The Journal of Laryngology & Otology
December 2000, Vol. **114,** pp. 978–979

Proliferative myositis arising in the tongue

A. Singh, B.Sc., M.B.B.S., J. M. Philpott, F.R.C.S., N. N. Patel, F.R.C.S., G. Mochloulis, M.D., C.C.S.T. (orl)

Abstract
We describe a rare case of proliferative myositis affecting the lateral border of the tongue. The site of the lesion and its gross pathological presentation were highly suggestive of a malignant process. Subsequent biopsy and characteristic histological appearance led to the diagnosis of this benign condition. This is the first reported case of a painful presentation of proliferative myositis involving the tongue. This report serves to remind the head and neck surgeon of the need to obtain histological diagnosis of malignancy before embarking upon radical surgical treatment. We recommend careful follow-up to ensure complete resolution of the lesion.

Key words: Myositis; Tongue

Introduction

Proliferative myositis is an uncommon benign reactive process affecting muscle and often associated with proliferative fasciitis.[1,2] The condition usually involves the flat muscles of the trunk and shoulder but distinct cases involving the head and neck have been described in the literature.[3] It usually presents as a rapidly growing nodular lesion with characteristic pathological appearance. Local excision is usually curative. We report a case of proliferative myositis involving the lateral tongue border with an atypical presentation leading to an initial suspicion of malignancy.

Case report

A 46-year-old lady previously well was admitted to hospital with an acute psychotic episode and a lobar pneumonia having been found in the streets eating mud. Her past psychiatric history included deliberate self-harm, alcohol and drug dependency. She was a heavy smoker of 40 cigarettes a day. On presentation, she complained of tongue pain and was initially diagnosed with a bacterial mucositis of the oral cavity involving the tongue. Her symptoms failed to settle on antibiotic therapy and her tongue pain increased interfering with her speech and swallowing.

She was referred for an urgent otorhinolaryngology opinion. Examination revealed a tender, large ulcerated lesion located on the left lateral border of the tongue (Figure 1). There was a similar but smaller lesion on the right side. Examination of the neck revealed bilateral cervical lymphadenopathy. The history of smoking and alcohol abuse coupled with the clinical appearance led to the provisional diagnosis of squamous cell carcinoma of the tongue. An examination under general anaesthetic confirmed the presence of an ulcerating lesion of the tongue. Biopsies were taken and sent for histological and microbiological analysis. Rigid endoscopic examination of the upper aerodigestive tract was otherwise normal.

Light microscopy demonstrated surface ulceration with granulation tissue underlying chronic inflammation with degeneration of striated muscle cells. Many cells demonstrated a ganglion-like appearance which is characteristic of this condition (Figure 2). No evidence of dysplasia or malignancy was seen. Microbiology staining and culture for bacteria and fungi was negative.

Following treatment for the acute psychotic state the patient was advised on regular oral hygiene and the importance of avoiding further trauma. Strong analgesics were prescribed to control pain. The patient was followed up closely in the out-patients department to ensure resolution of the lesion. At her last visit, three months post-presentation, she was asymptomatic and her tongue lesion had resolved completely.

Fig. 1
Proliferative myositis arising in the left lateral border of the tongue. The arrow points to the raised and ulcerated appearance of the lesion.

From the Department of Otorhinolaryngology–Head and Neck Surgery, St Mary's Hospital, Praed Street, London, UK.
Accepted for publication: 10 July 2000.

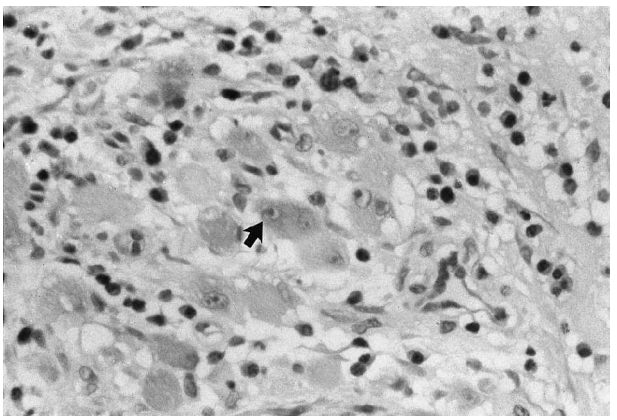

FIG. 2
Ganglion-like cells typical of proliferative myositis (arrow) (H&E; ×400).

Discussion

Proliferative myositis is an uncommon benign condition usually affecting the flat muscles of the trunk and shoulder. It is an intramuscular reactive proliferative process seen in conjunction with a proliferative fasciitis.[1,2] It was first described by Kern in 1960 as a pseudosarcomatous process.[4]

Typically, proliferative myositis occurs as a rapidly growing nodular lesion, which is not painful nor tender. It is generally seen in the over-45 age group. Grossly, lesions are poorly demarcated with scar-like induration. Microscopically, spindle cells with numerous ganglion-like or rhabdomyoblast-like cells and proliferating fibroblasts with circumferential vessel fibrosis are present. Occasional osteoid formation is seen. Notably, muscle bundles show atrophy with no attempt at regeneration. The majority of cells are vimentin and actin positive.[1]

Differential diagnosis includes rhabdomyosarcoma, nodular fasciitis, fibromatosis and focal myositis. In the tongue, myositis has been confused with squamous cell carcinoma, clinically and histologically.[5,6] One report describes an initial diagnosis of proliferative myositis based on biopsy findings but was subsequently revised to desmoplastic squamous cell carcinoma following electron microscopy studies.[6]

The clinical features seen in this case initially suggested a carcinoma of the tongue, however, the correct diagnosis was made by the typical histological findings on biopsy. Therefore, a diagnosis of proliferative myositis affecting the tongue should be made with care based on characteristic histological appearance and with follow up to ensure resolution of the lesion.

Although originally documented in the extremities, awareness has led to the diagnosis of proliferative myositis in other areas including the head and neck.[3,7] A review by Dent *et al*.[3] showed 10 per cent of cases arose in the head and neck region. The majority involved the sternocleidomastoid and masseter muscles.

Only one case has been described involving the tongue.[3] This was an incidental finding involving the dorsum of the tongue in the absence of trauma and treated with an excision biopsy. The current case contrasts with the previous report in several ways emphasizing the variability in clinical appearance of proliferative myositis in the tongue.

The painful ulcerated presentation described in this report is atypical for proliferative myositis, which is usually seen as a painless nodular lesion. Trauma caused by chewing mud whilst the patient was in her acute psychotic state was felt to instigate the disease process and may have contributed a secondary inflammatory component. In the series by Dent *et al*.[3] trauma has been suggested as an aetiological factor in 30 per cent of the cases.

The reported head and neck cases of proliferative myositis were all treated by complete or partial excision with no evidence of recurrence. The current case resolved completely with attention to oral hygiene along with adequate control of the patient's acute psychotic state. At follow-up there has been no evidence of recurrence of disease.

Conclusion

Proliferative myositis is an uncommon condition, rarely involving the head and neck region.

The clinical presentation described in this report demonstrate atypical features of proliferative myositis that raised the possibility of a malignant disease process.

Accurate diagnosis relies on characteristic histopathological findings and complete resolution of the lesion.

We hope that this report serves to highlight the care needed in diagnosing proliferative myositis. If correctly identified this is a condition easily treated. The authors suggest careful follow up to ensure complete resolution of the lesion, otherwise repeat biopsy is recommended.

References

1 Al-Nafussi A, Hughes DE. *Histological Diagnosis of Tumours by Pattern Analysis An A-Z guide*. 1st edn. London: Arnold, 1997
2 Enzinger FM, Weiss SW. *Soft Tissue Tumours*. 1st edn. St Louis: CV Mosby, 1983
3 Dent CD, DeBoom GW, Hamlin ML. Proliferative myositis of the head and neck. Report of a case and review of the literature. *Oral Surg Oral Med Oral Pathol* 1994;**78**:354–8
4 Kern WH. Proliferative myositis: a pseudosarcomatous reaction to injury. *Arch Pathol Lab Med* 1960;**69**:209–16
5 McLendon CL, Levine PA, Mills SE, Black WC. Squamous cell carcinoma masquerading as focal myositis of the tongue. *Head Neck* 1989;**11**:353–7
6 Norris CM, Mustoe TA, Ross JS, Goodman ML. Desmoplastic squamous cell carcinoma of the tongue simulating myositis or fasciitis. *Head Neck Surg* 1986;**9**:51–5
7 Donegan JO, Miller TH, Wirman JA. Proliferative myositis in the head and neck.*Am J Otolaryngol* 1983;**4**:442–4

Address for correspondence:
A. Singh,
Department of Otorhinolaryngology–Head and Neck Surgery,
St Mary's Hospital,
Praed Street,
London W1 2NY, UK.

Mr A Singh takes responsibility for the integrity of the content of the paper.
Competing interests: None declared