

Case Report

Giant cell tumor of the tendon sheath composed largely of epithelioid histiocytes

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Abstract: Giant cell tumor of the tendon sheath (GCTTS) is a relatively uncommon lesion. GCTTS composed largely epithelioid histiocytes are very rare. In the literature, the author could not find such cases. A 73-year-old man presented with a mass of right thumb, and resection of the mass was performed. Grossly, the mass was encapsulated and yellowish, and measured 1.5 x 2 x 2 cm. Microscopically, the mass was composed of cellular and hypocellular zones. The former was composed of spindle cells and osteoclast-like giant cells, while the latter of epithelioid clear histiocytes. The area of the former was 20%, and the latter 80%. Pigment was seen in the former elements. Mitotic figures were seen in 3/per 30 high power fields (HPFs) in the former element and 2/per 30 HPFs in the latter element. Histochemically, the pigment was hemosiderin positive with Prussian blue staining. Immunohistochemically, both the elements were negative for cytokeratin (CK) CE1/3, CK CAM5.2, CEA, HMB45, alpha-smooth muscle antigen, p53, CD10, TTF-1, and CDX2. Both the elements were positive for CD68 and Ki-67 (cellular element 30% and hypocellular element 20%). The histiocytes of the hypocellular element and osteoclast-like giant cell of the cellular element were positive for CD45. S100-protein positive Langerhans cells and CD45-positive lymphocytes were scattered. The pathological diagnosis was GCTTS. In the author's experience, GCTTS composed largely epithelioid histiocytes are very rare. In the literature, the author could not find such cases. Thus, the author reports herein this case.

Keywords: Giant cell tumor, tendon, sheath, epithelioid, histiocytes

Introduction

Giant cell tumor of the tendon sheath (GCTTS) is a relatively uncommon lesion. GCTTS affects mainly fingers of female [1]. GCTTS is classified into localized and diffuse types [1]. The localized GCTTS is a circumscribed proliferation of synovial-like mononuclear cells, accompanied by a variable numbers of multinucleate osteoclast-like cells form cells, siderophages and inflammatory cells, most commonly occurring in the digits [1]. In my experience, GCTTS composed largely epithelioid histiocytes are very rare. In the literature, the author could not find such cases.

Case report

A 73-year-old man presented with a mass of right thumb, and resection of the mass was performed. At the operation, the mass was attached to the tendon sheath. Grossly, the mass was encapsulated and yellowish, and measured 1.5 x 2 x 2 cm. Microscopically, the mass was

composed of cellular and hypocellular zones (**Figure 1**). The former was composed of spindle cells and osteoclast-like giant cells (**Figure 2**),

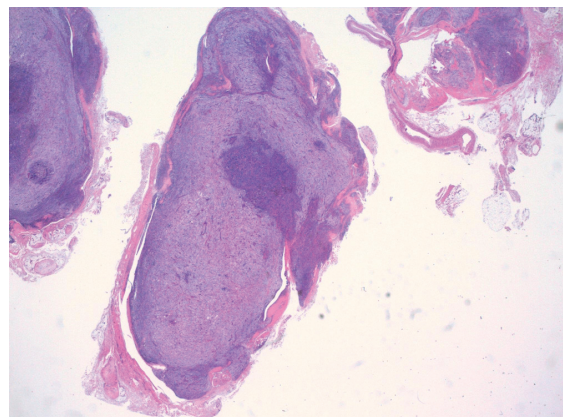


Figure 1. Very low power view of the thumb tumor. The tumor is encapsulated, and is composed of cellular (center) and hypocellular (periphery) zones. The former consists of spindle cells and osteoclast-like giant cells and the latter of epithelioid histiocytes. HE, x5

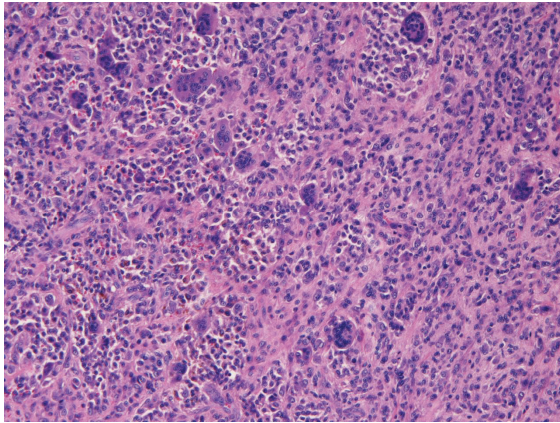


Figure 2. High power view of the cellular zone. It consists of spindle cells and osteoclast-like giant cells. Hemosiderin pigment is also seen. HE, x200

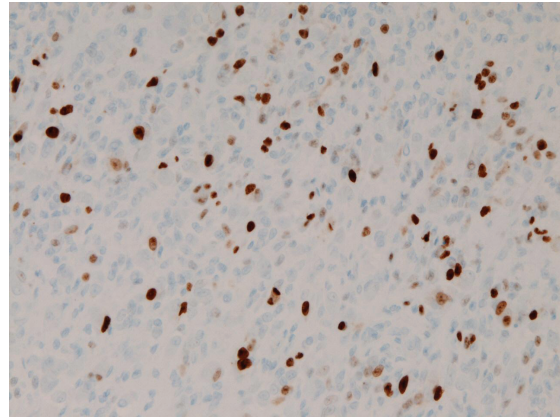


Figure 5. Ki-67 immunostaining of the peripheral zone. The Ki-67 labeling is relatively high. Immunostaining, x200

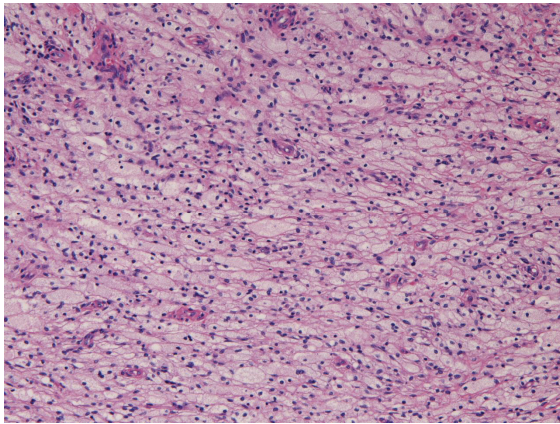


Figure 3. High power view of the hypocellular zone. It consists of epithelioid clear histiocytes. HE, x200

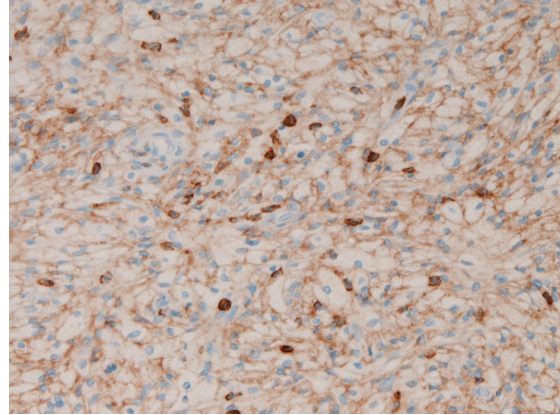


Figure 6. CD45 immunostaining of the peripheral zone. The histiocytes are positive for CD45. CD45-positive lymphocytes are scattered. Immunostaining, x200

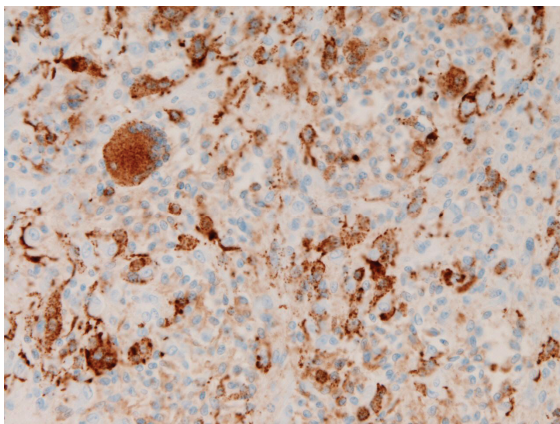


Figure 4. CD68 immunostaining of the cellular zone. The osteoclast-like giant cells and some spindle cells are positive for CD68. Immunostaining, x200

while the latter of epithelioid clear histiocytes (**Figure 3**). The area of the former was 20%, and the latter 80% (**Figure 1**). Pigment was seen in the former elements. Mitotic figures were seen in 3/per 30 high power fields (HPFs) in the former element and 2/per 30 HPFs in the latter element. Histochemically, the pigment was hemosiderin positive with Prussian blue staining. An immunohistochemical study was performed with the use of Dako's envision method, as previously described [2, 3]. Immunohistochemically, both the elements were negative for cytokeratin (CK) CE1/3, CK CAM5.2, CEA, HMB45, alpha-smooth muscle antigen, p53, CD10, TTF-1, and CDX2. Both the elements were positive for CD68 (cellular element, moderate; hypocel-

lular element, mild) (**Figure 4**) and Ki-67 (cellular element 30% and hypocellular element 20%) (**Figure 5**). The histiocytes of the hypocellular element and osteoclast-like giant cell of the cellular element were positive for CD45 (**Figure 6**). S100-protein positive Langerhans cells and CD45-positive lymphocytes were scattered.

Discussion

The author firstly thinks that the tumor is GCTTS, but immunohistochemical study was performed because of the unusual histology. Metastatic melanoma, clear cell sarcoma, metastatic renal cell carcinoma, metastatic carcinoma, primary carcinoma, leiomyomatous tumors, neurogenic tumors were excluded by negative CK, CEA, HMB45, alpha-smooth muscle antigen, p53, CD10, TTF-1 and CDX2. Thus, the present tumor appears GCTTS composed largely of epithelioid histiocytes. In the author's experience, GCTTS composed largely epithelioid histiocytes are very rare. In the literature, the author could not find such cases. Thus, the author reports herein this case.

Positive expression of the tumor cells for CD68 indicates that the present tumor is a histiocytic tumor. The relatively high Ki-67 labeling (cellular element, 30%, hypocellular element 20%) indicate rapid proliferation and that the histiocytes of the hypocellular zone are neoplastic. Negative p53 expression suggests benign nature of the present tumor. GCTTS was initially regarded as inflammatory process [1] and one X-inactivation study suggested polyclonarity [4]. However, recent evidence suggested that GCTTS is monoclonal and neoplastic [5, 6]. Chromosomal abnormalities are found in GCTTS [1]. In the present study, the epithelioid histiocytes predominated and showed relatively high Ki-67 labeling. These findings suggest that the epithelioid histiocytes in the current case are neoplastic cells.

In conclusion, the author reported a very rare case of GCTTS composed largely epithelioid histiocytes.

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