. Three cases of palatal polyps in infants. *Int J Paediatr Dent* 1999; **9**: 213–7.
14. Baden E, Doyle JL, Lederman DA. Leiomyoma of the oral cavity: a light microscopic and immunohistochemical study with review of the literature from 1884 to 1992. *Eur J Cancer B Oral Oncol* 1994; **30B**: 1–7.