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Histological characteristics of a giant cell tumor of the tendon sheaths associated with palmar fascial fibromatosis (case report)

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Abstract

Giant cell tumor of the tendon sheaths and palmar fascial fibromatosis are both fibrohistiocytic diseases. There is no information about their combination in the available literature. **Aim** To analyze the histological characteristics of a giant cell tumor of the tendon sheath associated with palmar fascial fibromatosis. **Materials and methods** Case history and pathomorphological examination of the surgical material of patient M with recurrent palmar fascial fibromatosis and Dupuytren's contracture of the 5th ray of the right hand in grade 3 and a neoplasm of the 2nd finger of the right hand **Results** A neoplasm with a thin fibrous capsule, permeated with blood vessels, many of them had abnormally thickened fibrous walls and completely obliterated lumens. The cellular composition of the tumor is mixed: fibroblast-like cells, small histiocytes, siderophages, osteoclast-like giant multinucleated cells, xanthoma and plasma cells. The frequency of mitoses is less than one per 10 fields of view, apoptotic bodies from zero to several per field of view. In the composition of fibromatous cords in the projection of the V beam, giant multinucleated cells (possibly cells of foreign bodies) were found. **Conclusion** For the first time, a clinical case of a giant cell tumor of the tendon sheaths, which developed against the background of a long-term recurrent palmar fascial fibromatosis, is presented. Taking into account the pathomorphological characteristics of the surgical material, the risk of its recurrence and malignancy in patient M. is low. However, the unpredictability of the histogenesis of this tumor known from the literature is an indication for active clinical observation.

Keywords: palmar fascial fibromatosis, Dupuytren's contracture, giant cell tumor of the tendon sheath, pathohistology

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INTRODUCTION

Giant cell tumors of the tendon sheaths (GCTTS) originate from the synovial cells of the tendon sheaths, the synovial membrane of the joints, and articular capsules. They can be intra- and extra-articular by location, and are localized or diffuse by the nature of extension [1]. Localized types are characteristic for the hand [2], and 80 % of the GCTTS develop on the fingers [3], more often on the index finger, less often on the thumb [4]. The female to male ratio among patients is approximately 3:2, the peak incidence occurs in the fifth decade of life [5]. According to the frequency of occurrence, GCTTS ranks second after tendon ganglia. A significant number of publications have been dedicated to this tumor [6]. The differential diagnosis of GCTTS includes more than two dozen tumors and tumor-like diseases of the hand with a similar clinical picture [7]. However, in the available literature, we did not find information about the combination of GCTTS with palmar fascial fibromatosis.

Purpose To analyze the histological characteristics of a giant cell tumor of the tendon sheath associated with palmar fascial fibromatosis.

MATERIAL AND METHODS

The surgical material (a fibrously altered fragment of the palmar aponeurosis and a neoplasm) was fixed in 10 % neutral formalin solution, dehydrated in alcohols of ascending concentrations (from 70º to absolute), impregnated with a sealing mixture, and embedded in paraffin. Histological sections 5–7 μm thick were prepared on a Riechard sledge microtome (Germany). Sections were stained with hematoxylin and eosin and Masson's trichrome method. The study of micropreparations and microphotography was carried out using an Axio-Scope A1 stereomicroscope with an AxioCam digital camera complete with Zen Blue software (Carl Zeiss MicroImaging GmbH, Germany).

RESULTS

Presentation of the clinical case Patient M., 66 years old, applied to the clinic of reconstructive plastic and hand surgery of the Ilizarov Center in November 2021 with complaints of flexion deformity of the 5th finger of the right hand and a neoplasm of the 2nd finger of the hand. According to the patient, she underwent surgery to excise the ganglion of the radial flexor of the wrist in her childhood and an operation for Dupuytren's contracture more than 30 years ago. In 2014, a relapse occurred in the form of a deformity of the fifth finger. In

2019, a neoplasm appeared on the dorsal ulnar surface of the second finger, which gradually increased in size. Physical examination revealed a subcutaneous cord on the palmar surface of the hand in the projection of the 5th ray, persistent flexion contracture of the proximal interphalangeal joint in grade III.

On the dorsal ulnar surface of the second finger in the projection of the distal interphalangeal joint, a node 1–2 cm in size was palpated, extending to the palmar surface of the finger. There were no signs of bone erosion on X-ray examination. According to the ultrasound examination, in the structure of the soft tissues of the middle phalanx of the second finger, along the medial and anteromedial surfaces, there was an isoechogenic avascular homogeneous formation with a hypoechoic rim, not adhered to the tendons, which was interpreted as a fibroma.

An operation was performed: excision of a fibrously altered palmar aponeurosis of the right hand, arthrolysis of the proximal interphalangeal joint of the fifth finger, transosseous fixation of the fifth finger with an Ilizarov mini-apparatus, excision of the neoplasm on the second finger of the right hand, neurolysis of the ulnar palmar neurovascular bundle of the second finger.

The neoplasm was located on the tendon of the deep flexor of the second finger under the neurovascular bundle, which had an atrophic overstretched appearance. The tumor had a lobed structure, yellow-brown coloring, elastic consistency, and measured 2 × 1.5 × 1 cm (Fig. 1).

In the postoperative period, smooth wound healing was achieved, complete extension of the fifth finger, and there were no sensory disturbances.

Histological examination of the pretendinous fibromatous cord of the palmar aponeurosis revealed fields of dense connective tissue characteristic of the residual stage of fibromatosis. In some areas, tendon-like fibromatous cords were straightened (Fig. 2, a), in the others they were twisted by contractions (Fig. 2, b). They were accompanied by blood vessels with thickened walls. Tendon-like strands and layers of loose connective tissue contained mainly cells of fibroblastic differon and myofibroblasts of a contractile phenotype, which at the light-optical level can be identified by characteristic cross-striated nuclei (Fig. 2, c). Macrophage-type cells were single, but there were giant multinucleated cells among them (Fig 2, d). Probably, those were cells of foreign bodies.

The neoplasm had a thin, well-defined fibrous capsule and was penetrated by numerous blood vessels (Fig. 3a), many of which had abnormally thickened fibrous walls with signs of fibrosis and hyalinosis, and some had completely obliterated lumens (Fig. 3, b, c). Collagenization of its stroma was expressed to varying degrees in different parts of the tumor.

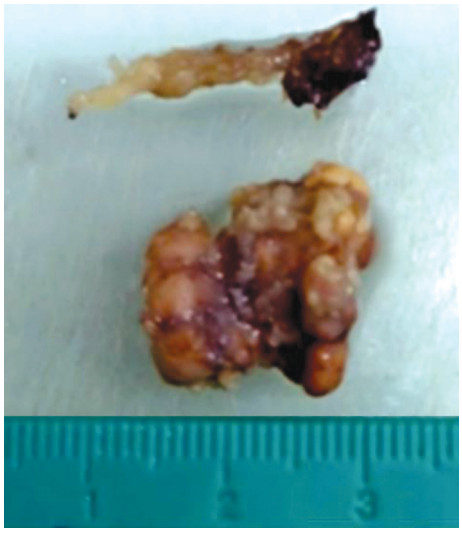

Fig. 1 Macropreparations. Upper view – pretendon cord of fibrous palmar aponeurosis of the 5th ray of the hand; bottom view – multilobular giant cell tumor of the tendon sheaths of the deep flexor of the second finger

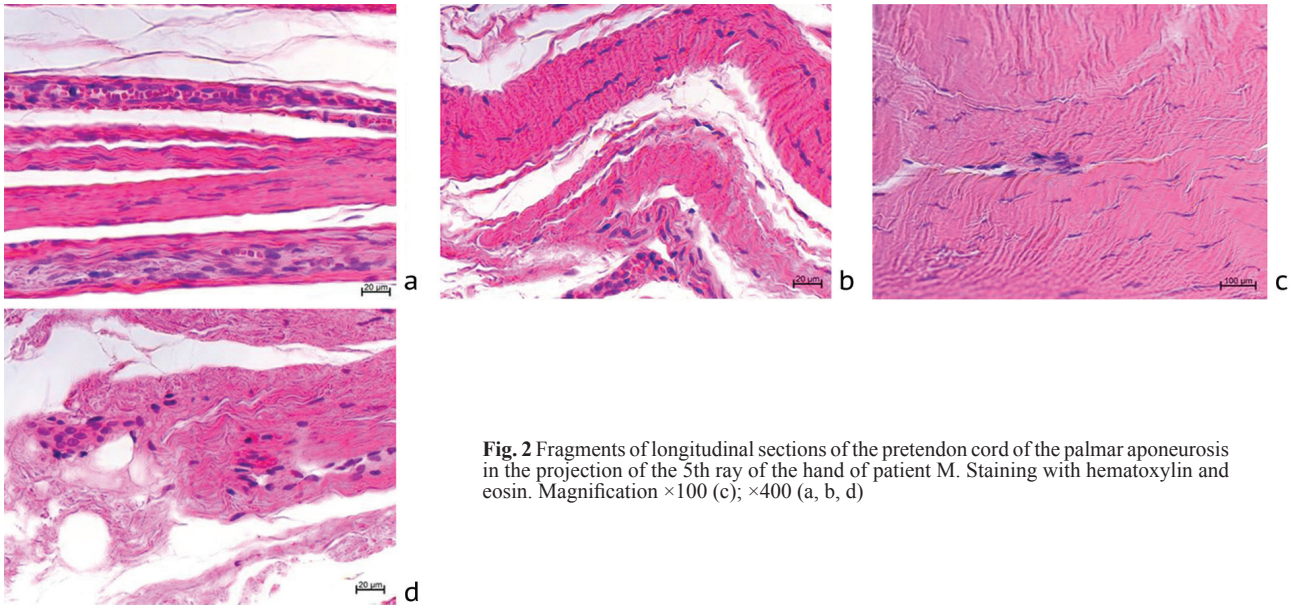

Fig. 2 Fragments of longitudinal sections of the pretendon cord of the palmar aponeurosis in the projection of the 5th ray of the hand of patient M. Staining with hematoxylin and eosin. Magnification ×100 (c); ×400 (a, b, d)

The cell composition of the tumor was mixed; along with fibroblast-like cells, it contained small histiocytes and larger mononuclear cells loaded with siderin (Fig. 3, b, d), osteoclast-like multinuclear cells (Fig. 3, d, e, f), xanthoma and plasma cells. Occasionally there were figures of mitotic division, less than one per 10 fields of view (Fig. 3, e), apoptotic bodies – from 0 to several per field of view (Fig. 3, f).

A cluster of three small bodies of Vater-Pacini and a nerve stem in a state of denervation, perineuritis, and death of perineural cells were found at the border of the tumor with the hypodermis (Fig. 4).

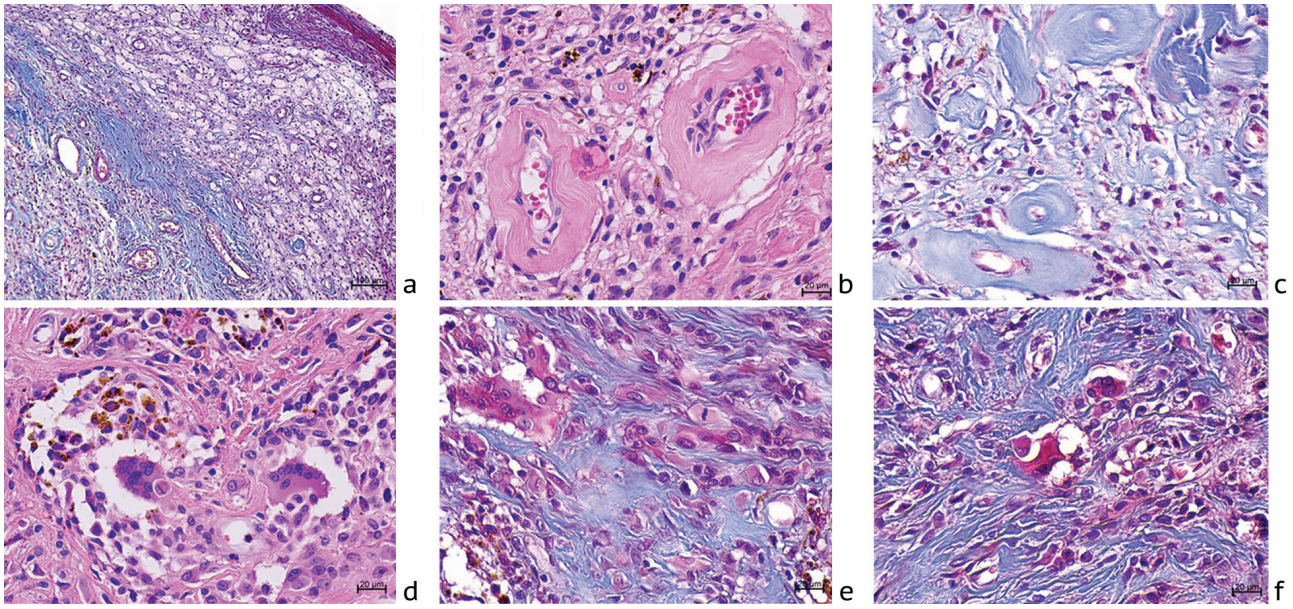

Fig. 3 Cell- and tissue composition of the neoplasm of the second finger in patient M. Staining with Masson's trichrome method (a, c, e, f), hematoxylin and eosin (b, d); magnification ×100 (a); ×400 (b, c, d, e, f)

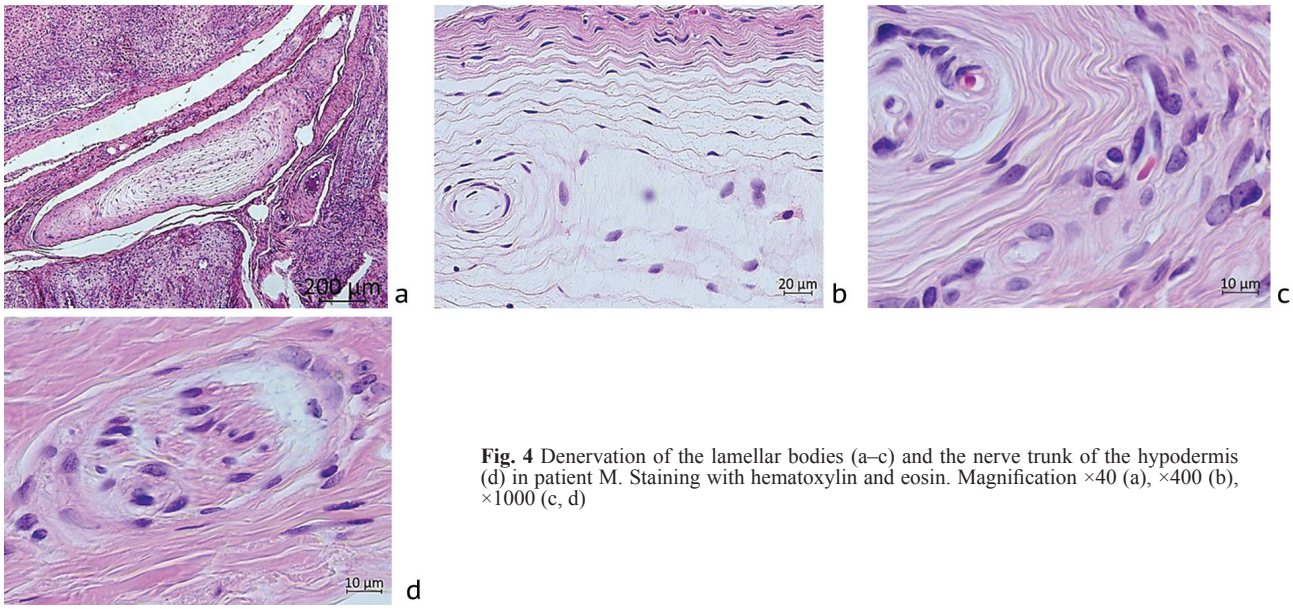

Fig. 4 Denervation of the lamellar bodies (a–c) and the nerve trunk of the hypodermis (d) in patient M. Staining with hematoxylin and eosin. Magnification ×40 (a), ×400 (b), ×1000 (c, d)

DISCUSSION

Different authors have defined GCTTS as fibrous histiocytoma, pigmented nodular tenosynovitis, tenosynovial giant cell tumor, localized nodular tenosynovitis, benign synovioma, and synovial fibrous xanthoma [2, 8, 9; 10, 11]. In the literature, for several decades, the question has been discussed whether GCTTS is a tumor or refers to tumor-like conditions. According to the consensus concept of etiopathogenesis, GCTTS, having both neoplastic and reactive features, is characterized by a landscape effect - neoplastic cells with chromosomal translocations and expression of macrophage colony-stimulating factor (CSF1) make up an insignificant part of the tumor, from 2 to 16 %, the bulk is represented by inflammatory cells, recruited by local overexpression of CSF1 [12]. In our observation, there were various macrophage-type cells,

including giant multinucleated ones. Macroscopically, the GCTTS had a characteristic appearance, a yellow dense lobulated-nodular mass, which corresponds to the descriptions available in the literature [8].

GCTTS originates from the tendon sheaths and is located superficially, but its extensions spread in the dorsal and ventral direction under various anatomical formations, including neurovascular bundles, often surrounding them, which creates technical difficulties for radical noninvasive removal [13]. According to a meta-analysis, only 4.57 % of patients (16 out of 350) with GCTTS of the fingers have sensory impairments [14]. However, there is no data on an instrumental study of specialized types of sensitivity. Our patient had no clinical signs of sensory disturbances, but histological study revealed signs of denervation of the lamellar bodies and nerve trunks of the hypodermis, possibly resulting from compression of these structures by a growing tumor. Similar histological manifestations of distal compression-ischemic neuropathy were found in all patients with palmar fascial fibromatosis in a large sample study [15].

A hand surgeon should suspect GCTTS not only in the presence of a subcutaneous neoplasm, but also in all cases of prolonged edema of the hand, since there are cases of unexpected detection of GCTTSS in acute synovitis [16].

Localized GCTTSs are regarded as benign neoplasms, but frequent recurrence is a problem: according to a meta-analysis, 14.8 % of GCTSs of fingers recurred [14]. Recurrence factors include localization in the area of the interphalangeal joints [11], degenerative joint diseases, radiological signs of bone erosion, and increased mitotic activity [17]. According to other authors, bone impressions are not associated with the risk of recurrence. Single tumors (type I) have a low risk of recurrence, multiple tumors (type II) have an increased risk, the number of mitoses in most GCTTSs is less than 2 in 10 fields of view, and this factor is not significant for recurrence [18]. Rare malignant variants were described among primary and recurrent GCTTSs. In the world literature, data on 49 patients were published in whom the tumor was characterized by locally destructive growth, distant metastases, and death [19]. Some authors refer GCTTSs to tumors with uncertain histogenesis [3], similar to giant cell tumor of bone [20].

Regardless of its location, GCTTS is characterized by a mixed cell composition: small histiocytes with rounded corrugated nuclei, larger mononuclear cells with eccentric nuclei and abundant eosinophilic cytoplasm with a hemosiderin rim, osteoclast-like giant multinucleated cells, xanthomatous macrophages, and plasma cells [19]. The ratio of cell types and the degree of stromal collagenization can be different.

In our observation, in addition to collagenization of the tumor stroma, collagenization of the thickened walls of its blood vessels was expressed, and many of them were obliterated. On the other hand, giant multinucleated cells were found in the composition of fibromatous cords in the projection of the 5th ray. Probably, these not quite usual signs are due to the combination of GCTTS with palmar fascial fibromatosis. Pronounced fibrosis of the matrix and of tumor vessels made ultrasound diagnosis difficult. At the same time, there is reason to consider GCTTS to be a transitional tumor with a possible transformation into a fibroma [21]. For the differential diagnosis of these GCTTSs and fibroma, the pathohistological characterization at the light-optical level is of decisive importance, the role of immunohistochemistry and electron microscopy is small [22].

CONCLUSION

This is the first clinical report on the giant cell tumor of tendon sheaths developed due to persistent recurrent palmar fascial fibromatosis. Judging from the pathomorphological characteristics of the surgical material, the risk of recurrence and malignancy of the tumor in patient M. is low. However, the uncertain histogenesis of GCTTS known from the literature is an indication for active clinical observation.

REFERENCES

1. Gouin F., Noailles T. Localized and diffuse forms of tenosynovial giant cell tumor (formerly giant cell tumor of the tendon sheath and pigmented villonodular synovitis). *Orthop. Traumatol. Surg. Res*., 2017, vol. 103, no. 1S, pp. S91-S97. DOI: 10.1016/j.otsr.2016.11.002.
2. Adams E.L., Yoder E.M., Kasdan M.L. Giant cell tumour of the tendon sheath: experience with 65 cases. *Eplasty*, 2012, vol. 12, pp. e50.
3. Monoghan H., Salter D.M., Al-Nafussi A. Giant cell tumour of tendon sheath (localized nodular tenosynovitis): clinopathological features of 71 cases. *J. Clin. Pathol*., 2001, vol. 54, no. 5, pp. 404-407. DOI: 10.1136/jcp.54.5.404.
4. Leneveu E., Prévot J., Dautel G., Lascombes P. Villonodular synovitis of the wrist. In regards to a case in an adolescent. *Ann. Chir. Main Memb. Super*., 1994, vol. 13, no. 4, pp. 274-277. (in French) DOI: 10.1016/s0753-9053(05)80007-9.
5. Lee K.R., Hyun C.L. Tenosynovial giant cell tumor showing severe bone erosion in the finger: case report and review of the imaging findings and their significance. *Investig. Magn. Reson. Imaging*, 2016, vol. 20, no. 2, pp. 127-131. DOI: 10.13104/imri.2016.20.2.127.
6. Lv Z., Liu J. Giant cell tumor of tendon sheath at the hand: A case report and literature review. *Ann. Med. Surg*. (Lond), 2020, vol. 58, pp. 143-146. DOI: 10.1016/j.amsu.2020.09.011.

7. Darwish F.M., Haddad W.H. Giant cell tumour of tendon sheath: experience with 52 cases. *Singapore Med J*., 2008, vol. 49, no. 11, pp. 879-882.
8. Hamdi M.F., Touati B., Zakhama A. Giant cell tumour of the flexor tendon sheath of the hand: analysis of 27 cases. *Musculosket. Sur*g., 2012, vol. 96, no. 1, pp. 29-33. DOI: 10.1007/s12306-011-0148-1.
9. Ozalp T., Yercan H., Kurt C., Ozdemir O., Coşkunol E. Giant-cell tumors of the tendon sheath involving the hand or the wrist: an analysis of 141 patients. *Acta Orthop. Traumatol. Turc*., 2004, vol. 38, no. 2, pp. 120-124. (in Turkish)
10. Messoudi A., Fnini S., Labsaili N., Ghrib S., Rafai M., Largab A. Giant cell tumors of the tendon sheath of the hand: 32 cases. *Chir. Main*, 2007, vol. 26, no. 3, pp. 165-169. (in French) DOI: 10.1016/j.main.2007.03.008.
11. Reilly K.E., Stern P.J., Dale J.A. Recurrent giant cell tumors of the tendon sheath. *J. Hand Surg. Am*., 1999, vol. 24, no. 6, pp. 1298-1302. DOI: 10.1053/jhsu.1999.1298.
12. West R.B., Rubin B.P., Miller M.A., Subramanian S., Kaygusuz G., Montgomery K., Zhu S., Marinelli R.J., De Luca A., Downs-Kelly E., Goldblum J.R., Corless C.L., Brown P.O., Gilks C.B., Nielsen T.O., Huntsman D., van de Rijn M. A landscape effect in tenosynovial giant-cell tumor from activation of CSF1 expression by a translocation in a minority of tumor cells. *Proc. Natl. Acad. Sci. USA*, 2006, vol. 103, no. 3, pp. 690-695. DOI: 10.1073/pnas.0507321103.
13. Wakely P.E. Jr., Frable W.J. Fine-needle aspiration biopsy cytology of giant-cell tumor of tendon sheath. *Am. J. Clin. Pathol*., 1994, vol. 102, no. 1, pp. 87-90. DOI: 10.1093/ajcp/102.1.87.
14. Fotiadis E., Papadopoulos A., Svarnas T., Akritopoulos P., Sachinis N.P., Chalidis B.E. Giant cell tumour of tendon sheath of the digits. A systematic review. *Hand* (N Y), 2011, vol. 6, no. 3, pp. 244-249. DOI: 10.1007/s11552-011-9341-9.
15. Shchudlo N.A., Kostin V.V. Pathogenesis of neuropathy in Dupuytren's contracture. *Genij Ortopedii*, 2019, vol. 25, no. 1, pp. 58-64. DOI: 10.18019/1028-4427-2019-25-1-58-64.
16. Kruithof E., Baeten D., Dierckxsens P., Veys E.M., De Keyser F. Tenosynovitis with acute onset: unexpected giant cell tumour of the tendon sheath. *Ann. Rheum. Dis*., 2002, vol. 61, no. 9, pp. 847. DOI: 10.1136/ard.61.9.847.
17. Bedir R., Balik M.S., Sehitoglu I., Güçer H., Yurdakul C. Giant Cell Tumour of the Tendon Sheath: Analysis of 35 Cases and their Ki-67 Proliferation Indexes. *J. Clin. Diagn. Res*., 2014, vol. 8, no. 12, pp. FC12-15. DOI: 10.7860/JCDR/2014/10553.5311.
18. Al-Qattan M.M. Giant cell tumours of tendon sheath: classification and recurrence rate. *J. Hand Surg. Br*., 2001, vol. 26, no. 1, pp. 72-75. DOI: 10.1054/jhsb.2000.0522.
19. Al-Ibraheemi A., Ahrens W.A., Fritchie K., Dong J., Oliveira A.M., Balzer B., Folpe A.L. Malignant Tenosynovial Giant Cell Tumor: The True "Synovial Sarcoma"? A Clinicopathologic, Immunohistochemical, and Molecular Cytogenetic Study of 10 Cases, Supporting Origin from Synoviocytes. *Mod. Pathol*., 2019, vol. 32, no. 2, pp. 242-251. DOI: 10.1038/s41379-018-0129-0.
20. Minasov B.Sh., Biktasheva E.M., Valeev M.M., Yakupov R.R., Minasov T.B., Mavlyutov T.R. Circular resection for giant-cell tumor of the ulna combined with vascularized fibilar fragment autoplasty. *Genij Ortopedii*, 2019, T. 25, No 3, pp. 388-392. DOI: 10.18019/1028-4427-2019-25-3-388-392.
21. Satti M.B. Tendon sheath tumours: a pathological study of the relationship between giant cell tumour and fibroma of tendon sheath. *Histopathology*, 1992, vol. 20, no. 3, pp. 213-220. DOI: 10.1111/j.1365-2559.1992.tb00959.x.
22. Batista K., de Moura H., Lima M.I., Abe K.T. Fibroma and giant-cell tumor of tendon sheath: a case report. *Orthop. Res. Rev*., 2014, vol. 6, pp. 41-46. DOI: 10.2147/ORR.S46531.

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