The Journal of Laryngology and Otology
December 1995, Vol. **109**, pp. 1190–1191

Leiomyomatous hamartoma of the posterior tongue: a case report

P. GOLDSMITH*, M.A., B.M., B.CH., J. V. SOAMES†, PH.D., F.D.S., F.R.C.PATH., D. MEIKLE‡, F.R.C.S.

Abstract
A case of a rare leiomyomatous hamartoma arising in the posterior tongue of a sixteen-month-old male is reported. There has been no recurrence following simple excision and presenting symptoms of choking on swallowing have resolved. Most other leiomyomatous hamartomas in the upper aerodigestive tract have been reported in Japanese patients and have involved the maxillary gingiva and hard palate.

Key words: Tongue neoplasms; Hamartoma, leiomyomatous

Introduction

Hamartomas, named after the Greek for error and tumour, are focal, tumour-like overgrowths of tissue which are normal to the site of occurrence. Their growth parallels that of the surrounding tissues, although sometimes at a greater rate, but is limited and classically described as stopping when the corresponding normal tissues cease growing, or soon thereafter (Ritchie, 1990). Several common lesions are considered as hamartomatous such as melanotic naevi and angiomatous malformations. However, with the exception of the latter, hamartomas are uncommon in the upper aerodigestive tract, at least as judged from a review of the literature. The purpose of this paper is to report a rare case of a leiomyomatous hamartoma originating in the posterior part of the tongue in a young child.

Case report

An otherwise healthy and developmentally normal sixteen-month-old boy was referred to the ENT Department because of his tendency to choke on swallowing both solid and liquid foods. On examination a pink polypoid mass was evident on the posterior part of his tongue. The lesion was excised. Healing was uneventful and symptoms have resolved.

Histological examination showed a tumour-like mass comprising a core of loose oedematous fibrous tissue running throughout which were randomly orientated fascicles and isolated strands of smooth muscle (Figure 1). A few dilated thin-walled vascular channels and anomalous vessels with thicker walls (including smooth muscle) were also identified, together with normal minor salivary glands. The essential component was the disorganized smooth muscle proliferation consistent with a leiomyomatous hamartoma. There was no evidence of malignancy. Although lesional tissue extended to the base of the lesion, to date, eighteen months after operation, there has been no recurrence.

Discussion

Hamartomas in the oronasopharynx (other than haemangiomas) are uncommon. When present they are usually situated in the midline of the tongue, palate or oropharynx and consist of various components including muscle, fibrous, neural and epithelial tissues (Semba *et al.*, 1993). Majumder *et al.* (1977) reported on an unusual angiomatous hamartoma with areas of cartilage and reviewed the early literature regarding hamartomas of the head and neck and Baille and Batsakis (1974) described a rare glandular (seromucinous) hamartoma of the nasopharynx. The leiomyomatous variety is also rare. Five Japanese cases have been reported involving the maxillary palatal gingiva/anterior palate (Mushimoto *et al.*, 1982; Kajiyama *et al.*, 1983; Kanekawa, 1990; Ng *et al.*, 1992; Semba *et al.*, 1993) and Semba *et al.* (1993) refer to a further case originally reported by Takahashi *et al.* (1962) as a congenital epulis. We have been able to find only one

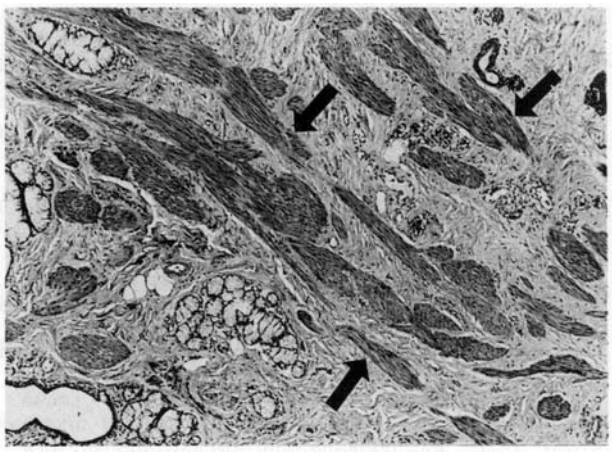

FIG. 1
Leiomyomatous hamartoma showing bundles of smooth muscle (arrows) coursing between lobules of minor salivary glands (Masson trichrome, × 50).

From the National Hospital for Neurology & Neurosurgery*, Queen Square, London, the Department of Oral Pathology†, University of Newcastle upon Tyne, and the Department of Otolaryngology‡, Freeman Hospital, Newcastle upon Tyne.
Accepted for publication: 15 July 1995.

other case reported in the tongue, again in a Japanese patient (Kanekawa, 1990). Reasons for the preponderance of cases in Japanese patients are unknown.

Hamartomas behave in a benign fashion and typically have a limited capacity for growth co-ordinated with that of the host, but in some cases they may be difficult to distinguish from a benign neoplasm. Neoplasms are believed to be caused by mutations, particularly in genes coding for cellular growth and many chromosomal abnormalities have been reported which may account for inappropriate activation of genes. Although hamartomas lack the autonomy for growth of a benign neoplasm, abnormal karyotypes have been detected in some pulmonary (Fletcher *et al.*, 1991; Johansson *et al.*, 1992; 1993) and hepatic (Speleman *et al.*, 1989; Mascarello and Kraus, 1992) hamartomas. These findings lend weight to the concept that some hamartomas may be true neoplasms and Fletcher *et al.* (1991) suggest that the pulmonary hamartoma should perhaps be renamed as 'pulmonary chondroma'. It is possible that when the abnormal karyotype results in loss of control of tissue architecture a hamartoma develops but when control over cell division is affected a neoplasm results. However, whether or not abnormal karyotypes are associated with other hamartomas, particularly those in the oropharyngeal region, is unknown.

References

Baillie, E. E., Batsakis, J. G. (1974) Glandular (seromucinous) hamartoma of the nasopharynx. *Oral Surgery, Oral Medicine, Oral Pathology* **38:** 760–762.

Fletcher, J. A., Pinkus, G. S., Weidner, N., Morton, C. C. (1991). Lineage-restricted clonality in biphasic solid tumours. *American Journal of Pathology* **138:** 1199–2007.

Johansson, M., Dietrich, C., Mandahl, N., Hambraeus, G., Johansson, L., Clausen, P. P., Mitelman, F., Heim, S. (1993) Recombinations of chromosomal bands 6p21 and 14q24 characterize pulmonary hamartomas. *British Journal of Cancer* **67:** 1236–1241.

Johansson, M., Heim, S., Mandahl, N., Johansson, L., Hambraeus, G., Mitelman, F. (1992) t(3;6;14) (p21; p21;q24) as the sole clonal chromosomal abnormality in a hamartoma of the lung. *Cancer Genetics and Cytogenetics* **60:** 219–220.

Kajiyama, M., Iino, E., Kurokawa, H., Fukuyama, H., Ueno, M. (1983) Leiomyomatous hamartoma in the region of the upper medial alveolar gingiva: report of a case. *Japanese Journal of Oral and Maxillofacial Surgery* **29:** 1520–1524.

Kanekawa, A. (1990) A case of hamartoma occurring in tongue and upper gingiva. *Japanese Journal of Oral and Maxillofacial Surgery* **36:** 317–320.

Majumder, N. K., Venkataramaniah, N. K., Gupta, K. R., Gapalakrishan, S. (1977) Hamartoma of nasopharynx. *Journal of Laryngology and Otology* **91:** 723–727.

Mascarello, J. T., Kraus, H. F. (1992) Second report of a translocation involving 19q13.4 in a mesenchymal hamartoma of the liver. *Cancer Genetics and Cytogenetics* **58:** 141–142.

Mushimoto, K., Kakudo, K., Ueno, S., Uenaka, H., Shirasu, R., Takasu, J. (1982) Leiomyomatous hamartoma of the gingiva: report of a case. *Japanese Journal of Oral and Maxillofacial Surgery* **28:** 493–496.

Ng, K.H., Siar, C. H., Latif, H. A. (1992) Leiomyoma of the incisive papilla region: case report. *Annals of Dentistry* **51:** 29–31.

Ritchie, A. C. (1990) Boyd's textbook of pathology. 9th Edition. Lea and Febiger, Philadelphia/London.

Semba, I., Kitano, M., Mimeva, T. (1993) Gingival leiomyomatous hamartoma: immunohistochemical and ultrastructural observations. *Journal of Oral Pathology and Medicine* **22:** 468–470.

Speleman, F., De Telder, V., De Potter, K. R., Dal Cin, P., Van Daele, S., Benoit, Y., Leroy, J. G., Van den Berghe, H. (1989) Cytogenetic analysis of a mesenchymal hamartoma of the liver. *Cancer Genetics and Cytogenetics* **40:** 29–32.

Takahashi, S., Koukita, Y., Naruke, J., Watanabe, J., Suzuki, A. (1962) Congenital epulis: a case report. *Shikagakuho* **62:** 55–59.

Address for correspondence:
Dr P. Goldsmith,
National Hospital of Neurology and Neurosurgery,
Queen Square,
London WC1N 3BG.