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INTRAMUSKULARNI MIKSOM NATKOLJENICE LEČEN U TERCIJARNOM CENTRU U MALOJ DRŽAVI

Authors Zoran Terzic*, Batric Vukcevic†, Marinko Paunovic*, Boban Djordjevic‡, Stojan Terzic†, Vojnosanitetski pregled (2020); Online First May, 2020.

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Authors: Zoran Terzic*, Batric Vukcevic†, Marinko Paunovic*, Boban Djordjevic‡, Stojan Terzic*
* – Center for Plastic and Reconstructive Surgery, Clinical Center Montenegro
† – Faculty of Medicine, University of Montenegro
‡ - Military Medicine Academy

Corresponding author:
Batric Vukcevic, MD
General surgery resident
Teaching Assistant in Anatomy
e-mail: batricvukcevic@gmail.com
telephone number: +382 69 799 114

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Abstract
**Background:** Myxoid lesions may present as benign, locally invasive or malignant tumors. The incidence of intramuscular myxoma is nearly 1 case per 1,000,000 population.

**Case report:** A 73-year-old man presented to our clinic with a painless, subcutaneous tumor of the adductor region of the left thigh. Computed tomography and magnetic resonance imaging showed a cystic tumor with thin septae located in the adductor muscles. The tumor was extirpated in toto, with the histopathological confirmation of an intramuscular myxoma.

**Conclusions:** This example of a successful treatment of intramuscular myxoma may serve to increase the awareness of surgeons and radiologists in small countries regarding myxoid tumors.

**Key words:** intramuscular; myxoma; magnetic resonance imaging; surgery; thigh

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**Abstrakt**

**Uvod:** Miksoidni tumori mogu biti benigni, lokalno invazivni ili maligni. Incidenca intramuskularnog miksoma je oko 1 slučaj na 1,000,000 stankovnika.

**Prikaz slučaja:** Prezentovan je slučaj 73-godišnjeg mučkarca sa bezbolnim, potkožnim tumorom aduktorne regije lijeve natkoljenice. Kompjuterizovana tomografija i magnetna rezonanca su pokazale cistični tumor sa tankim septama lociran u aduktornoj muskulaturi. Tumor je ekstirpiran u cjeloti, a patohistološki nalaz je pokazao da se radi o intramuskularnom miksomu.

**Zaključak:** Ovaj primer uspešnog lečenja intramuskularnog miksoma može služiti da skrene pažnju hirurzima i radioloških u malim zemljama kada se u pitanju miksoidni tumori.

**Ključne reči:** intramuskularni; miksom; magnetna rezonanca; natkoljenica; hirurgija

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**Introduction**

Myxoid soft-tissue tumors represent a group of neoplasms consisting of rich extracellular gelatinous mucopolysaccharide matrix actively secreted by tumor cells. They usually affect the extremities and can be benign (including the locally invasive tumors) or malignant. Among benign and locally aggressive myxomas, intramuscular myxoma is the most frequent type; while aggressive angiomyxoma, superficial
angiomyxoma, myxolipoma and dermal myxoma are less common. The incidence of intramuscular myxoma is around 1 case per 1,000,000 population. Due to the hypocellularity of the lesion, excisional biopsy is indicated (instead of fine needle aspiration cytology); while complete excision is almost always curative. We present a case of an asymptomatic intramuscular myxoma in a patient diagnosed and treated in a tertiary care center in Podgorica, Montenegro.

Case report

A 73-year-old man presented to our clinic with a tumor of the left thigh. He stated that the mass was growing slowly over the course of several years, without pain or any other symptoms. His previous medical history included: arterial hypertension; transurethral resection of the prostate for benign prostatic hyperplasia, laparoscopic cholecystectomy for chronic calculous cholecystitis; right-sided inguinal hernia repair; as well as extirpation of the right great saphenous vein due to venous varices. The laboratory results were unremarkable.

The physical examination revealed a subcutaneous tumor on the medial aspect of the superior third of the left thigh. The tumor was solid, irregularly ball-shaped and around 6-7 cm in its widest diameter. It was not painful on palpation, and there was no neurovascular deficit on the affected leg. Ultrasonography of the left thigh showed a tumor located among the adductor muscles, heterogenous in echosonographic appearance. Color Doppler imaging did not show any tumor blood vessels; and there were no pathological findings on the arterial and venous vessels of the left leg. Computed tomography (CT) showed a 95 x 90 mm tumor in the adductor region of the left thigh adjacent to the inferior ramus of the left pubic bone; resembling a cystic lesion (Fig. 1). Magnetic resonance imaging (MRI) also suggested the cystic nature of the tumor, with thin septae (Fig. 2 and 3). No bone or vascular lesions were seen on CT or MRI.

Surgery revealed an encapsulated tumor measuring 9 x 7 x 6.5 cm in size, located in the adductor muscles of the thigh, arising from the medial plane of the femoral sheath (adjacent to the adventitial layer of the femoral vein) (Fig. 4). The tumor was extirpated in toto and sent to histopathological examination. The patient's recovery was uneventful and there was no recurrence of the tumor in the next 6 months after surgery. Histopathology showed an overall regular histological and cytological appearance - a tumor consisting of myxomatous stroma, oval and spindle cells without mitoses. Immunohistochemistry was
negative for CK, S100, CD34 and actin, while it was positive for vimentin (Fig. 5). Therefore, the tumor was diagnosed as a benign myxoma.

Discussion

Intramuscular myxoma usually occurs in patients 50-60 years of age, somewhat more often in women; most commonly affecting the muscles of the thigh. The tumor is rarely located in the intermuscular planes, and more often in the muscle tissue itself. CT image is nonspecific, showing a well-defined hypodense lesion in the intramuscular space. MRI shows homogeneous, (81-100%), hypointense lesions on T1 sequence and hyperintense lesions on T2 sequence, owing to the liquid contents of the tumor; as well as perilesional rind of fat or edema.

Aggressive angiomyxomas usually occur in women, affecting the pelvis or perineum. They exhibit a swirling pattern of infiltration without visceral involvement. Myxofibrosarcoma is a malignant lesion affecting the extremities, with exual sex predilection and common local recurrence due to incomplete resection. It exhibits an infiltrative border with centrifugal spreading along fascial and vascular planes. The tumor is heterogeneous on both T1 and T2 sequences, with a T2-hyperintense curvilinear „tail sign“ projection from the primary lesion into the adjacent tissue. The „tail sign“ has moderate sensitivity (64-77%) and specificity (79-90%) for this diagnosis, and it should be differentiated from perifocal edema by the presence of contrast enhancement. Due to its heterogeneity, myxofibrosarcoma is most difficult to distinguish from myxoid liposarcoma (intralesional hemorrhage might mimic fat tissue – but fat-suppressed T1 images are of great value in these cases).

Harish et al. proposed several factors to help determine malignant from benign cyst-like lesions on MR: heterogeneity on T1 sequence, average tumor size ≥ 7 cm, with the largest tumor size ≥ 10 cm. Peterson et al. suggested that bening myxoid lesions exhibit the following characteristics: uniform low signal intensity on T1 sequence and increased signal intensity on T2 sequence, homogeneous enhancement, sufficient circumscription, and intramuscular localization. In a 2016 study on 95 myxoid tumors (26 benign and 69 malignant), Crombe et al. identified several MR characteristics of malignant lesions: ill-defined margin, hemorrhagic component, fibrosis, “tail sign”, and intra-tumoral fat. In their study, malignant lesions were misdiagnosed due to the concomitant absence of all the
The radiographic and histopathologic descriptions of the tumor presented herein implied that it is a benign intramuscular myxoma. The absence of distant metastases - as well as the lack of local recurrence after resection - confirm the nature of the tumor.

Montenegro is inhabited by roughly 600,000 people, and the aforementioned incidence of intramuscular myxoma makes it an unique case in this country. While there is a sufficient number of case reports and research articles published on benign and malignant myxoid tumors worldwide, there are not many case reports on this subject originating from Balkan countries. Therefore, we feel that the awareness of these tumors among the surgeons and the radiologists from this region should be increased.

Conflict of interest: none declared.

References


Figure legends:
Fig. 1 Axial computed tomography scan (white arrowhead indicating the tumor).
Fig. 2 Axial magnetic resonance image (white arrowhead indicating the tumor); a – T1 sequence; b – T2 sequence.
Fig. 3 Coronal magnetic resonance image (white arrowhead indicating the tumor); a – T1 sequence; b – T2 sequence.
Fig. 4 Intraoperative view of the tumor
Fig. 5 a – Uniform tumor cells without mitoses (HE, x200); b – Vimentin positivity on immunohistochemistry (x400)