

Non-Traumatic Myositis Ossificans Case First Detected in Terms of LocalizationKaygusuz U¹, Akyer SP², Kurbetli N^{3*}, Aygün D⁴ and Özdemir MB⁵¹Ear, Nose and Throat Specialist, Private Odak Hospital, Ear Nose Throat (Otorhinolaryngology) Diseases, Turkey²Assoc. Prof, Pamukkale University, Medical Faculty, Department of Anatomy, Turkey³Pamukkale University, Medical Faculty, Department of Anatomy, Turkey⁴Pamukkale University, Medical Faculty, Department of Anatomy, Turkey⁵Prof, Pamukkale University, Medical Faculty, Department of Anatomy, Turkey***Corresponding author:**

Nuriye Kurbetli,
 Pamukkale University, Medical Faculty,
 Department of Anatomy, Tel: 0090 5054006271,
 Turkey, E-mail: nurkurbetli@gmail.com

Received: 05 Oct 2021

Accepted: 12 Oct 2021

Published: 17 Oct 2021

Copyright:

©2021 Kurbetli N. This is an open access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and build upon your work non-commercially.

Keywords:

Myositis Ossificans; Non-Traumatic Myositis Ossificans; Rare Localization; Regio Cervicalis Lateralis; Three-Dimensional Imaging

&Authors Contribution:

Kaygusuz U, Akyer SP, Kurbetli N, Aygün D, Özdemir MB all are equally contributed to this work

Citation:

Kurbetli N. Non-Traumatic Myositis Ossificans Case First Detected in Terms of Localization. *Ame J Surg Clin Case Rep.* 2021; 3(14): 1-6

1. Abstract**1.1. Objectives**

Myositis ossificans (MO) is a heterotopic bone formation in muscle tissue that usually occurs due to trauma. The reported without trauma of it is rare. In this case, MO was detected in the supraclavicular portion of the plexus brachialis and between the musculus (m.) scalenus anterior and m. scalenus medius. A previously described case was not found in the literature.

1.2. Methods

This case was seen in a 22-year-old female patient with no history of trauma and neck pain. A painful mass was revealed in the regio cervicalis lateralis on the left side of her neck, adhering to the scalene muscles according to physical examination and radiological evaluations (MRI). The mass was excised under appropriate anesthetic conditions. Mature lamellar bone tissue at the periphery, increased cellularity at the central, swollen osteoblasts in the middle zone and osteoid formation were observed in the pathology report. Based on these findings, myositis ossificans was diagnosed.

1.3. Results

This case is a type of hereditary, progressive myositis ossificans that is both risky and uncommonly developed without trauma.

This mass was formed between m. scalenus anterior and m. scalenus medius, and in the fascia or aponeurosis above the supraclavicular part of the plexus brachialis.

1.4. Conclusion

The identification of such rare cases is important in guiding physicians in the related disciplines.

2. Introduction

Myositis ossificans (MO) is a non-inflammatory disease that results in heterotopic bone formation in muscle tissue [1]. MO may develop at any age, but is most commonly seen in adolescents and young adults [2,3].

MO is a self-limiting, benign disease. The diagnosis using imaging techniques is important for distinguishing MO from malignancy or infectious processes [1,4]. The conditions to be considered in the differential diagnosis are progressive fibrodysplasia ossificans, ossified fibromixoid tumour, osteochondroma, infections and sarcoma [5].

In 75% of cases, MO usually has a history of trauma, while the remainder may not have a history of trauma. The probability of developing MO varies with the severity of trauma [6]. Except traumatic type, other common types of MO are pseudo malign / non

traumatic myositis ossificans and progressive myositis ossificans [7].

Patients are usually asymptomatic, but may have pain, erythema, and sensitive focal swelling. In some patients, calcifications may heal spontaneously and symptoms may be significantly reduced [8].

Here, we report a 22-year-old female patient with myositis ossificans in the left regio cervicalis lateralis without trauma.

3. Case Report

A 22-year-old female patient presented to our clinic with a sudden onset of pain in the left side of the neck for a month. The history revealed that the patient lived a sedentary life, had no trauma, and did not engage in any sporting or compelling activity.

It was thought that the appearance in infectious diseases may be related to infectious and neoplastic processes or posttraumatic muscle and soft tissue changes and hematoma. The infectious diseases clinic consulted us for the differentiation of abscess and neoplasia. In our physical examination; we found a hard, fixed and extremely painful, 4 × 5 cm mass lesion palpated deeply in the left lateral regio cervicalis.

The patient was sampled by fine needle aspiration biopsy due to considering the presence of a neoplastic lesion. The infection parameters were negative. There was no response to empirical antibiotherapy. As the patient's pain unresponsive to analgesics continued, surgical exploration of the neck was decided for diagnostic and therapeutic purposes.

Radiological evaluations (MRI) revealed a calcified and irregular mass on the left side of the regio cervicalis lateralis, adhering to the scalene muscles. In the surgical neck exploration, a very hard mass was detected between the m. scalenus anterior and the m. scalenus medius adhering to the surrounding tissue in the upper part of the supraclavicular part of the plexus brachialis (Figure 1).

The mass was excised by dissection from the fascia outside without damaging the N. accessorius and plexus brachialis (Figure 4-5).

The pathological examination revealed a brown material (3.5 cm in diameter) with gray nodules on the outer face, and calcified gray-brown lesion on sections of this material. In addition, two adipose tissue materials (one with a diameter of 3 cm and the other with a diameter of 1 cm) and two nodular tissues (5 mm in diameter) were observed in the sections made on these materials.

In microscopic findings, zonal phenomenon is observed. Peripheral mature lamellar bone tissue, increased cellularity at the central and swollen osteoblasts in the middle zone and osteoid formation were observed (Figure 6).

Myositis ossificans was diagnosed by supporting histopathological findings with clinical and radiological findings.

In order to understand its relationship with the surrounding tissues, two-dimensional MR images were rendered three-dimensional using Ossirix program (Figure 2-3). The surface area and volume of the mass were calculated (Table 1).

The patient whose postoperative pain decreased dramatically was discharged with stretching exercise and close control.

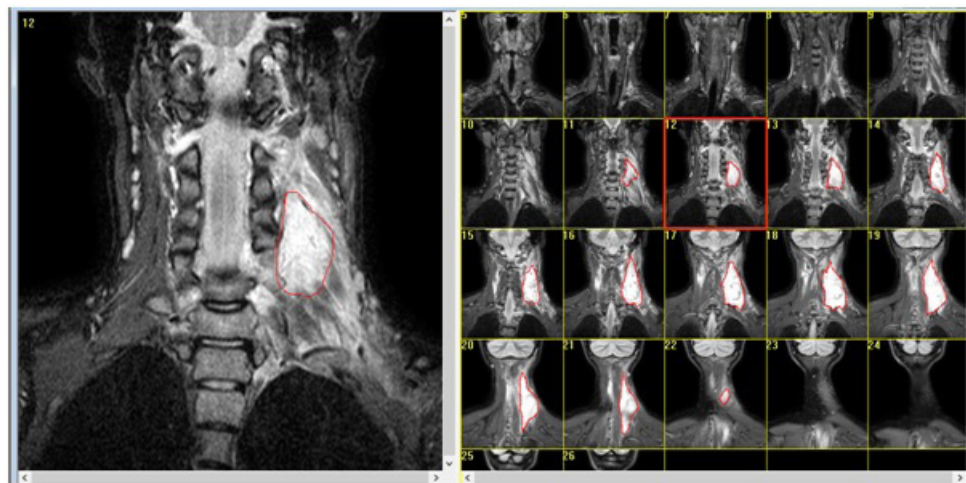


Figure 1. Plain radiography showing a defined mass in the upper part of the supraclavicular region of the brachial plexus in the left regio cervicalis lateralis.

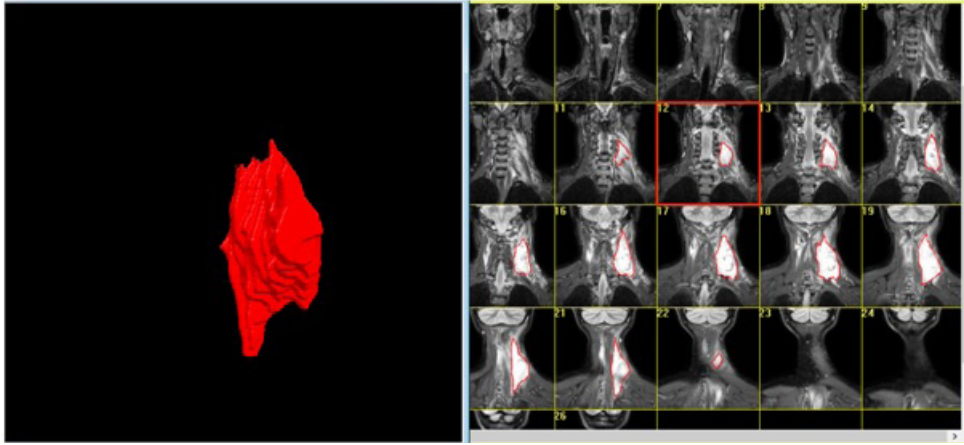


Figure 2. Selecting MR images in Ossirix program to obtain a three-dimensional image of the mass.

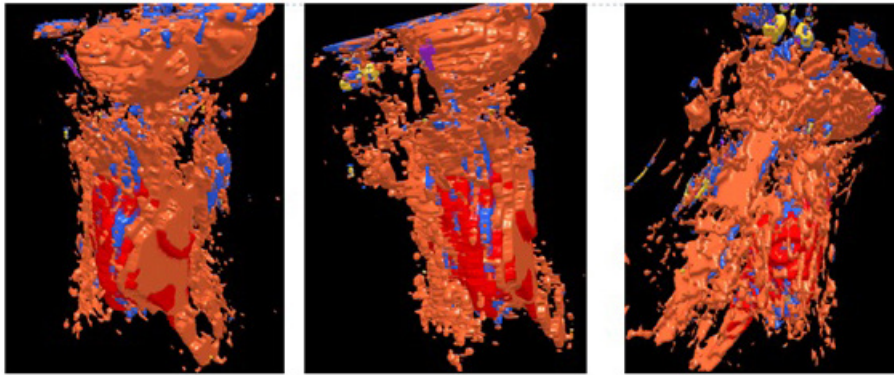


Figure 3. The mass of myositis ossificans was selected in red. Relation of mass with neck structures.

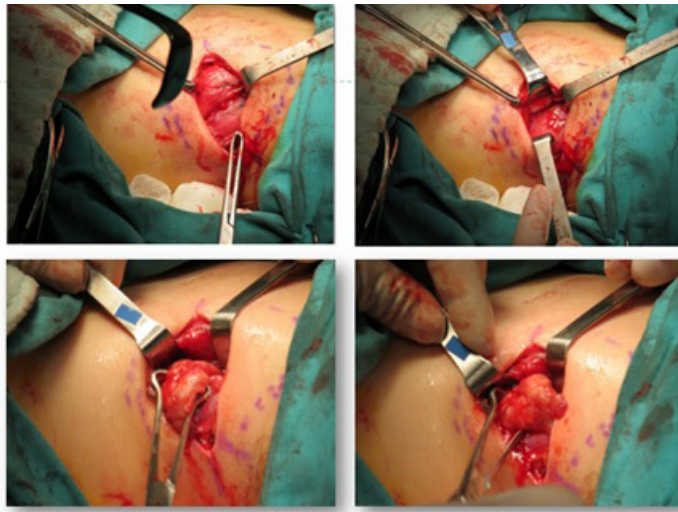


Figure 4. Intraoperative image showing a well-circumscribed mass.



Figure 5. The appearance of the postoperative mass (4×5 cm).

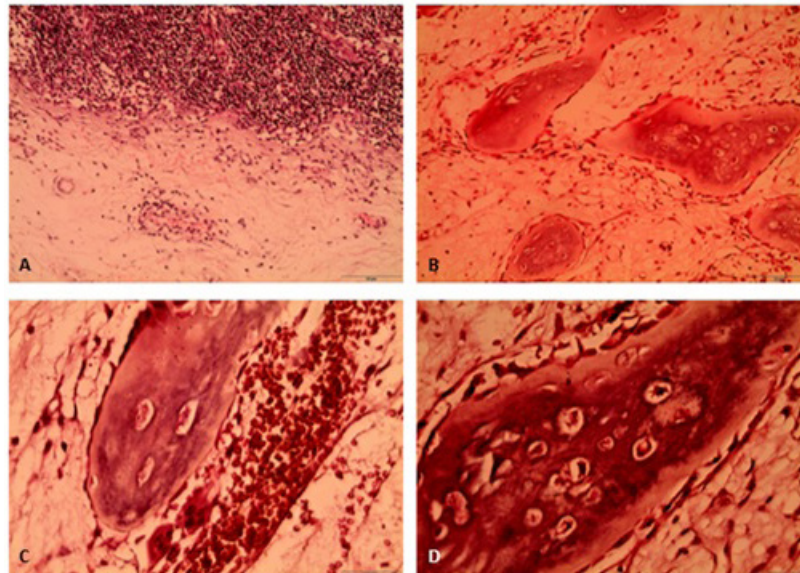


Figure 6. Microscopic appearance of the lesion. A and B 20 \times . C and D 100 \times .

Table 1: Volume and surface area of the mass in three-dimensional evaluation.

	volume	surface
myositis ossificans	98964,11mm ³	16394,35 mm ²

4. Discussion

Myositis ossificans is a benign formation in which heterotopic calcification develops in soft tissue. It occurs in adolescents and young athletic adults, especially males. It is most commonly seen in the anterior and posterior proximal locations of the thigh and calf. However, lesions in the gluteal muscles, pectoral muscles, m. brachialis, m. temporalis, tendons, breasts, elbows and knees have been reported [9, 10].

The patient in this case was a 22-year-old woman. In this patient, myositis ossificans was found in the left regio cervicalis lateralis. The mass is located between the m. scalenus anterior and m. scalenus medius. Arteria subclavia and plexus brachialis are located between these two muscles. The anterior branches of the spinal nerves form the plexus brachialis, and these branches are called radix. Radixes combine to form trunks. Radixes and trunks are located in the supraclavicular part of the plexus brachialis. The nervus (n.) dorsalis scapulae originating from the C 5 radix and the n. thoracicus longus (Bell nerve) from the C 5 - 7 radixes penetrate the m. scalenus medius. N. suprascapularis from the truncus innervates m. supraspinatus and m. infraspinatus. M. supraspinatus is the muscle that makes the first 15° abduction of the arm.

The mass in this region presses the roots and truncus. In such cases, there may be no signs other than pain and swelling. However, the mass may cause clinical pictures resembling torticollis or neck hernia due to the pressure exerted on the site. Removal of the mass removes the pain that causes MO. In this case, it was thought that this pressure effect was the reason for the patient's difficulty in

abducting the arm and severe pain spreading from shoulder to arm.

When MO occurs in areas outside the extremities, the diagnosis may be difficult, especially if there is no history of trauma [11]. The neck region is a place where MO is rare. It is difficult to distinguish MO from bone and soft tissue malignancies when making a diagnosis. However, excellent results can be achieved with appropriate treatment with the correct diagnosis. One of these treatment methods is surgical removal of the mass. When atraumatic myositis ossificans develops in the neck or shoulder region, it is important to consider fibrodysplasia ossificans progressiva (FOP) or Munchmeyer's disease in the differential diagnosis [12,13].

FOP is characterized by progressive heterotopic ossification of connective tissue and congenital malformation of the big toe. In a study by Cohen et al, the mean age of onset of ossification in 44 patients with FOP was 5 [14]. The most common sites of early heterotopic ossification were neck, spine and shoulder girdle. Heterotopic ossification progresses from proximal to distal, cranial to caudal, or from axial to appendicular. Other publications have reported that FOP starts in the first decade of life, especially in the head and neck region [15-17]. Our patient was 22 years old and had no congenital malformation of the big toe. According to Sferopoulos et al, non-traumatic idiopathic / pseudomalign MO may be a rough form of FOP [18].

In 1999, Sarac et al. published a case of post-traumatic MO involving the m. scalenus medius of a 53-year-old man; the authors stated that their cases were the first cases of MO originating from the m. scalenus medius [19]. Our case is also the first reported

case of MO between m. scalenus anterior and m. scalenus medius without trauma.

In the literature, although the number of cases reported after trauma is high [20,21,22], cases reported without trauma are rare [23,24]. There was no coercive activity or trauma in the history of our case. Although MO may occur at any age, it is most commonly seen at the age of 20-30; m. quadriceps femoris, m. brachialis and adductor muscles are the most commonly affected areas [25]. The case is compatible with the literature in terms of age, but it will be a new case in the literature in terms of its appearance.

The informations on the pathophysiology of MO is still unclear. The causes of MO may be calcium accumulation in adjacent skeletal or soft tissue edema, tissue hypoxia due to circulatory reduction, or local factors like accumulation of osteoblastic mesenchymal cells, and interactions among unknown systemic factors. There are also theories involving intramuscular hematoma transfer to bone, hematoma calcification, intramuscular bone formation from separated periosteal flaps, osteoblast proliferation from periosteal rupture, metaplasia of intramuscular connective tissue cells, and individual predisposition [26]. MO may be confused with different diagnoses due to these pathophysiological features. Neoplastic and infectious pathologies such as osteosarcoma, osteochondroma, posttraumatic periostitis, osteomyelitis, tumoral calcinosis should be considered in the differential diagnosis [27]. In this case, MRI images were evaluated and laboratory tests and fine needle biopsy were performed to make differential diagnosis. As a result, neoplastic and infectious diagnoses were excluded.

Diagnosis of MO may be difficult, even in multiple imaging methods, especially when developing atypical areas such as the neck and in the absence of trauma [28]. MO can be confused with bone and soft tissue tumors and infection. Good evaluation of radiological imaging features is important in the differential diagnosis of tumors such as parosteal osteosarcoma, osteochondroma, synovial sarcoma, chondrosarcoma, fibrosarcoma, and tumoral calcinosis [29]. Our case confirms the good prognosis of MO. It supports that the diagnosis of MO should be considered in patients presenting with painful soft tissue swelling.

5. Conclusion

As a result, it is easy to diagnose when MO occurs in areas where it is frequently seen. However, when it occurs in very rare regions such as this case, a good history, physical examination, laboratory tests and radiological examinations are necessary for differential diagnosis. In this case, a young female patient was diagnosed with non-traumatic MO in the neck region. The definition of this case will raise awareness in the literature and contribute to clinician.

6. Acknowledgment

All our work was carried out with the approval of Pamukkale University Non-Interventional Clinical Research Ethics Committee.

References

1. Lau J, Hartin CW, Ozgediz DE. Myositis ossificans requires multiple diagnostic modalities. *J Pediatr Surg*. 2012; 47: 1763-6.
2. Parikh J, Hyare H, Saifuddin A. The imaging features of post-traumatic myositis ossificans, with emphasis on MRI. *Clin Radiol*. 2002; 57: 1058-66.
3. Hanquinet S, Ngo L, Anooshiravani M, Garcia J, Bugmann P. Magnetic resonance imaging helps in the early diagnosis of myositis ossificans in children. *Pediatr Surg Int*. 1999; 15: 287-9.
4. Sirvanci M, Ganiyusufoglu AK, Karaman K, Tezer M, Hamzaoglu A. Myositis ossificans of psoas muscle: magnetic resonance imaging findings. *Acta Radiol*. 2004; 45: 523-5.
5. Onen MR, Varol E, Tosun Mİ, Naderi S. Non-traumatic myositis ossificans as an uncommon cause of scoliosis: a case report and discussion of literature. *World Neurosurg*. 2019; 123: 208-11.
6. Kaya T. Kas İskelet-Yumuşak Doku Radyolojisi. *Nobel&Güneş* 2008;279-282.
7. Parikh J, Hyare H, Saifuddin A. The imaging features of post-traumatic myositis ossificans, with emphasis on MRI. *Clin Radiol*. 2002; 57: 1058-66.
8. Hait G, Boswick Jr JA, Stone NH. Heterotopic bone formation secondary to trauma (myositis ossificans traumatica). *J Trauma*. 1970; 10: 405-11.
9. Hanna SL, Magill HL, Brooks MT et al. Cases of the day. *Radiographics*1990; 10: 945-9.
10. Galanis N, Stavraka Chara, Valavani MR, E, Kirkos J. Unsupervised Exercise-Induced Myositis Ossificans in the Brachialis Muscle of a Young Healthy Male Orthop J Sports Med. 2017; 5: 2325967117718780.
11. Dubuisson A, Lombard A, Otto B. Pseudomalignant Myositis Ossificans of the Neck in a Child: Case Report and Review of the Literature. *World Neurosurg*. 2019; 130: 95-97.
12. Jouve JL, Cottalorda J, Bollini G, Scheiner C, Daoud A. Myositis ossificans: report of seven cases in children. *J Pediatr Orthop B*. 1997; 6: 33-41.
13. Sferopoulos NK, Kotakidou R, Petropoulos AS. Myositis ossificans in children: a review. *Eur J Orthop Surg Traumatol*. 2017; 27: 491-502.
14. Cohen RB, Hahn GV, Tabas JA, Peeper J, Levitz CL, Sando A, et al. The natural history of heterotopic ossification in patients who have fibrodysplasia ossificans progressiva. A study of forty-four patients. *J Bone Joint Surg Am*. 1993; 75: 215-9.
15. Civan M, Bilgili F, Kilic A, Uyguner Z, Toksoy G. A case of fibrodysplasia ossificans progressiva in a 5-year-old boy with all musculoskeletal features and review of the literature. *J Orthop Case Rep*. 2018; 8: 36-39.
16. García Callejo FJ, Morant Ventura A, Orts Alborch MH, Blay Galaud L, Marco Algarra J. Head and neck lesions caused by systemic progressive ossifying fibrodysplasia (Munchmeyer's disease). *Acta Otorrinolaringol Esp*. 2000; 51: 646-54.

17. Kaplan FS, Xu M, Glaser DL, Collins F, Connor M, Kitterman J, et al. Early diagnosis of fibrodysplasia ossificans progressiva. *Pediatrics*. 2008; 121: e1295-e1300.
18. Sferopoulos NK, Kotakidou R, Petropoulos AS. Myositis ossificans in children: a review. *Eur J Orthop Surg Traumatol*. 2017; 27: 491-502.
19. Sarac S, Sennaroglu L, Hosal AS, Sozeri B. Myositis ossificans in the neck. *Eur Arch Otorhinolaryngol*. 1999; 256: 199-201.
20. Booth DW, Westers BM: The management of athletes with myositis ossificans traumatica. *Can J Sport Sci* 1989; 14: 10-6.
21. Muir B. Myositis ossificans traumatica of the deltoid ligament in a 34-year-old recreational ice hockey player with a 15-year post-trauma follow-up: a case report and review of the literature. *J Can Chiropr Assoc*. 2010; 54: 229-42.
22. Wang SY, Lomasney LM, Demos TC, Hopkinson WJ. Radiologic case study. Traumatic myositis ossificans. *Orthopedics* 1999; 22: 991-5.
23. Ogilvie-Harris DJ, Fornasier VL. Pseudomalignant myositis ossificans: heterotopic new-bone formation without a history of trauma. *J Bone Joint Surg Am*. 1980; 62: 1274-83.
24. Samuelson KM, Coleman SS. Nontraumatic myositis ossificans in healthy individuals. *JAMA*. 1976; 235: 1132-3.
25. Danchik JJ, Yochum TR, Aspegren DD. Myositis ossificans traumatica. *J Manipulative Physiol Ther* 1993; 16: 605-14.
26. Mestan MA, Bassano JM. Fractured heterotopic bone in myositis ossificans traumatica. *J Manipulative Physiol Ther*. 2001; 24: 296-9.
27. Le Roux DA. Chondrosarcoma and myositis ossificans. *J Manipulative Physiol Ther*. 1998. 21: 640-48.
28. Simmonds J, Taki N, Chilton I, Vecchiotti M. A rare case of pediatric Nontraumatic Myositis Ossificans in the posterior triangle. *International Journal of Pediatric Otorhinolaryngology*. 2016; 84: 116-118.
29. Kokkosis AA, Balsam D, Lee T. K, Z. Schreiber J. Pediatric non-traumatic myositis ossificans of the neck. *Pediatr Radiol*. 2009; 39: 409-12.