

Spindle-cell variant of intralingual lipoma – report of a case with literature review

SUNIL N. DUTT, M.S., D.N.B., F.R.C.S. (ED.), DAVID M. EAST, M.A., F.R.C.S., YOUSUF SALEEM, M.S., D.N.B., D.L.O., E. L. JONES, M.D., F.R.C.PATH.*

Abstract

A true lipoma is a rare lesion in the oral cavity. A histologically distinct variant is the spindle-cell variety, which is an innocuous lesion that can simulate a myxoid liposarcoma.

We report a case of intra-oral spindle-cell lipoma in a 42-year-old female and have reviewed the literature pertaining to this unusual histopathological entity. Awareness of the condition is essential for both clinicians and pathologists to avoid any misinterpretation of the benign nature of this condition.

Key words: Lipoma; Mouth; Tongue neoplasms

Introduction

Lipomas are benign neoplasms of adipocytes. Approximately 13 per cent of all lipomas occur in the head and neck region (Barnes, 1985). Oral lipomas account for about 4.4 per cent of all benign oral tumours (Wilson *et al.*, 1990). The cheek is the most common location of intraoral lipomas accounting for 32 to 50 per cent of the cases while the other sites in descending order of frequency are the tongue, floor of the mouth, buccal sulcus and vestibule, palate, lips and the gingivae (Barnes, 1985).

Spindle-cell lipoma is an uncommon and histologically distinct entity, which is extremely rare in the oral cavity. A review of the literature revealed six cases in the oral cavity and only two in the tongue (Tosios *et al.*, 1995), although there have been reports of various other types of lipomas in the tongue. It is important to differentiate this variety from an 'atypical lipoma' and from a liposarcoma, both of these entities having different growth potential and behaviour as low-grade malignancies.

Case report

A 42-year-old female presented with a gradually enlarging painless swelling in the tongue which she had first noted many years previously. The patient denied any history of trauma or inflammation of the lesion but reported some degree of difficulty in chewing food. Examination of the oral cavity revealed a well-circumscribed, 30 × 20 × 10 mm swelling in the anterior and left lateral aspect of the tongue, soft in consistency and covered by intact lingual mucosa (Figure 1). There was no evidence of inflammation or restriction of mobility of the tongue. A clinical diagnosis of a probably benign soft tissue tumour was made and an excisional biopsy of the lesion was undertaken.

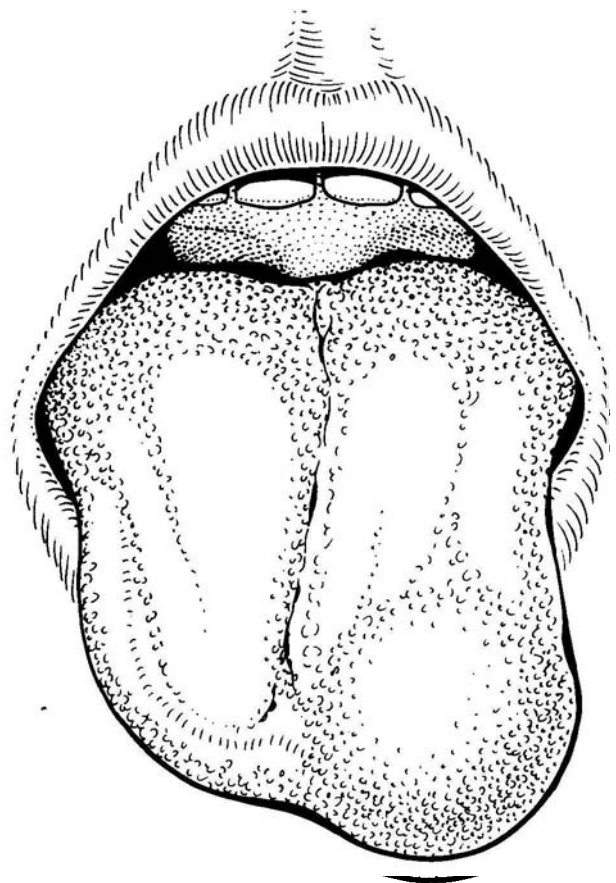


FIG. 1

Schematic diagram showing the tongue with an ovoid, well circumscribed swelling in the anterior and left lateral aspect.

From The Department of Otolaryngology, Walsall Manor Hospital, Walsall, West Midlands and the Department of Pathology*, Birmingham University Medical School, Birmingham, UK.

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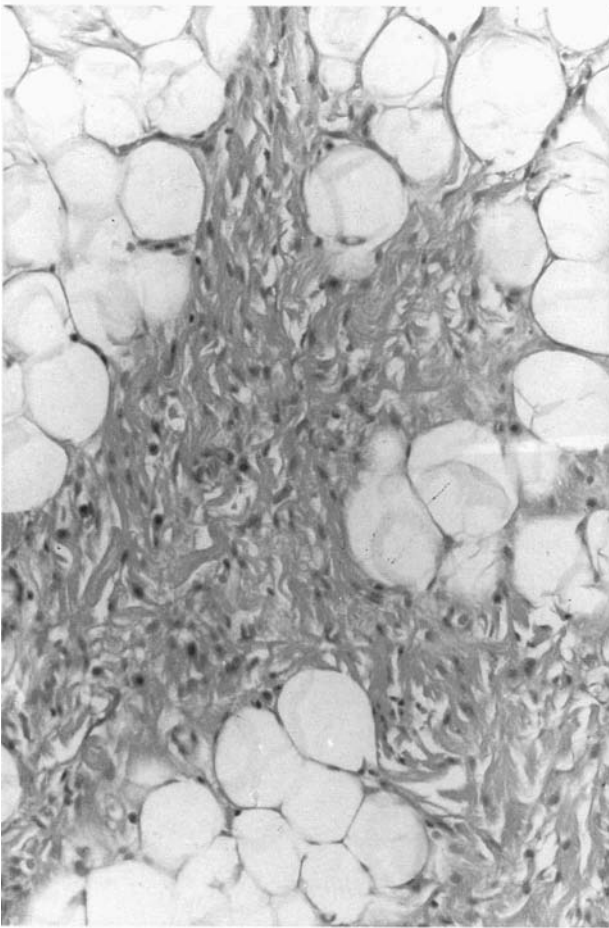


FIG. 2

Spindle-cell lipoma showing a mixture of mature fat cells and uniform spindle cells (H & E; $\times 187$)

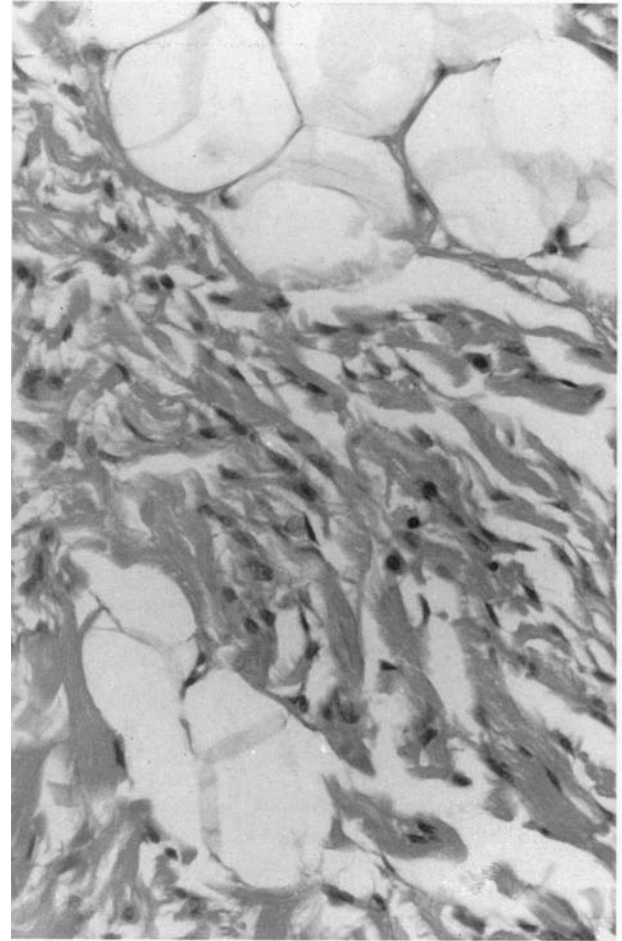


FIG. 3

Spindle-cell lipoma showing well-orientated spindle cells showing no evidence of cellular pleomorphism with interspersed collagen fibres and mature fat cells. (H & E; $\times 300$)

Histopathology

A portion of the tongue measuring $3 \times 2 \times 1$ cm was received. The cut surface showed a well-circumscribed yellow lesion, measuring 2×1 cm, extending up to the resection margins. The histological sections showed normal squamous epithelium covering a benign connective tissue tumour composed of a mixture of mature fat cells and more cellular areas of spindle cells with eosinophilic cytoplasm and varying numbers of collagen fibres (Figure 2). The spindle cells tended to be well aligned and showed no evidence of cellular pleomorphism (Figure 3). A myxoid matrix was not a particular feature of this tumour. The spindle cells were negative for S-100 protein immunostaining. The appearances were those of a spindle-cell lipoma and there was no evidence of malignancy.

Discussion

Lipomas are benign ubiquitous tumours occurring in almost any part of the body containing adipose tissue. Several histological varieties of lipoma have been described including simple lipoma, fibro-lipoma, spindle-cell lipoma, pleomorphic lipoma, myxoid lipoma, angiolipoma, angiomyolipoma, myelolipoma, lipoblastomatosis, hibernoma, atypical lipoma and infiltrating lipoma (Enzinger and Weiss, 1995).

Enzinger and Harvey described spindle-cell lipoma as a distinct entity in 1975. They described 114 cases with a range of microscopic features from mature adipose tissue with mucinous material and scattered spindle cells to a cellular mass of spindle cells with a small number of

TABLE I
CLINICAL DATA OF ALL REPORTED CASES OF INTRA-ORAL SPINDLE CELL LIPOMAS

Authors	Age (yrs)	Sex	Location of lipoma	Size (mm)	Duration	Follow-up
McDaniel <i>et al.</i> (1984)	33	F	Anterior floor of mouth	10	2 yrs	–
	52	M	Tongue	–	–	–
Christopoulos <i>et al.</i> (1989)	58	M	Hard palate	$20 \times 20 \times 10$	–	2 yrs NSR
Levy and Goding (1989)	74	F	Anterior floor of mouth	45×35	1 yr	–
Lombardi and Odell (1994)	68	F	Dorsum of tongue	$15 \times 13 \times 10$	–	–
Tosios <i>et al.</i> (1995)	52	M	Cheek	$40 \times 20 \times 12$	'yrs'	2 yrs NSR
Present case	42	F	Lateral margin of tongue	$30 \times 20 \times 10$	'yrs'	10 mths NSR

NSR – No sign of recurrence.

mature adipocytes. The majority of these tumours are of the intermediate variety with an admixture of adipocytes, spindle cells, mucinous material and bundles of collagen. There have been several additional reports of this tumour since its original description (Angervall *et al.*, 1976; Kitano *et al.*, 1979; Fletcher and Martin-Bates, 1987). Spindle-cell lipoma accounts for about 1.5 per cent of all adipose tumours. Clinically and morphologically, spindle-cell lipomas are related to pleomorphic lipomas and transitional forms can exist. The tumour can occur in any part of the body as a painless subcutaneous nodule and the described sites in the literature hitherto include the shoulders, nape of the neck, trunk, face and extremities. The age range is as wide as 24 to 81 years and 75 to 90 per cent of these occur in men (Enzinger and Harvey, 1975).

A literature review revealed only six cases of this variant occurring in the oral cavity and only two in the tongue (McDaniel *et al.*, 1984; Christopoulos *et al.*, 1989; Levy and Goding, 1989; Lombardi and Odell, 1994 and Tosios *et al.*, 1995). The clinical features and relevant clinical information are summarized in Table I.

The relative proportions of adipocytes and spindle cells in these tumours may vary markedly in different areas of the tumour. In most tumours these are present in equal proportions and the presence of numerous mast cells is a constant feature of the lesion (Angervall *et al.*, 1976; McDaniel *et al.*, 1984). Immunohistochemically, the spindle cells do not usually react with antibodies to S-100 protein and factor VIII related antigen thus excluding an origin from nerve sheath cells or endothelial cells respectively. Negative staining is also reported for the myeloid-histiocyte antigen MAC-38, collagen type IV and laminin (Beham *et al.*, 1989). It is hypothesized that spindle cells are analogous to the non-lipoblastic stellate mesenchymal cells of the primitive fat lobules which have lost their ability to differentiate to lipocytes but are capable of collagen synthesis (Bolen and Thorning, 1981; Beham *et al.*, 1989). It is obvious that these tumours have the potential to differentiate to both fat-storing and collagen-producing cells.

The differential diagnosis from a liposarcoma is based on its superficial submucosal location, well-circumscribed nature, the uniformity and the association of the spindle cells with mature and regular collagen fibres and the absence of lipoblasts or mitotic figures. Spindle cell lipoma is not usually encapsulated and may sometimes infiltrate adjacent muscle (Angervall *et al.*, 1976). Only one case of recurrence has been reported (Fletcher and Martin-Bates, 1987) and recurrent lesions should not be mistaken for liposarcomas. Definitive diagnosis of spindle cell lipoma depends on accurate correlation between the histological and the clinical features and it is important to recognize that this is a benign neoplasm and avoid the potential diagnostic hazard of interpreting the lesion as a myxoid liposarcoma with differing prognostic implications.

Acknowledgement

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References

- Angervall, L., Dahl, I., Kindblom, L. G., Save-Soderbergh, J. (1976) Spindle cell lipoma. *Acta Pathologica Microbiologica et Immunologica Scandinavica Section A* **84**: 477-487.
- Barnes, L. (1985) Tumours and tumour-like lesions of the soft tissues. In *Surgical Pathology of the Head and Neck*. (Barnes, L., ed.) Marcel Dekker, New York, pp 725-880.
- Beham, A., Schmid, C., Hodl, S., Fletcher, C. D. M. (1989) Spindle cell and pleomorphic lipoma: an immunohistochemical study and histogenetic analysis. *Journal of Pathology* **157**: 219-222.
- Bolen, J. W., Thorning, D. (1981) Spindle cell lipoma: A clinical, light- and electron-microscopical study. *American Journal of Surgical Pathology* **5**: 435-441.
- Christopoulos, P., Nicolatou, O., Patrikiou, A. (1989) Oral spindle cell lipoma: Report of a case. *International Journal of Oral and Maxillofacial Surgery* **18**: 208-209.
- Enzinger, F. M., Harvey, D. A. (1975) Spindle cell lipoma. *Cancer* **36**: 1852-1859.
- Enzinger, F. M., Weiss, S. W. (1995) Spindle cell lipoma. In *Soft Tissue Tumours*. (Enzinger, F. M., Weiss, S. W., eds.) 3rd Edition. CV Mosby, St Louis, MO. pp 395-399.
- Fletcher, C. D. M., Martin-Bates, E. (1987) Spindle cell lipoma: a clinicopathological study with some original observations. *Histopathology* **11**: 803-817.
- Kitano, M., Enjoji, M., Iwasaki, H. (1979) Spindle cell lipoma. A clinicopathological analysis of 12 cases. *Acta Pathologica Japonica* **29**: 891-899.
- Levy, F. E., Goding, G. S. (1989) Spindle cell lipoma: an unusual oral presentation. *Otolaryngology - Head and Neck Surgery* **101**: 601-603.
- Lombardi, T., Odell, E. W. (1994) Spindle cell lipoma of the oral cavity: report of a case. *Journal of Oral Pathology and Medicine* **23**: 237-239.
- McDaniel, R. K., Newland, J. R., Chiles, D. G. (1984) Intraoral spindle cell lipoma: case report with correlated light and electron microscopy. *Oral Surgery* **57**: 52-57.
- Tosios, K., Papanicolaou, S. I., Kapranos, N., Papadogeorgakis, N. (1995) Spindle cell lipoma of the oral cavity. *International Journal of Oral and Maxillofacial Surgery* **24**: 363-364.
- Wilson, G. W., Braun, T. W., Smith, R. L. (1990) Nodular mass in the anterior floor of the mouth. *Journal of Oral and Maxillofacial Surgery* **48**: 492-495.

Address for correspondence:

Mr Sunil N. Dutt, F.R.C.S.(Ed.),
37 Jacoby Place,
Priory Road,
Edgbaston,
Birmingham B5 7UN.