Case Reports

Focal Myositis – an atypical presentation of a rare disease

José Barata*, Nuno Carvalho*

Abstract

A clinical case of tumoral lesion in a paravertebral location leading to a focal myositis diagnosis is presented. This disease seldom occurs triggering as a rule a number of doubts in the

Introduction

Focal myositis is a pseudotumoral lesion of the skeletal muscle, with an inflammatory character of benign course, unknown etiology, compromising the extremities selectively. This nosological entity identified in 1977 is deemed a rare clinical condition and only around one hundred cases have been published in the world literature. The diagnosis is essentially clinical and histological, with a differential diagnosis of malignant tumor of muscle origin, focal infection or venous thrombosis.

Clinical case

Female patient, 67 years old, Caucasian, a gardener, widower. She mentions a painful tumefaction appearing on the left paravertebral region, growing progressively in the last 10 months, followed by a spontaneous moderate local pain, getting worst on dorsal decubitus. She denied any other focal or general symptomatology. She went to her physician, underwent analyses, and thorax and abdomen CAT scans, being referred to Surgery for a lesion biopsy.

Accepted for publication on the 20th April 2008

differential diagnosis which in the current case has the particularity of a emerging in a rare topography.

Key words: focal myositis, striated muscle, pseudotumor.

She was then referred to the Medicine consultation for reassessment and evaluation of the histological result.

In her personal history it was highlighted controlled and medicated hypertension; surgical intervention for stress urinary incontinence at 47 years of age; bilateral carpal tunnel surgically corrected 7 years ago; hysterectomy due to a glandular uterine polyp 2 years ago. No diseases of a family hereditary characteristic. The family history was irrelevant.

In the objective exam there were no changes in the general condition, only to highlight the presence of an ovoid form tumefaction located on the left paravertebral region, about 1 cm [0.4 inch] off the spine, spreading from D12-L4, with a longitudinal diameter about 11 cm [4.3 inch] and a transversal of 8 cm [3.15 inch], without visible inflammatory signs, on which it could be seen a surgical scar 7 cm [2.75 inch] long, after biopsy. It had well defined limits, elastic consistency and was painful to palpation (*Fig.1*).

In the auxiliary exams it could be verified a moderate increase on CK (459 UI/L), with the reminder hematologic and biochemical values within normal ranges. Thorax and abdomen CAT scan has shown dorsolumbar muscles asymmetry through a suggestive process of fat infiltration on the left dorsal plan reaching the whole *erector spinae*, which is kept confined to the aponeurosis, although it makes it slightly rounded (*Fig. 2*).

The muscular biopsy has identified material made up of striated muscular tissue where focal necrosis of muscular fibre, its degenerative aspects, adipose regression and focal fibrosis are observed. It can also be seen an inflammatory infiltrate made up by mononucleated elements. Such aspects were considered

^{*}Department of Medicine

Garcia de Orta Hospital, Almada

Received for publication on the 9th January 2007

Paper presented as Poster on the 12nd Internal Medicine National Congress (Porto, 2006)







compatible with an inflammatory myopathy process (*Fig. 3 and 4*). It was accepted a focal myositis diagnosis and the patient was medicated with prednisone, dosage of 40 mg/day, with a total lesion remission after 4 weeks of therapy. The choice of corticotherapy was based on the slow progression of the disease and the painful symptomatology triggered by dorsal decubitus.

Discussion

Focal myositis was separated as an autonomous nosological entity by Heffner, in 1977¹. It is considered a rare clinical condition, and around one hundred cases are described in the global literature.^{2,3} Clinically, it is translated by an inflammatory process located in the striated muscle, of pseudotumoral expression and

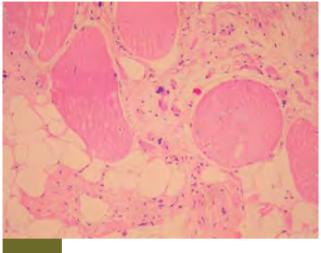
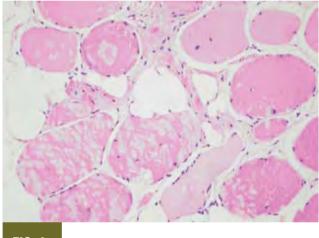


FIG. 3





benign character, with slow growth (weeks/months), with sizes ranging from 2 to 10 cm [0.8 to 3.9 inch].^{1,4-} ⁸ It is usually located in the extremities, having as pathognomonic feature the absence of simultaneous systemic symptomatology.^{1,4,5,6,8,9} Local pain, usually mild, is a frequent symptom, however difficult painful conditions related with the compression of adjacent structures can arise.^{3,10,11} There is not a preferential distribution based on gender or age range, and cases are known both in children as in the elderly.^{1,4,5,6} It is characteristic the compromise of an isolated muscle, usually the lower limbs (75% of cases),^{1,3,4,5,6} although, there are other locations described, namely the sternocleidomastoid, rectus abdominis, facial and tongue muscles, and seldom paravertebral muscles.^{1,3,4,6,8,} ^{9,12,13} It usually evolves with a surprising analytical

normality, namely in the inflammatory processes and sedimentation rate, emerging in some cases, a moderate increase on CPK.^{1,3,4,6,7,8,9,12,14,15} The clinical evolution tends to a spontaneous regression^{3,5,7,8} although a number of recurring situations have been described.^{16,17,18,19} Corticosteroids and immunosuppressants are recommended in the long term, compressive symptomatology and also in recurring conditions.^{3,4,7,10,11,14,15,16,20}

Some authors suggest, in a non consensual manner, the possibility of this nosological entity evolving to systemic polymyositis.^{3,4,6,9,15,18,21-24} It is a clinical and histological diagnosis, although the imagiology aspects (CAT scan and Nuclear Magnetic Resonance imagiology) of muscle tumefaction and adipose infiltration are considered very suggestive.^{5,6,8,15,20,25} The etiology is unknown, with a speculating discussion on the infectious pathogenesis (viral) and immunologic mediation or genetic predisposition.^{1,3,4,5,26,27} The differential diagnosis is made with striated muscle malignant neoplasm, focal infection or venous thrombotic lesion.^{3,4,7,9}

In the current case, it should be noticed the rarity of this nosological entity without any case known being previously published in Portugal, and to be highlighted the unusual lesion topography with only 4 similar locations identified in the literature.

Acknowledgements

The authors would like to express their appreciation to Dr. Paula Borralho, from the Anatomic Pathology Service of Garcia da Orta Hospital, for all the support given in the current case study.

References

1. Heffner RR Jr, Armbrustmacher VW, Earle KM. Focal myositis. Cancer. 1977; 40(1):301-306.

2. Yanmaz Alnigenis MN, Kolasinski SL, Kalovidouris AE. Focal myositis: a review of 100 previously published cases and a report of 2 new cases. Clin Exp Rheumatol 1999;17(5):631.

3. Mielnik P, Chwali ska-Sadowska H, Wagner T. Focal myositis. Case Rep Clin Pract Rev 2003; 4(3):157-159.

4. Kocanaogullari H, Ozdemir E, Keser G et al. Focal myositis. Clin Rheumatol. 1998;17(1):65-67.

5. Llauger J, Bagué S, Palmer J et al. Focal myositis of the thigh: unusual MR pattern. Skeletal Radiol 2002; 31:307–310.

6. Kransdorf M J, Temple HT, Sweet D E. Focal myositis. Radiol 1998; 27:283-287.

7. Cheng N, Taylor SM, Bullock M, Hanly J.Focal myositis of the sternocleidomastoid muscle. Otolaryngol Head Neck Surg 2005;132(1):150-151.

8. Galloway HR, Dahlstrom JE, Bennett GM.Focal myositis. Australas Radiol

2001;45(3):347-349.

9. Gordon MM, Madhok R.Recurrent focal myositis. Rheumatology 1999; 38: 1295-1296(Letter)

10. Alzagatiti BI, Bertorini TE, Horner LH et al T. Focal myositis presenting with radial nerve palsy. Muscle Nerve. 1999 ; 22(7):956-9.

11. Streichenberger N, Meyronet D, Fiere V, Pellissier JF, Petiot P. Focal myositis associated with S-1 radiculopathy: report of two cases. Muscle Nerve 2004 ;29(3):443-446.

12. Hepburn A, Damani N, Sandison A, Pandit N.Idiopathic focal myositis in pregnancy. Rheumatology (Oxford) 2000 ;39(2):211-213.

13. Wunderlich S, Csoti I, Reiners K, Gunthner-Lengsfeld T, Schneider C, Becker G, Naumann M.Camptocormia in Parkinson's disease mimicked by focal myositis of the paraspinal muscles. Mov Disord 2002;17(3):598-600.

14. Georgalas C, Kapoor L, Chau H, Bhattacharyya A.Inflammatory focal myositis of the sternomastoid muscle: is there an absolute indication for biopsy? A case report and review of the literature. Eur Arch Otorhinolaryngol 2006; 263(2):149-151.

15. Kalden P, Krause T, Volk B, Peter HH, von Kempis J.Myositis of small foot muscles. Rheumatol Int 1998;18(2):79-82.

16. Grace KL, Wilson YG, Collins CM, Kirwan JR, Baird RN.Focal myositis - a new presentation. Eur J Vasc Endovasc Surg 2000;19(1):90-91.

17. Kisielinski K, Miltner O, Sellhaus B, Kruger S, Goost H, Siebert CH. Recurrent focal myositis of the peroneal muscles. Rheumatology (Oxford) 2002;41(11):1318-1322.

18. Misu T, Tateyama M, Nakashima I, Shiga Y, Fujihara K, Itoyama Y.Relapsing focal myositis: the localization detected by gallium citrate Ga 67 scintigraphy. Arch Neurol 2005;62(12):1930-1931.

19. Revaz S, Theumann N, Lobrinus JA, So AK, Dudler J. Leg pain due to bilateral focal recurrent myositis in a hemodialysis patient. Am J Kidney Dis. 2005;45(1):e7-11.

20. Gaeta M, Mazziotti S, Toscano A, Rodolico C et al. "Dropped-head" syndrome due to isolated myositis of neck extensor muscles: MRI findings. Skletal Radiology 2006;35(2): 110 -112.

21. Sekiguchi K, Kanda F, Oishi K, Hamaguchi H, Nakazawa K, Maeda N, Ishihara H, Chihara K.HLA typing in focal myositis. J Neurol Sci 2004;227(1):21-25.

22. Flaisler F, Blin D, Asencio G, Lopez FM, Combe B.Focal myositis: a localized form of polymyositis? J Rheumatol 1993;20(8):1414-1416.

23. Smith AG, Urbanits S, Blaivas M, Grisold W, Russell JW. Clinical and pathologic features of focal myositis. Muscle Nerve 2000;23(10):1569-1575.

24. McCluggage WG, Mirakhur M. Focal myositis of the floor of mouth: report of two cases and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1996;81(5):573-575.

25. Moskovic E, Fisher C, Westbury G, Parsons C. Focal myositis, a benign inflammatory pseudotumour: CT appearances. The British Journal of Radiology 1991; 64 (62): 489-493.

26. Toti P, Romano L, Villanova M, Zazzi M, Luzi P. Focal myositis: a polymerase chain reaction analysis for a viral etiology. Hum Pathol 1997;28(1):111-113.

27. Naggar EA, Kanda F, Okuda S, Maeda N, Nakazawa K, Oishi K, Sekiguchi K, Ishihara H, Chihara K. Focal myositis in monozygotic twins. Intern Med 2004;43(7):599-601.