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Miscellaneous

Inflammatory focal myositis of the sternomastoid muscle: is there an absolute indication for biopsy? A case report and review of the literature

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Abstract Focal myositis is a localised inflammatory process affecting skeletal muscles belonging to the pathological group of inflammatory pseudo tumours of soft tissue that includes myositis ossificans, proliferative myositis and nodular pseudosarcomatous fasciitis. Very rarely, it may affect one of the neck muscles and present as a neck lump, in which case both the clinical and pathological picture can mimic a sarcoma. We describe a case of focal myositis of the sternocleidomastoid muscle, present a review of this rare condition and debate the necessity of biopsy.

Keywords MESH · Neck muscles · Myositis

Introduction

The differential diagnosis of an enlarging neck lump includes early, localized presentation of polymyositis as well as the inflammatory pseudotumors of the neck muscles (myositis ossificans, proliferative myositis and nodular pseudosarcomatous fasciitis). Focal myositis is a relatively rare,

recent addition to this pathological group. There have been four reported cases of focal myositis affecting the sternomastoid muscle and presenting as a neck mass in an adult patient. We present a new case and review the current literature while debating whether biopsy is invariably indicated.

Case report

A 41-year-old Caucasian male presented with a painful, progressively enlarging neck mass of 10-day duration. He had completed a 7-day course of erythromycin, as prescribed by his general practitioner, with no improvement. He had given a history of a mild sore throat 3 weeks before, but denied any history of local trauma or recent surgery, and did not complain of dysphagia or proximal muscle weakness. On examination he had moderate pyrexia (38°C). An ill-defined, tender, non-fluctuant mass measuring approximately 6×6 cm in diameter involved the upper two-thirds of his left sternocleidomastoid muscle (Fig. 1). The overlying skin was normal in appearance. He had no muscle weakness, although head rotation to the affected side was painful. Laboratory haematological investigations showed a slightly elevated white blood cell count ($12 \times 10^9/l$) and raised C-reactive protein (37 mg/dl), while the remaining biochemical results (including creatine phosphokinase) were within normal range. An urgent ultrasound revealed diffuse swelling of the sternomastoid muscle with no evidence of a collection. It was felt that an infective process accounted for his symptoms, and administration of intravenous benzyl penicillin and metronidazole was initiated. Despite 2 days of treatment, his pyrexia persisted, the mass was unchanged and the inflammatory markers continued to be elevated. The advice of microbiologists was sought and intravenous levofloxacin was added, with the aim of covering gram-negative organisms, including *pseudomonas aeruginosa*. Twenty-four hours later there was no improvement in the patient's symptoms so further imaging in the form of a computed tomography of the neck was undertaken and fine-needle aspiration of the mass was performed. Cytology of the aspirate showed serous fluid and inflammatory cells, including neutrophils, while the scan revealed isolated oedema of the sternomastoid muscle. The fascial planes were intact and there was no collection or cervical lymphadenopathy (Fig. 2). On the basis of the FNA and CT findings a tentative diagnosis of focal myositis was made. The patient was started on oral prednisolone. The following day there was a dramatic reduction in the size of the lump as well as improvement in his overall condition. Two days later the mass was half the original size, and the patient was discharged on a 5-day decreasing course of oral prednisolone. On follow-up a month later, the mass had completely disappeared. Over the subsequent outpatient visits 2 and 4 months after his discharge the patient

remained well with no further attacks of this or other groups of muscles while no neck mass was palpable. Repeated biochemical and immunological investigations, including rheumatoid factor, anti-nuclear antibodies and creatine phosphokinase, were normal. The patient remains on routine 6-month follow-up.



Fig. 1 Clinical photograph of the patient showing a diffuse swelling involving the superior part of the sternocleidomastoid muscle

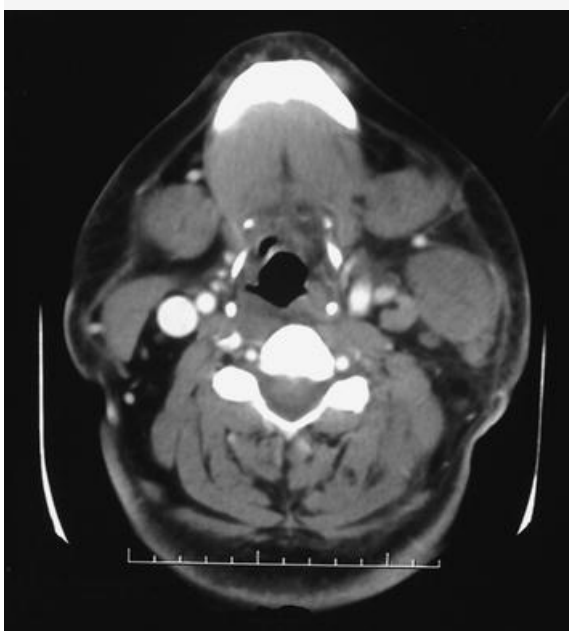


Fig. 2 Axial computed tomography scan showing isolated oedema of the sternomastoid muscle, with intact fascial planes and absence of collection or lymphadenopathy

Discussion

Focal myositis, as described by Heffner [1] in 1977, is a self-limiting, localized inflammation of skeletal muscles of unknown aetiology (although viruses [2] and muscle denervation [3] have been implicated in its pathogenesis). It was initially described affecting the lower extremity muscles. There have been about 30 cases of focal myositis in the English-language literature, with only four cases affecting the sternomastoid muscle [4, 5, 6, 7] in adults. In one of these cases it was associated with orbital myositis and in another with mixed connective tissue disease. Clinically focal myositis presents as a painful swelling of a skeletal muscle with minimal systemic symptoms or signs (which differentiates it from an early, localised form of polymyositis [8] and no history of trauma (as opposed to myositis ossificans [9]. In most cases WBC count and ESR are either normal or minimally elevated while CPK and aldolase are invariably within normal limits. Macroscopically the affected muscle appears pale, rubbery and oedematous, with no involvement of the fascia, tendon or subcutaneous tissue and raises the possibility of a sarcoma. The histological picture is characteristic, with lymphocytic and other inflammatory cell infiltration of necrosed and regenerating muscle fibres. In more chronic cases, there is connective tissue production in the endomysium and perimysium. This histological differentiation between an early, acute stage and more chronic lesions is also reflected in clinical presentation and inflammatory markers: Although in the original case series of Heffner all patients were afebrile and WBC as well as CRP and ESR were not elevated; in subsequent reports some patients had moderate pyrexia [6], while the white blood cell count and ESR were elevated [5, 6], in keeping with an earlier, acute stage and the presence of the mass for a smaller time period. In these early cases fine-needle aspiration cytology suggested [5] or confirmed [10] the diagnosis in two cases by showing inflammatory cells and failing to show any neoplastic cells.

Neck lumps are one of the most common presentations to an otolaryngology clinic. Neoplastic, infectious, traumatic, congenital, inflammatory and metabolic causes are included in the differential diagnosis; however, a short history of a painful neck lump in a patient with external symptoms and signs of inflammation would direct most clinicians towards an infectious cause. Following appropriate antibiotic treatment and exclusion of a collection and necrotising fasciitis, the logical next step would be, in most cases, the performance of fine-needle aspiration and (after exclusion

of squamous cell carcinoma) an open biopsy with or without panendoscopy in order to exclude a neoplastic process. However, what most otolaryngologists tend to overlook are non-infective, inflammatory causes of neck lumps. A mass within a muscle that could be a neoplasm could as well be a pseudo sarcomatous lesion of soft tissue—notably myositis ossificans, proliferative myositis, nodular pseudosarcomatous fasciitis and focal myositis. In our case, the history as well as clinical examination pointed towards an inflammatory process, while imaging confirmed the presence of a lesion confined within the sternomastoid muscle. Normal CPK as well as lack of other muscle involvement directed us away from systemic myopathies including polymyositis or dermatomyositis, both of which can initially present as a localised process [11]. From the group of localised pseudo sarcomatous lesions, myositis ossificans characteristically follows trauma, while the radiological picture is pathognomonic [9]. Radiology and FNA could also point away from nodular fasciitis and proliferative myositis (absence of involvement of muscle fascia and inflammatory cells rather than hypercellular material in FNA). The dramatic resolution of the mass following the administration of steroids confirmed our diagnosis. Sarcoma was a very real possibility, however, and neither the mass' response to steroids nor the negative FNA could exclude it. However, the fact that the mass disappeared completely after a week of steroid treatment and remained non-palpable subsequently, effectively rule it out. Although fine-needle aspiration can be very helpful in the diagnosis of pseudo sarcomatous muscle lesions [12], we believe, like Wong et al. [13], that any suspicious lesion that does not regress within 4 to 6 weeks should be subjected to biopsy.

We believe that the combined role of imaging and fine-needle aspiration can point a clinician towards the diagnosis of focal myositis. In this context a therapeutic trial of steroids is indicated. However one should always keep in mind the possibility of neoplasia. In these cases the importance of careful follow-up cannot be overemphasized.

References

1. Heffner RR, Armbrustmacher VW, Earle KM (1977) Focal myositis. *Cancer* 40:301–306
2. Toti P, Romano L, Villanova M, Zazzi M, Luzi P (1997) Focal myositis: a polymerase chain reaction analysis for a viral etiology. *Hum Pathol* 28:111–113
3. Heffner RR Jr, Barron SA (1980) Denervating changes in focal myositis, a benign inflammatory pseudotumor. *Arch Pathol Lab Med* 104:261–264
4. Ho KL, Konno ET, Chason JL (1979) Focal myositis of the neck. *Hum Pathol* 10:353–356
5. Josephson GD, de Blasi H, McCormick S, Sabini P, Goldberg J, Pincus R (1996) Focal myositis of the sternocleidomastoid muscle: a case report and review of the literature. *Am J Otolaryngol* 17:215–217

6. Rivest C, Miller FW, Love LA, Turgeon PP, Blier C, Senecal JL (1996) Focal myositis presenting as pseudothrombophlebitis of the neck in a patient with mixed connective tissue disease. *Arthritis Rheum* 39:1254–1258
7. Cain AJ, Michie BA, Davis BC, Ram B (1998) Focal myositis of the sternocleidomastoid muscle. *J Laryngol Otol* 112:687–689
8. Heffner RR Jr, Barron SA (1981) Polymyositis beginning as a focal process. *Arch Neurol* 38:439–442
9. Sarac S, Sennaroglu L, Hosal AS, Sozeri B (1999) Myositis ossificans in the neck. *Eur Arch Otorhinolaryngol* 256:199–201
10. Dhanasekar G, Rajan MS, Nirmal Kumar B, Watson SD (2002) Focal myositis of the neck with idiopathic orbital myositis. *J Laryngol Otol* 116:314–316
11. Wanamaker JR, Wanamaker HH, Lavertu P (1992) Polymyositis presenting as a neck mass. *Arch Otolaryngol Head Neck Surg* 118:318–320
12. Dodd LG, Martinez S (2001) Fine-needle aspiration cytology of pseudosarcomatous lesions of soft tissue. *Diagn Cytopathol* 24:28–35
13. Wong NL (2002) Fine needle aspiration cytology of pseudosarcomatous reactive proliferative lesions of soft tissue. *Acta Cytol* 46:1049–1055