Proliferative Myositis: An Uncommon Cause of Recurrent Trismus and Facial Swelling

Kevin J. Quinn, MD[1]; Lexie L. Wang, MD[1]; Evan R. Reiter, MD[1]
[1]Department of Otolaryngology – Head & Neck Surgery
Virginia Commonwealth University School of Medicine, Richmond, Virginia, USA

Case Presentation

History and Examination:

- 56-year-old otherwise healthy female with no history of tobacco or alcohol use presented with recurrent, acute onset left facial swelling and trismus without paresthesia, weakness, headache, or vision changes.
- No evidence of odontogenic pathology on panoramic radiograph evaluation with oral-maxillofacial surgery, and no purulence on attempted intraoral incision and drainage.
- Treatment with oral dicloxacillin, amoxicillin-clavulanate, clindamycin, and prolonged ciprofloxacin did not result in lasting improvement.
- Physical exam notable for trismus to 1 cm at the incisors, no visible/palpable oral or oropharyngeal abnormalities, tonsils 1+ without crypts/debris, no cranial neuropathies.

Diagnostic Testing:

- Selected laboratory values are shown in **Table 1**. Comprehensive metabolic panel and complete blood count were otherwise unremarkable. IgA, IgE, IgG, and IgM levels were normal at the second lab draw.
- Computed tomography (CT) Imaging (obtained prior to presentation): **Figure 1**
- Magnetic Resonance Imaging (MRI): **Figure 2**
- Incisional biopsy: Marked fibrosis of the submucosal tissue adjacent to the retromolar trigone, as well as the anterior border of the masseter muscle.
- Histologic findings: **Figure 3**

Treatment:

- Started prednisone (40mg daily for 1 week with subsequent taper) during post-operative course with significant improvement in her discomfort and trismus.
- Additional month of prednisone 10mg daily with ibuprofen for pain control prescribed per Rheumatology recommendation.
- Follow-up several months later revealed complete resolution of pain, swelling, and trismus.

Laboratory Values	Following first visit	Following treatment
White blood cell count	8.4 10e9/L	11.2 10e9/L
Erythrocyte sedimentation rate	**102mm in 1 hour**	**57mm in 1 hour**
C-reactive protein	**10.8 mg/dL**	**0.4 mg/dL**
ANA Screen	Negative	Not repeated
Cytoplasmic ANCA	<1:20 titer	Not repeated
Perinuclear ANCA	<1:20 titer	Not repeated
Atypical ANCA	<1:20 titer	Not repeated
Rheumatoid factor	<15 IU/mL	Not repeated

Table 1: Laboratory values following first clinical visit and after treatment with steroids. Significant values in bold/red. ANA = antineutrophil antibody ANCA = antineutrophil cytoplasmic antibody IU = international units

Imaging

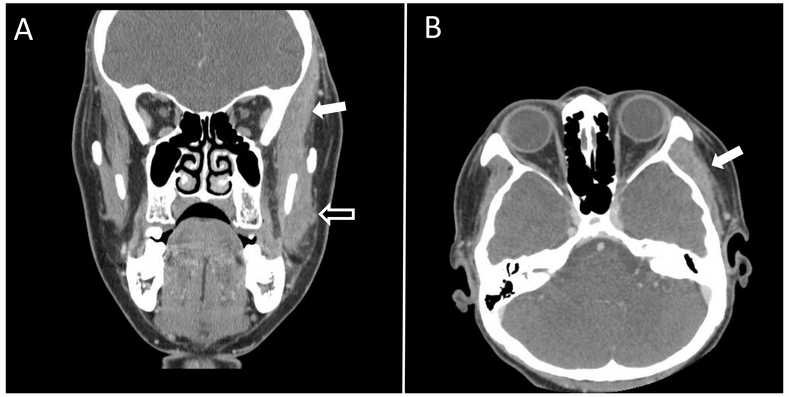

Figure 1: A) Coronal CT and B) axial CT imaging with contrast demonstrating diffuse thickening of the left temporalis (solid arrow) and masseter (hollow arrow) muscles without any discrete lesion. There is minor associated fat stranding in the left buccal subcutaneous fat.

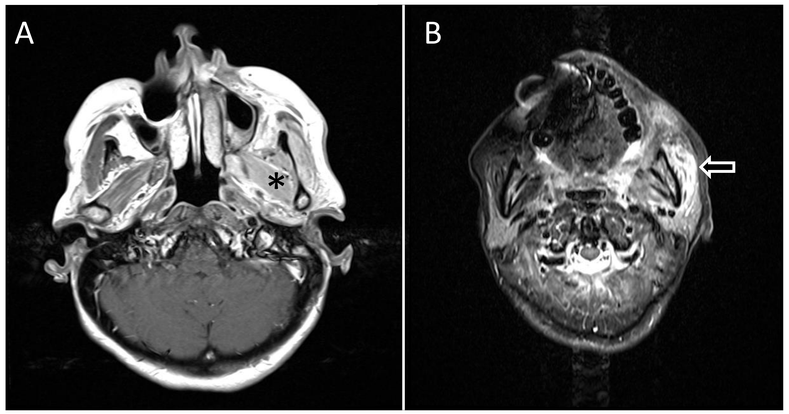

Figure 2: MRI demonstrating left lateral pterygoid (asterisk) and masseter (hollow arrow) muscle enhancement without any discrete lesion. A) T1-weighted post-contrast. B) T2-weighted STIR short-tau inversion recovery (STIR).

Histology

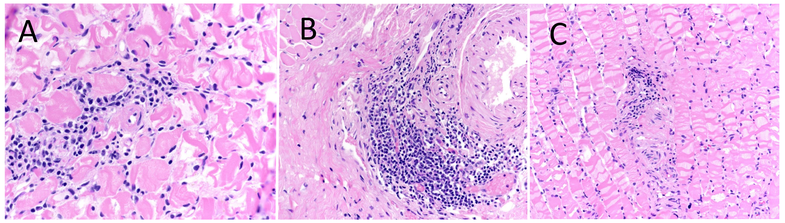

Figure 3: H&E-stained sections from incisional biopsy. A) 400x magnification. Rare focus of chronic perivascular inflammation comprised of small mature lymphocytes and plasma cells. The vessel does not demonstrate edema, fibrosis, or other sequelae of chronic inflammation. B) 100x magnification. Similar to A, with longitudinally oriented muscle fibers demonstrates contraction bands predominately as a result of surgical removal. C) 400z Magnification. A rare focus of chronic inflammation aggressive towards muscle fibers. The inflammation is comprised of small mature lymphocytes and plasma cells. This inflammation is aggressive towards muscle fibers which resultantly demonstrate necrosis. H&E = hematoxylin and eosin

Discussion

Proliferative myositis is an uncommon benign fibroproliferative pathology which rarely presents in the head and neck.[1] It will typically present as a rapidly growing intramuscular lesion in adults, often raising suspicion for sarcoma or other malignant process.[2] Involvement of the muscles of mastication can result in trismus, while sternocleidomastoid involvement can result in diminished neck mobility.[3] Historically, overly aggressive interventions including wide local excision have been performed due to concern for the potential of a malignant neoplasm.[4] Typical MRI findings are a hyperintense muscular mass with preserved muscle fascicles on T1 weighted images with fat suppression.[5] Characteristic histologic findings include immature myofibroblastic proliferation within skeletal muscle with associated basophilic, ganglion-like giant cells.[2] Treatment is focused on analgesic antipyretics including non-steroidal anti-inflammatory medications with corticosteroids for refractory or more severe cases, as the majority of patients will spontaneously improve within several weeks.[6] Differential diagnoses to consider include proliferative fasciitis, nodular fasciitis, myositis ossificans, sarcoma, phlegmon, and abscess.

Contact Information:
Kevin Quinn, MD
Otolaryngology – Head & Neck Surgery
VCU School of Medicine
Email: kevin.quinn@vcuhealth.org
The authors have no relevant disclosures.

References:

1. Fauser C, Nahrig J, Niedermeyer HP, Arnold W. Proliferative myositis: A rare pseudomalignant tumor of the head and neck. *Arch Otolaryngol Head Neck Surg. 2008;134:437–40.*
2. Gan S, Xie D, Dai H, et al. Proliferative myositis and nodular fasciitis: a retrospective study with clinicopathologic and radiologic correlation. *Int J Clin Exp Pathol*. 2019;12(12):4319-4328.
3. Sumi I, Kawabata I, Watanabe K, Kinoshita M. A case report of proliferative myositis of bilateral sternocleidomastoid muscles. *Pract Otorhinolaryngol. 1998;91:851–56.*
4. Enzinger FM, Dulcey F. Proliferative myositis. Report of thirty-three cases. *Cancer. 1967;20:2213–23.*
5. Jarraya M, Parva P, Stone M, Klein MJ, Guermazi A. Atypical proliferative myositis: original MR description with pathologic correlation: Case report. *Skeletal Radiol*. 2014;43(8):1155-1159.
6. Nishi TM, Yamashita S, Hirakawa YN, Katsuki NE, Tago M, Yamashita SI. Proliferative Fasciitis/Myositis Involving the Facial Muscles Including the Masseter Muscle: A Rare Cause of Trismus. *Am J Case Rep*. 2019;20:1411-1417.