

Elastofibroma in the rearfoot. Report of a rare case

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Elastofibroma in extremely rare location of rearfoot is described. The tumor occurred in 57-ys-old man with recent history of arthrosis of the 1st metatarsophalangeal joint and with history of unspecified trauma of rearfoot in childhood. The 1.2 cm lesion showed typical histological and immunohistochemically features of elastofibroma. It was paucicellular and composed of bland fibroblasts and mature appearing fat cells, with dense intercellular collagenous matrix that contained numerous elastic fibers. The fibroblasts of the lesion expressed CD34 and were negative for alpha smooth muscle actin, desmin, S100 protein and beta-catenin. The case demonstrates that the diagnosis of elastofibroma should be considered also by examination of non-periscapular lesions. From pathogenetic point of view, the tumor in present case was caused probably by chronic minor trauma, like it is supposed for common periscapular elastofibromas.

Keywords: elastofibroma, soft tissue, rearfoot, CD34, arthrosis, trauma

Elastofibróm na päte. Popis zriedkavého nálezu

V kazuistike je popísaný elastofibróm v enormne zriedkavej lokalizácii na päte. Jednalo sa o 1,5 cm-ový tumor pravej päty u 57-ročného muža, s trojročnou anamnézou artrózy prvého metatarzofalangeálneho kĺbu a s históriou úrazu päty v mladosti. Tumor bol priemeru 1,2 cm a mal typické histologické a imunohistochemické znaky elastofibrómu, s hypocelulárnou blandnou populáciou fibroblastov, zrelými adipocytmi, početnými elastickými vláknami, imunohistochemickou expresiou CD34 a negativitou na aktín, desmín a beta-katenín. Prípad ukazuje, že diagnózu elastofibrómu je potrebné zvažovať aj pri léziách mimo typickej periskapulárnej lokalizácie. Pravdepodobnou patogenézou popísaného tumoru je chronická mechanická traumatizácia tkaniva, podobne ako sa predpokladá u „konvenčných“ periskapulárnych elastofibrómov.

Kľúčové slová: elastofibróm, mäkké tkanivá, päta, artróza, trauma

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Introduction

Elastofibroma, described firstly by Jarvi and Saxen^(1,2) is a benign soft tissue lesion composed of fibroblasts which produce abundant elastic type fibers^(3,4). It occurs usually in the subscapular region in the elderly population. According original studies, it is predominant among females⁽¹⁻³⁾; but another large series of 122 cases shows mild male predominance⁽⁴⁾. Subscapular region is typical for elastofibroma (it is almost diagnostic of it), whereas other locations are very rare⁽⁴⁾. Recently, we have seen elastofibroma arising atypically in the rearfoot. To our knowledge, only one case was reported in this location before⁽⁵⁾. We would like to present our case here.

Report of the case

In a 57-ys-old man, extirpation of the right rearfoot tumor along with arthrodesis of 1st metatarsophalangeal joint were performed. The patient had arthrosis of 1st metatarsophalangeal joint and long-lasting snapping sensations and mild pain of the rearfoot especially by walking. He mentioned rearfoot trauma by intense landing after a jump in his youth, with subsequent long lasting pain. Magnetic resonance imaging of the right foot revealed 1,2 cm tumoroid lesion in the

region of the origin of the plantar band and adjacent soft tissues (**figure 1**).

Figure 1. Coronal magnetic resonance image of the foot shows tumoroid lesion (arrow) with signal density that corresponds with lesion's fibroadipous histology.



Figure 2. Elastofibroma showing typical features, such as paucicellular population of bland fibroblastic cells, abundant and deeply eosinophilic elastic fibers, and mature adipose tissue.

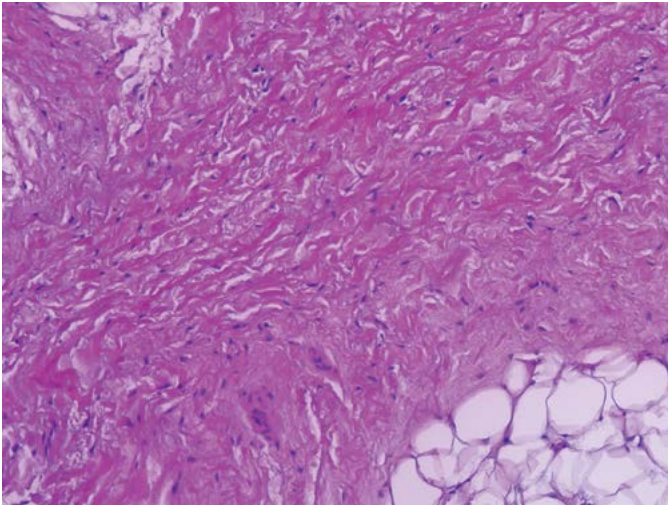


Figure 3. Elastofibroma contains numerous elastic fibers which are positive by orcein stain.

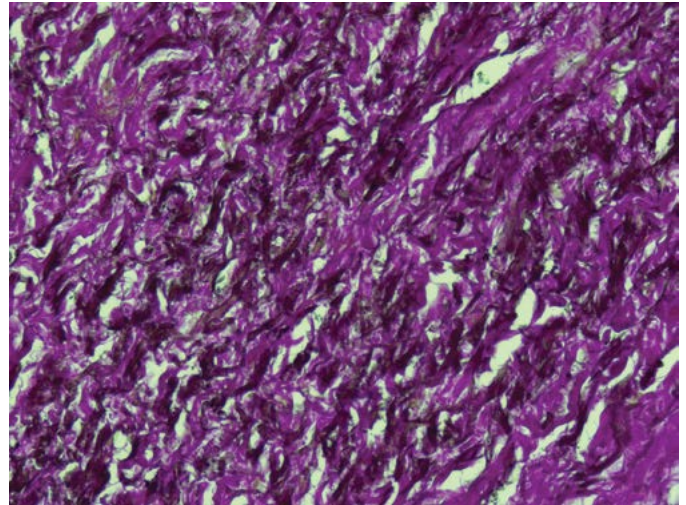


Figure 4. Immunohistochemical expression of CD34 in fibroblastic cells of elastofibroma.

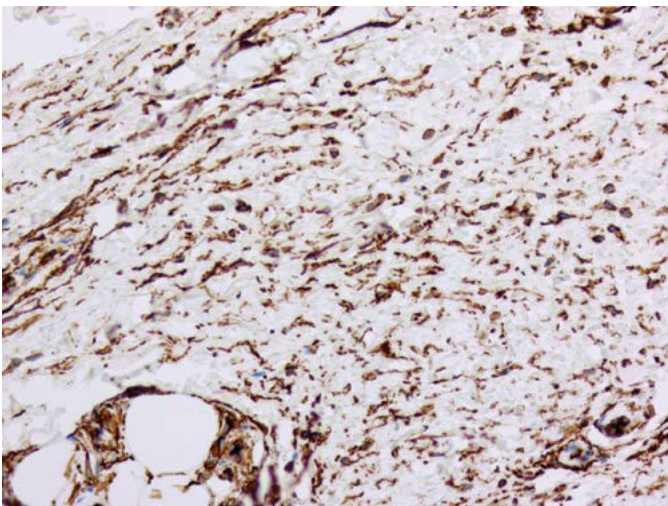
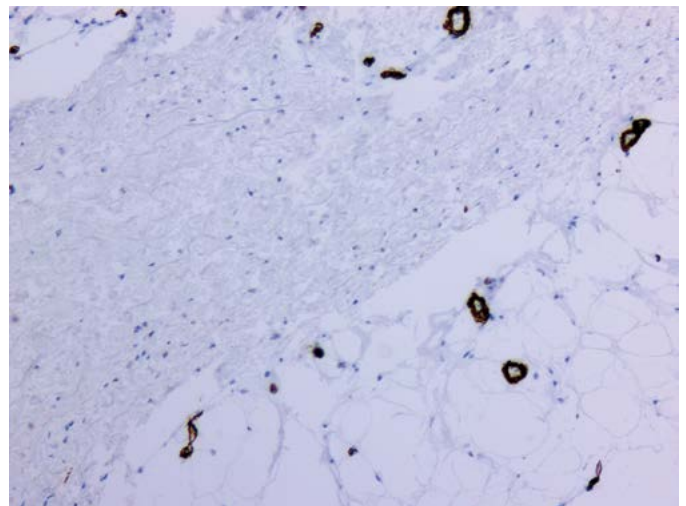


Figure 5. Alpha-smooth muscle actin is negative, with positivity limited to pericytes (serving as internal control).



Other medical history of the patient included autoimmune thyroiditis, stage 2 hypertension, celiac disease, duodenal ulcer, and sigmoid diverticulosis. Family history of the patient did not include any soft tissue lesions.

After the extirpation of the rearfoot lesion, two pieces of tissue were submitted for histologic examination. **Grossly**, the fragments were of irregular shape, measuring 1,5 cm x 1 cm x 0,5 cm and 1,5 cm x 1 cm x 0,5 cm, respectively. They were of fibrous and lipomatous appearance, without necrosis and hemorrhage. **Histologically**, the lesion was unencapsulated and it was composed of paucicellular population of bland fibroblasts, with collagenous matrix containing abundant elastic fibers (stained positively by orcein stain) (**figures 2 and 3**). Groups of adipocytes were seen between strands of mentioned fibrous tissue. Immunohistochemically, the fibroblastic cells were positive for CD34 (**figure 4**), and were negative for alpha-smooth muscle actin (**figure 5**), desmin, S100 protein and beta-catenin. Adipocytes expressed S100

protein. Based upon these findings the diagnosis of elastofibroma was rendered.

Discussion

In our case, pathologic finding was typical for elastofibroma. The lesion was paucicellular and composed of bland fibroblasts and mature appearing fat cells, with dense intercellular collagenous matrix that contained numerous elastic fibers. Immunohistochemically, the fibroblasts were positive for CD34, and negative for muscle markers such as alpha-smooth muscle actin and desmin. This phenotype is characteristic for elastofibroma^(4,6).

Interesting finding in our case represents the atypical location in the rearfoot. As mentioned above, common elastofibromas occur in subscapular area. Rare cases were reported in other regions, such as the hand⁽⁷⁾, thigh⁽⁸⁾, deltoid muscle⁽⁹⁾, gluteal region⁽¹⁰⁾, axilla⁽¹¹⁾, neck⁽¹²⁾, face⁽¹³⁾, and oral mucosa⁽¹⁴⁾, old thoracotomy scar⁽¹⁵⁾, in the shoulder intra-ar-

ticularly⁽¹⁶⁾, at the umbilicus and aortic valve⁽¹⁷⁾ and in the colon⁽¹⁸⁾. Three cases were reported in the forefoot⁽¹⁹⁻²¹⁾. In the rearfoot, only one case was described before⁽⁵⁾. Pirak et al. reported elastofibroma of the rearfoot in a 79-year-old female. Unlike our case, this patient had no history of trauma or arthrosis, and etiology of the lesion in their case appears to be unclear. In our case, the patient history includes 3 years lasting arthrosis of 1st metatarsophalangeal joint and mild pain of the rearfoot by walk. In addition, the patient stated the he had any jump-landing trauma of the rearfoot in his youth. We suppose that an evolution of elastofibroma in our case is explainable by the chronic minor traumatization of the area due to mild anatomical and functional deviation, caused by arthrosis and posttraumatic changes. This is consistent with suggested pathogenesis of common elastofibromas of periscapular region. This currently proposed pathogenesis consists of repeated mechanical trauma, elastic degeneration of collagen fibers, and contributing genetic

factors⁽¹⁻⁴⁾. It is interesting, however, that some recent studies indicate that elastofibroma can represent a true neoplasm. Elastofibromas showed chromosomal instability⁽²²⁻²⁵⁾, and analysis of human androgen receptor (HUMARA) locus has revealed nonrandom X-chromosome inactivation, supporting clonal nature of the lesion⁽⁶⁾. It is possible that chronic irritation of fibrous tissue give rise to clonal neoplastic proliferation of fibroblastic cell. For more exact knowledge of the pathogenesis, additional studies (particularly molecular genetic) are needed.

In sum, we described elastofibroma in rare location of the rearfoot. The case demonstrates that diagnosis of elastofibroma should be considered also by examination of non-periscapular lesions. In our case, the lesion showed typical histological and immunohistochemical features of this tumor. It seems that also from pathogenetic point of view the tumor in our case was caused by chronic minor trauma, like it is supposed for common periscapular elastofibromas.

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