

CASE REPORT

Intramuscular lipomatosis of tongue

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Postgrad Med J 2002;**78**:295–297

A rare case of intramuscular lipomatosis of the tongue with intramuscular invasion in a 42 year old man is presented. The literature is reviewed and the clinical features, pathology, and treatment are discussed briefly.

The majority of tumours of the tongue are malignant.¹ Of the benign tumours lipomas constitute a very small proportion, with very few cases reported. A case of large multiple lipomatosis of the tongue with intramuscular invasion is presented, and the current literature is reviewed and treatment is discussed.

CASE REPORT

A 42 year old man presented with gradually progressive, diffuse painless swellings on both lateral borders of the tongue. Swelling on the right lateral border measured 4 × 4 cm and that on the left measured 3 × 2.5 cm. The swellings had a yellowish colour with a smooth shining surface and a few dilated vessels over it. There was no dysphagia, ankyloglossia, or stridor. However his speech was not clear because of the bulk of the tongue. There was no history of any relevant medical illness, drug intake, or similar illness in the family. The swellings were soft to firm in consistency, non-tender, and became harder in consistency on protrusion of the tongue. There were no significant lymph nodes in the neck and no other swellings anywhere in the body.

Fine needle aspiration was inconclusive. The swellings were excised and submitted to frozen section examination and reported as suggestive of lipoma. Subsequently the histopathological report confirmed intramuscular lipoma. The patient had an uneventful postoperative period and is disease-free after one and a half years of follow up. His speech and appearance of the tongue are now normal.



Figure 1 Cut surface of the excised tumour showing multiple yellowish lobules of varying sizes.

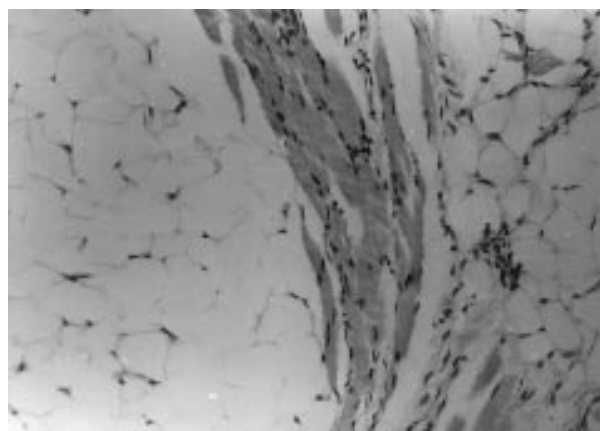


Figure 2 Intramuscular lipoma showing sheets of mature fat cells infiltrating striated muscle tissues (haematoxylin and eosin stain × 250).

PATHOLOGICAL FINDINGS

The excised portion of tongue measured 5.5 × 3 × 2 cm. The surface appeared vaguely nodular with an intact mucosa. The cut surface revealed multiple yellowish lobules of varying sizes, the largest of the lobules measuring about 3 × 2 cm in size (fig 1).

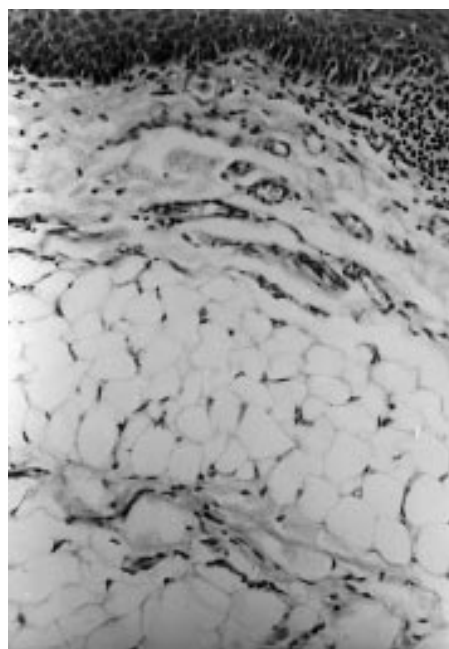


Figure 3 Intramuscular lipoma showing sheets of mature adipocytes underlying an intact squamous epithelium (haematoxylin and eosin stain × 250).

Box 1: Differential diagnosis of macroglossia*Congenital*

- Down's syndrome.
- Cretinism.
- Beckwith-Wiedemann syndrome or exomphalos.
- Macroglossia or gigantism syndrome.

Inflammatory

- Lymphoedema.

Trauma

- Lingual haematoma.

Neoplasm

- Benign (for example, neurofibroma, haemangioma, lymphangioma).
- Malignant (for example, squamous cell carcinoma).

Metabolic

- Glycogen storage disorders.
- Amyloidosis.

Endocrine

- Acromegaly.
- Myxoedema.

Miscellaneous

- Angioneurotic oedema.

Box 2: Benign tumours of the tongue

- Papilloma.
- Fibroepithelial polyp.
- Pregnancy tumour.
- Haemangioma.
- Lymphangioma.
- Plexiform neuroma.
- Neurofibroma.
- Lipoma.
- Osteoma.
- Chondroma.
- Sublingual varicosities.
- Granular cell myoblastoma.
- Juvenile fibroma.

Histology showed lobules and sheets of mature adipocytes infiltrating between the muscle fibres (fig 2). The nuclei of the fat cells appeared bland. The muscle fibres in areas showed varying degrees of atrophy. The overlying squamous epithelium appeared intact and normal (fig 3).

DISCUSSION

The differential diagnosis of macroglossia is given in box 1. Possible effects of macroglossia include cosmetic deformity, dysarthria, dysphagia, stridor, malocclusion, and dentofacial anomaly. Benign tumours of the tongue are listed in box 2. They are uncommon compared with the malignant tumours.¹ Benign tumours that commonly account for macroglossia are neurofibromatosis, cavernous haemangioma, and lymphangioma.

Lipoma of the tongue is rare and often presents as a single, superficial, pedunculated, or sessile lesion. Presentation as multiple tumours, infiltrating tumours, or macroglossia are infrequent. Diagnosis of lipoma of the tongue is essentially clinical. However radiological investigations including computed tomography and magnetic resonance imaging (MRI) may be helpful, with fat suppressed MRI images being very useful in establishing a provisional diagnosis.² Histologically the majority of lipomas are benign and ordinary.³ Important histological variants of lipomas are spindle cell lipoma,⁴ pleomorphic lipoma,⁵ angioliipoma,⁶ fibrolipoma,⁷ and myxoid lipoma.⁸ An important differential diagnosis of lipoma is well

Learning points

- Majority of tumours of the tongue are malignant.
- Tongue is a rare site for lipoma.
- Histologically most of the tongue lipomas are benign ordinary ones, although several histological variants of lipoma have been reported from other sites.
- Intramuscular lipomas of the tongue are very rare and tend to recur if not excised sufficiently wide.
- A well differentiated liposarcoma can mimic a benign lipoma histologically.

differentiated lipoma-like liposarcoma. The possibility of associated malignancy (liposarcoma) can only be ruled out after studying several cross sections of the lesion, although certain pathological characteristics, including the presence of lipoblasts and intramuscular invasion of the lesion, may point towards malignancy.⁹

Extensive review of the literature revealed very few previous reports of lipomas of the tongue.^{3,8,10} The present case is unique because of its combined multiplicity of origin¹¹ and its infiltrating nature.^{3,10} We also believe that it is the bulkiest lipomatous lesion ever reported within the tongue¹²; the anatomical and functional significance being macroglossia and dysarthria.

Intramuscular extension in lipoma is clinically diagnosed by the change in consistency of the swelling on the contraction of the muscle involved.¹³ They account for 1.8% of fatty tumours and arise predominantly in middle to late adult life.¹⁴ They are divided into well circumscribed and the infiltrative types. Infiltrative type tends to recur¹⁴ and often resemble a well differentiated liposarcoma histopathologically. Hence an anticipatory wide excision of the tumour may have to be undertaken. Further follow up will depend on the histopathological report. If the report is that of a well differentiated lipoma-like liposarcoma with a margin of clearance less than 1 cm, further surgery is mandatory. After an adequate tumour clearance of more than 1.5 cm is achieved, follow up twice a month for one year, every six months for the next two years, and yearly for the next seven years is recommended because of the high probability of distant metastasis. The role of adjuvant radiotherapy or chemotherapy is very limited.

CONCLUSION

Lipomatosis of the tongue is a rare phenomenon that can account for macroglossia. Occurrence of lipomas and many of its variants have been reported sporadically in the tongue and other sites within the oral cavity. Meticulous wide excision is the key to management of large lipomas of the oral cavity with intramuscular infiltration, because of the distinct possibility of a well differentiated lipoma-like liposarcoma.

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Submitted 9 May 2001

Accepted 10 December 2001

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