INTRAORAL PROLIFERATIVE MYOSITIS: CASE REPORT AND LITERATURE REVIEW

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Abstract: Background. Proliferative myositis is a rare, benign, reactive intramuscular lesion of fibroblastic/myofibroblastic origin: an identical lesion in a subcutaneous or fascial location is referred to as proliferative fasciitis. The rapid growth rate and unusual histopathologic features have frequently been mistaken for a malignant process and have promoted unnecessary invasive procedures. Here we present only the third oral case of proliferative myositis, arising from the tongue of a 65-year-old man.

Methods and Results. Histologically, the resected lesion was composed of numerous fibroblastic or myofibroblastic spindle cells and variable numbers of large ganglion-like cells infiltrating between and around muscle fascicles, resembling a “checkerboard” configuration. A demographic profile of proliferative myositis of the head and neck is also provided, compiled from 19 patients culled from an English-language literature review and this report.

Conclusions. Incisional biopsy or fine-needle aspiration biopsy of proliferative myositis of the head and neck should lead to spontaneous resolution and is, therefore, sufficient to render the diagnosis and to provide conservative treatment. Recurrence is extremely rare. ©2006 Wiley Periodicals, Inc. Head Neck 29: 416–420, 2007

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Fibroblastic and myofibroblastic tumors comprise a group of tumefactions exhibiting biologic diversity ranging from reactive, pseudosarcomatous proliferations to intermediate lesions, such as the fibromatoses, to malignant neoplasms, including fibrosarcoma.1–3 Nodular fasciitis, proliferative fasciitis, and proliferative myositis represent related reparative reactions with many histologic similarities.2,4 The hallmark feature of these lesions is the alarming growth rate, often masquerading as a malignant process, such as rhabdomyosarcoma or other sarcomas. Although nodular fasciitis is considered relatively common, proliferative fasciitis and proliferative myositis are rare lesions that are identical in appearance with 1 exception: the former is a fascial or subcutaneous process, while the latter is intramuscular.

To date, only 2 patients with proliferative myositis have been reported within the oral cavity, both involving the tongue.4,5 Here we present a third patient with proliferative myositis of the tongue, including microscopic and immunohistochemical findings.
CASE REPORT

A 65-year-old man was seen with a slightly tender growth of the tongue of 2 weeks duration. The patient denied any episodes of trauma to the region. The medical history was only significant for osteoarthritis and allergic rhinitis, for which the patient took celecoxib and fluticasone, respectively, and an allergy to penicillin. Clinical examination revealed a 5-×5-mm firm, minimally raised, round lesion of normal color along the right dorsum of the tongue. An excisional biopsy was performed with approximately 2 mm of margin and submitted for pathologic review. The patient maintained an uncomplicated postoperative course and has manifested no clinical signs of recurrence at a 5-month follow-up examination.

On gross examination, the surgical specimen consisted of a 1.0-×0.5-×0.3-cm tan-colored soft tissue mass. Histopathologic examination revealed an unencapsulated, deep, soft tissue lesion occurring in and among the skeletal muscle fibers. A focal area of surface ulceration, covered by a fibropurulent membrane, was noticed with attendant granulation tissue containing acute and chronic inflammatory cells. Below the ulceration, the lesion was composed of a population of 2 cell types, consisting of fibroblastic/myofibroblastic spindle cells and large ganglion-like cells with abundant basophilic cytoplasm, and large nuclei containing 1 to 2 prominent nucleoli. Focal aggregates of scant inflammatory cells were noted within the lesion. The lesional cells were set in a loose connective tissue background and expanded the space between normal and focally degenerating skeletal muscle cell fibers, producing a localized “checkerboard” pattern (Figure 1). Osseous or cartilaginous foci and necrotic areas were absent.

Immunohistochemical studies were performed to determine the cellular origin of this lesion using SMA (smooth muscle actin), vimentin, CD68, S-100 protein, pan-cytokeratin, and desmin. Immunostaining was graded according to the stain intensity as weak, moderate, or strong and according to the degree of positivity as follows: − (0% to 5% positive cells), + (5% to 25% positive cells), ++ (25% to 50% positive cells), and +++ (50% to 100% positive cells), following previously described protocols.6 The spindle cell populations demonstrated strong diffuse (+++) staining for vimentin, moderate focal (+) immunostaining for SMA, and strong focal (+) staining for CD68. In contrast, the ganglion-like cells were negative for SMA and CD68 and only showed diffuse strong positivity for vimentin. S-100 protein, desmin, and pan-cytokeratin expression was negative in the tumor cells, highlighting only nerve fibers, striated muscle fibers, and the overlying epithelium, respectively (Figure 2).

DISCUSSION

At least 100 cases of proliferative myositis have appeared in the English-language literature, pre-
dominately affecting the musculature of the trunk and extremities. Lesions of the head and neck are rarely encountered, and with the inclusion of the present report, total only 20 documented cases.\textsuperscript{4,5,7–19} Despite the limited number of patients with proliferative myositis of the head and neck, demographic trends can be established.

Patient age ranged from 9 to 82 years, with a mean of 56 years, with no appreciable gender predisposition (11 male, 9 female). The most commonly affected site was the sternocleidomastoid muscle (40\%, 8/20), with fewer cases arising in the masseter (15\%, 3/20), neck—not otherwise specified (10\%, 2/20), and singular lesions (5\%) involving the mylohyoid, trapezius within the posterior triangle of the neck, or the buccinator muscle. Intraoral lesions are extremely rare, and with the addition of our report, amount to only 3 cases, all involving the tongue.\textsuperscript{4,5} Ostensibly, the tongue has been the exclusive oral site affected with proliferative myositis, owing to the abundance of skeletal muscle within this structure.

Sixty-nine percent of patients (9/13) were seen with a painful or tender, well-defined mass; 2 of them manifested lesional ulceration, including the present patient.\textsuperscript{5,7,9,11–15} Tumorous consistency was described as firm or hard in 50\% (7/14) of patients,\textsuperscript{4,10,13–15,19} including the present patient, with 14\% (2/14) reported as fixed to the underlying structure,\textsuperscript{10,18} and 14\% (2/14) were noted to be mobile.\textsuperscript{14,19} Rapid enlargement of 2 months duration or less was reported in 77\% (10/13) of patients, including the present patient.\textsuperscript{7–11,13,15,18} One patient with proliferative myositis arising from the masseter muscle complained of trismus and chewing difficulty,\textsuperscript{11} while another patient with a painful glossal mass experienced interference with speech and swallowing.\textsuperscript{5}

The etiopathogenesis of proliferative myositis has not been thoroughly elucidated, although there is general agreement in the literature that proliferative myositis represents a reactive lesion. Recent trauma to variously affected bodily lesions was specifically designated with 42\% (10/24) of patients.\textsuperscript{8} Similarly, antecedent injury had been acknowledged in 27\% (4/15) of cohorts with head and neck lesions.\textsuperscript{5,8,15} The implication that trauma may play any constitutive role in oral development is debatable, considering the paucity of cases of glossal proliferative myositis and the overall frequency of accidental biting of the tongue in the population at large.

Histopathologic assessment of proliferative myositis demonstrates infiltration of abundant plump
fibroblastic and myofibroblastic spindle cell populations and large ganglion-like cells between and around relatively intact skeletal muscle fascicles, resulting in a “checkerboard” pattern. The ganglion-like cells vary in number and may be evenly or randomly distributed, displaying circular, singular nuclei with conspicuous nucleoli arrayed in a spacious basophilic cytoplasm; some of these giant cells may be multi-nucleated. Both the spindle cell and ganglion-like cells may reveal increased mitotic figures without atypia. The stromal appearance ranges from myxoid to collagenous with occasional lymphocytes, focal areas of necrosis, secondary muscle atrophy, or metaplastic bone or cartilage formation. The periphery of the lesion is usually poorly circumscribed and unencapsulated. Childhood lesions possess greater cellularity, reduced myxoid and collagenous background with less infiltrative growth and better delineated borders; in contrast, lesions of adult onset manifest a dominance of ganglion-like cells in the expense of the slender fibroblasts, with frequent focal necrosis and higher mitotic activity. Immunohistochemically, lesions invariably stain positive for SMA, muscle specific actin, and vimentin, with generally focal CD68 (KP1) expression; the ganglion-like cells may be negative for actin. Desmin, myoglobinulin, and factor XIIIa positivity is seen only in isolated cases, whereas protein staining for S-100 protein, cytokeratin, and factor VIII-related antigen has not been observed. The histogenesis of the ganglion-like cells is controversial, with investigators suggesting a fibroblastic, histiocytic, myofibroblastic, or an osteoblastic origin.

The differential diagnosis of proliferative myositis should include various benign and malignant processes such as nodular fasciitis, proliferative fasciitis, focal myositis, myositis ossificans, desmoid fibromatosis, ganglioneuroma, inflammatory myofibroblastic tumor, rhabdomyoma, intramuscular hemangioma, rhabdomyosarcoma, and desmoplastic squamous cell carcinoma. Earlier cases of proliferative myositis were frequently misdiagnosed as a malignant process, attributed to the unusual cellularity and exuberant behavior, and treated excessively, often with radical excision, and occasionally in conjunction with lymphadenectomy, chemotherapy, or radiation therapy. Subsequent series and numerous case reports have established the lesion’s self-limiting and self-healing tendency and lack of recurrent or metastatic potential, leading a growing number of clinicians to advocate only conservative histologic evaluation. In fact, a recent review of the literature revealed 45% (36/80) of cases of proliferative myositis were successfully managed with either incisional biopsy or fine-needle aspiration biopsy (FNAB), resulting in spontaneous resolution. Thus, incisional biopsy or FNAB may serve as both the diagnostic procedure and a conservative, tissue-sparing modality of treatment.

With regard to modalities of treatment for proliferative myositis of the head and neck, 70% (14/20) of patients had undergone excisional biopsy, including the present one, while 15% (3/20) each were managed with incisional biopsy and FNAB. The decision to perform an excisional biopsy with the present patient was based on the diminutive size of the mass and the lack of concern for contiguous vital structures. Postsurgical evaluation was reported in 80% (16/20) of head and neck cases, ranging from 4 weeks to 7 years, with an average of 17 months, and none of these lesions had recurred. Nevertheless, rare aggressive behavior of proliferative myositis of the head and neck with extension into surrounding structures has been reported, observed in 15% (3/20) of the reported cases, 2 of which showed vascular compromise. Overall, patients maintain an excellent prognosis, and recurrence of proliferative myositis is extremely rare, irrespective of the anatomic site affected.

REFERENCES

Intraoral Proliferative Myositis


