

Atypical intramuscular lipoma of the tongue

ASHUTOSH KACKER, M.D.*, METIN TASKIN, M.D.†

Abstract

Lipomatous tumours of the tongue are rare and most of them are benign ordinary lipomas. In this report, we describe the first case of atypical lipoma of the tongue in a 78-year-old male, and review the current literature and controversies surrounding the nomenclature and biology of this unusual lesion.

Key words: Tongue neoplasms; Lipoma

Case report

A 78-year-old black man presented with a six-year history of painless enlargement of the tongue associated with progressive dysarthria and mild dysphagia. Physical examination revealed a soft globular sessile lesion measur-

ing 6 × 4 × 4 cm protruding from the right lateral border of the tongue. The overlying mucosa was intact with a yellowish tinge and dilated veins. The base and dorsum of the tongue were unremarkable. The patient had mild dysarthria and difficulty in chewing and swallowing but

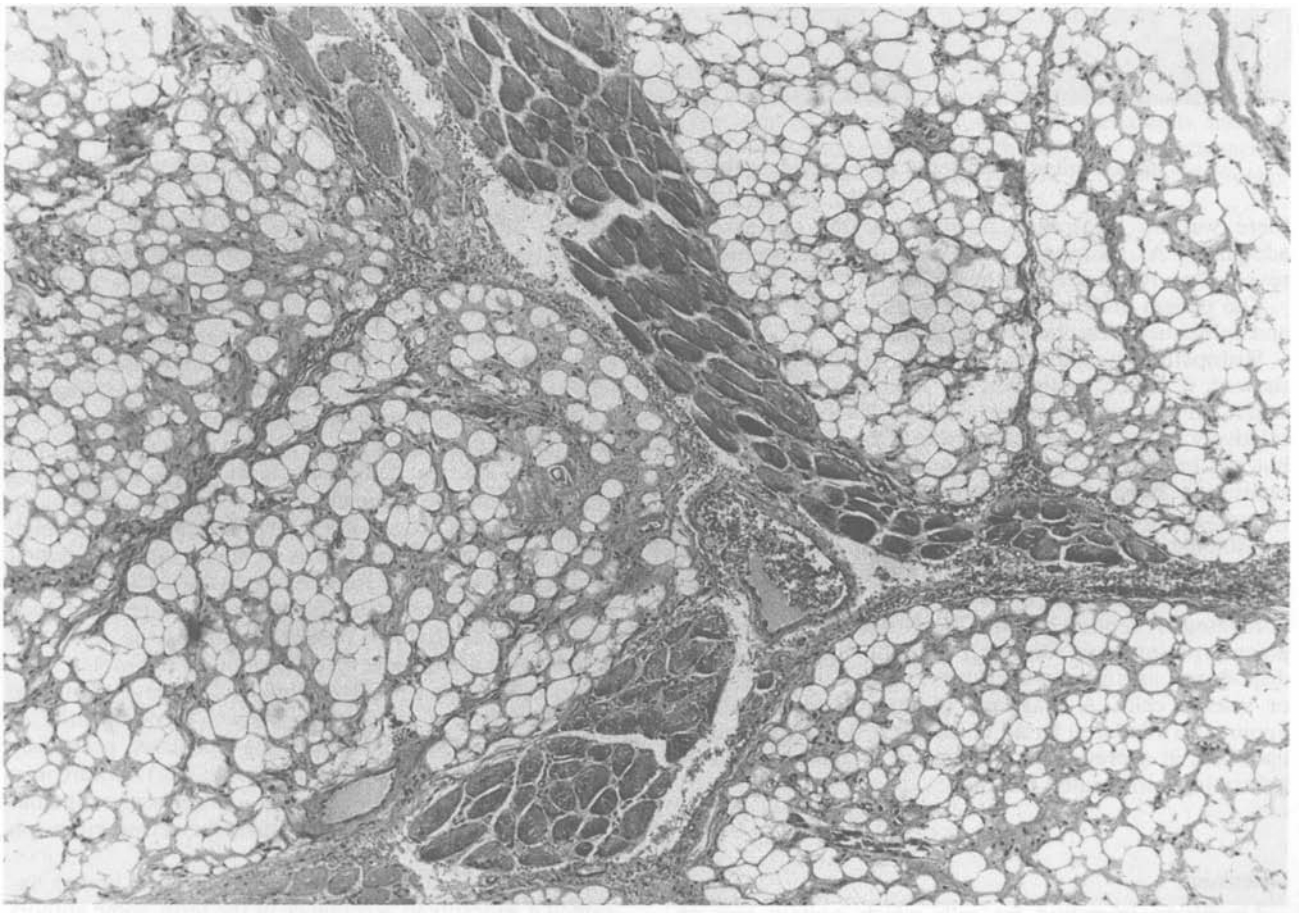


FIG. 1

Lobules of fatty tissue separated by fibrovascular septae. The tumour is broadly infiltrating the underlying muscle, widely separating bundles of partially atrophic muscle fibres (H & E; low magnification).

From the Departments of Otolaryngology* and Pathology†, Lenox Hill Hospital, New York, NY, USA.
Accepted for publication: 12 November 1995.

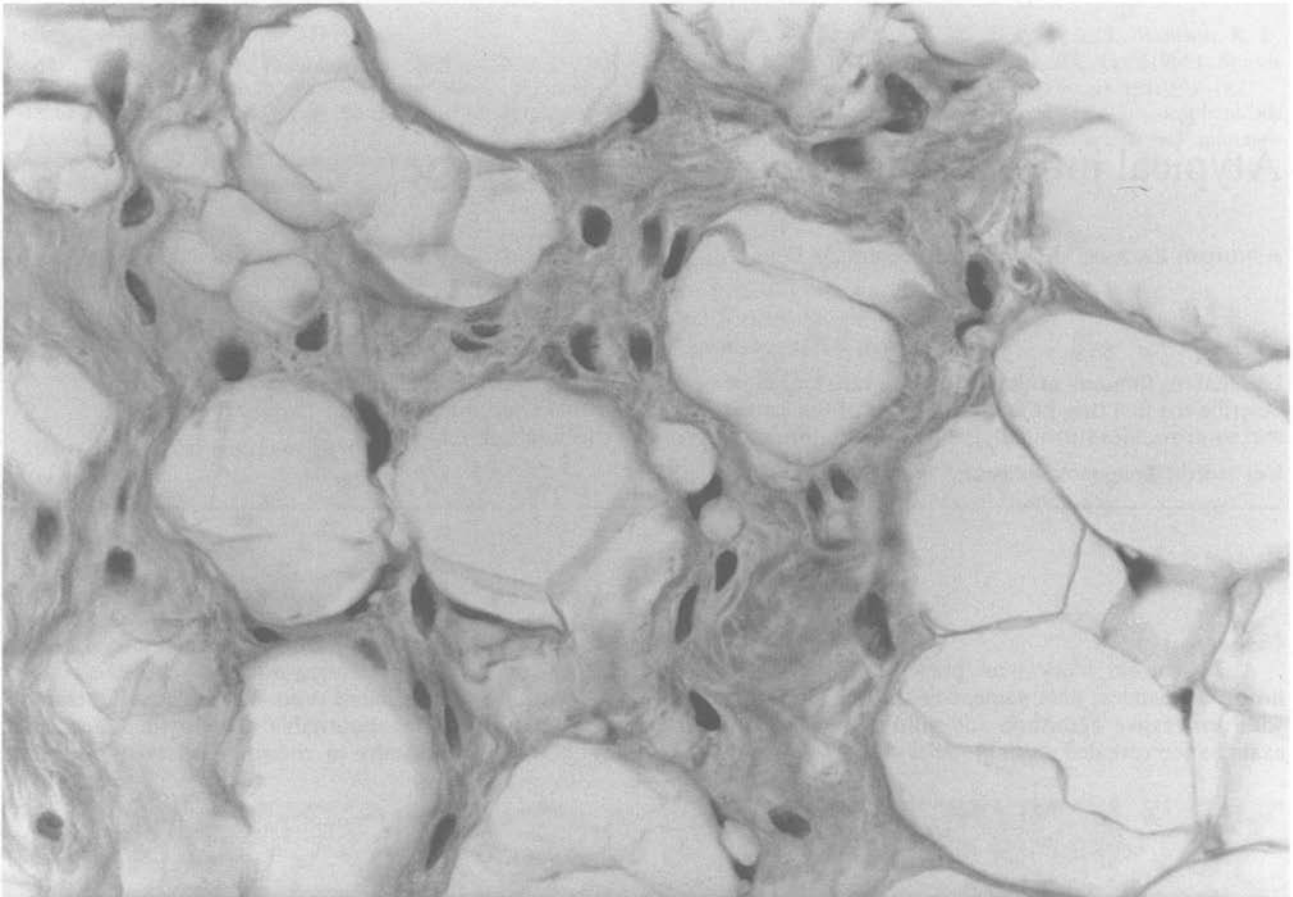


FIG. 2

A small number of scattered lipoblasts identified along the fibrovascular septae (H & E; high magnification).

there was no respiratory distress and the airway was adequate. A thorough head and neck examination revealed no lymphadenopathy.

The patient underwent an uneventful superficial biopsy and subsequent resection of the tumour.

Histopathological examination of the resected tumour showed it to be composed of lobules of fatty tissue separated by fibrovascular septae. The tumour was relatively well circumscribed towards the lingual mucosa, but was broadly infiltrating the underlying muscle, widely separating bundles of partially atrophic muscle fibres (Figure 1).

Although the neoplasm was composed almost exclusively of mature fatty tissue with low cellularity a small number of scattered lipoblasts were identified along the fibrovascular septae (Figure 2). No mitotic figures or pleomorphism were identified and the tumour did not display any significant admixture of vessels, myxoid tissue or spindle cells. A diagnosis of atypical intramuscular lipoma was rendered based on the presence of occasional lipoblasts in a predominantly mature lipomatous tumour infiltrating into striated muscle.

Discussion

Lipoblasts, immature fat cells which contain one or more fat vacuoles that indent the nucleus, are the diagnostic hallmark of liposarcomas but their presence is not restricted to malignant fatty tumours and a few scattered lipoblasts can also be encountered in atypical lipomatous tumours. The biological behaviour of lipomatous tumours depends not only on their histology but also

on their superficial or deep location. Tumours composed predominantly of mature fatty tissue can be broadly divided into lipomas and atypical lipomatous tumours, which besides mature fat cells also contain various amounts of lipoblasts, as in the atypical lipoma, spindle cells, as in the spindle-cell lipoma or multinucleated floret-like giant cells as in the pleomorphic lipoma. When in superficial (subcutaneous) location these atypical lipomatous tumours behave in a benign fashion, with low recurrence rates and no metastases, while histologically identical tumours, occurring deep in the retroperitoneum or inguinal area are capable of dedifferentiation into high grade sarcomas and metastases (Evans *et al.*, 1979; Azumi *et al.*, 1987; Weiss and Rao, 1992). Intramuscular lipomas and atypical intramuscular lipomas have a high recurrence rate (Azumi *et al.*, 1987) possibly related to the difficulties in achieving a complete resection but do not metastasize. These differences in biological behaviour have led to the reclassification of tumours composed predominantly of mature fatty tissue but containing lipoblasts, previously classified as well-differentiated (lipoma-like) liposarcoma, as atypical lipomas and atypical intramuscular lipomas when they occur in sites other than the retroperitoneum (Evans *et al.*, 1979).

Although the diagnostic category of atypical lipoma has gained a widespread acceptance of the term, some authors favour the use of terms such as borderline adipose tissue neoplasm (Hajdu and D'Ambrosio, 1993) to designate these tumours to stress their high recurrence rate. The differential diagnosis of atypical lipomas is made by careful exclusion of lipomas and their variants – angiolipoma and fibrolipoma, which do not contain lipoblasts, and of

dedifferentiated liposarcomas which contain juxtaposed areas of high grade sarcoma. Other lipoblast-containing lesions such as high grade liposarcomas (pleomorphic or round-cell) are excluded by the predominance of mature fatty tissue and lack of anaplasia; myxoid liposarcoma by the absence of myxoid change and lipoblastoma and lipoblastomatosis by the exclusive occurrence of these benign conditions in infancy and early childhood.

Although lipomas are among the most common benign tumours their occurrence in the oral cavity has been reported to be infrequent, representing between 2.2 per cent and 4.4 per cent of benign tumours of the oral cavity (Dockerty *et al.*, 1968; Hatziotis, 1971). Lipomas of the tongue account for approximately 20 per cent of intraoral lipomas (Hatziotis, 1971) and are usually small (less than 1 cm in diameter), submucosal, sessile or pedunculated tumours that can cause local discomfort and, when unusually large, interfere with mastication, deglutition and respiration. Most lesions occur singly but cases of multiple lipomas have been reported (Hatziotis, 1971) and must be differentiated from the rare lipomatosis of the tongue (Katou *et al.*, 1993). Although reported to occur at any age, most reported cases have occurred in middle-aged and elderly adults, some studies suggesting a slight male predominance (Hatziotis, 1971).

Approximately 80 per cent of these lipomas are of the 'ordinary' type, composed exclusively of mature fatty tissue (Gouillou *et al.*, 1986). Most of the remaining 20 per cent show a significant fibrous or vascular component and are classified as fibrolipomas and angiolipomas, respectively. All other histological variants of lipoma are exceptionally rare in the oral cavity and tongue as are liposarcomas. To date, only single cases of infiltrating (intramuscular) lipoma (Gouillou *et al.*, 1986), pleomorphic lipoma (Garavaglia and Gnepp, 1987) and liposarcoma (Guest, 1992) have been reported to arise in the tongue. This is, to our knowledge, the first case of atypical lipoma of the tongue to be reported. Like other lipomatous tumours, the treatment of this tumour is exclusively surgical and the clinical behaviour is best predicted by the completeness of surgical resection, as attested by surgical margins histologically free of tumour.

Acknowledgements

The authors wish to thank Rajesh Kakani, M.D. and Stefan E. Pambuccian, M.D. for their contribution and reviewing the manuscript.

References

- Azumi, N., Curtis, J., Kempson, R. L., Hendrickson, M. R. (1987) Atypical and malignant neoplasms showing lipomatous differentiation. A study of 111 cases. *American Journal of Surgical Pathology* **11**(3): 161–183.
- Dockerty, M. B., Parkhill, E. M., Dahlin, D. C., Woolner, L. B., Soule, E. H., Harrison, E. G. (1968) Tumors of the oral cavity and pharynx. In *Atlas of Tumor Pathology*. Fascicle 10b, Armed Forces Institute of Pathology, Washington, D.C., p 83.
- Evans, H. L., Soule, E. H., Winkelmann, R. K. (1979) Atypical lipoma, atypical intramuscular lipoma and well-differentiated retroperitoneal liposarcoma. A reappraisal of 30 cases formerly classified as well-differentiated liposarcoma. *Cancer* **43**(2): 574–584.
- Garavaglia, J., Gnepp, D. R. (1987) Intramuscular (infiltrating) lipoma of the tongue. *Oral Surgery, Oral Medicine, Oral Pathology* **63**(3): 348–350.
- Guest, P. G. (1992) Liposarcoma of the tongue. *British Journal of Oral and Maxillofacial Surgery* **30**(4): 268–269.
- Gouillou, L., Dehon, A., Charlin, B., Madarnas, P. (1986) Pleomorphic lipoma of the tongue. Case report and literature review. *Journal of Otolaryngology* **15**(5): 313–316.
- Hajdu, S. I., D'Ambrosio, F. G. (1993) Histopathologic classification of limb sarcomas in relation to prognosis. *Surgical Oncology Clinics of North America* **2**(4): 509–535.
- Hatziotis, J. C. (1971) Lipoma of the oral cavity. *Oral Surgery, Oral Medicine, Oral Pathology* **31**(4): 511–524.
- Katou, F., Shirai, N., Motegi, K., Satoh, R., Satoh, S. (1993) Symmetrical lipomatosis of the tongue presenting as macroglossia. Report of two cases. *Journal of Cranio-Maxillo-Facial Surgery* **21**(7): 298–301.
- Weiss, S. W., Rao, V. K. (1992) Well-differentiated liposarcoma (atypical lipoma) of deep soft tissues of the extremities, retroperitoneum and miscellaneous sites. A follow-up study of 92 cases with analysis of the incidence of dedifferentiation. *American Journal of Surgical Pathology* **16**(11): 1051–1058.

Address for correspondence:
Ashutosh Kacker,
Department of Otolaryngology,
Lenox Hill Hospital,
New York,
NY 10021, USA.

Fax: 212-434-2382