Giant Cell Tumor of Tendon Sheath

— A case Report —

Jang Oh Kim, M.D., Tae Hyung Kim, M.D.,
Hyuk Jin Kweon, M.D., Sang Won Kim, M.D.

Department of Dermatology, Taegu Catholic University School of Medicine,
Taegu, Korea

We report a case of giant cell tumor of the tendon sheath in a 36-year-old male, who presented an asymptomatic, firm, 1.0 × 0.8 × 0.4cm-sized nodule, involving the volar aspect of the distal phalanx of the right index finger about 1 year ago. Histopathologically, the enucleated lesion showed four lobules surrounded by thin connective tissue, each of which demonstrated the variable cellularity and the polymorphic cell population consisting of foam cells, spindle-shaped fibroblasts, histiocytelike cells and multinucleated giant cells with heavy hemosiderin deposits near the periphery, in the collagenous stroma.

He has done well with no recurrence during one year of follow-up since the operation.

Key Words : Giant cell tumor of tendon sheath, Index finger

Giant cell tumor of tendon sheath (GCTTS), first introduced by Chassaingac (1850)\(^1\), is the second most common nonepithelial tumor of the fingers and hands, and far less frequently involves the ankles, feet, wrists or elbows\(^2\). GCTTS has a strong association with degenerative joint disease or rheumatoid arthritis and shows occasionally bony erosions and cyst adjacent to the lesion in the minority of cases\(^3\). It appears relatively common to surgical services, but has been rarely described in the dermatologic literature. Korean reported cases are currently four including one in orthopedic literature\(^4\) and three in the dermatologic literature\(^5\).

We presented an additional case of GCTTS occurring on the finger and reviewed the literature.

REPORT OF A CASE

A 36-year-old male patient presented to the dermatology with a painless, deep-seated firm nodule involving the volar aspect of distal phalanx of the right index finger. One day he noticed a bean-sized movable nodule covered with the overlying normal skin around 1 year ago (Fig. 1). It tended not to increase in size. His occupation was engineering handling machinery. There had been repeated antecedent traumas to the involved finger, but he had no history of rheumatoid arthritis. A recent history of allergy was not noted. Family history was non-contributory. On physical examination, no specific findings were demonstrated except the phalangeal lesion. Laboratory studies, including complete blood cell count, erythrocyte sedimentation rate, liver function test, urinalysis, and VDRL test, were all within normal limits or negative except blood eosinophilia (840/mm\(^3\)). Rheumatoid factor was negative. He refused the radiologic examination. An initial clinical diagnosis was not made.

The enucleated lesion was a firm, yellowish 1.0 × 0.8 × 0.4cm-sized, subcutaneous mass. Histopatho-
logic examination revealed four well-defined lobules surrounded by thin connective tissue, each of which showed variable cellularity in the stroma (Fig. 2 A,B). Proliferation of spindle-shaped fibroblasts, together with numerous multinucleated giant cells and foam cells, were seen randomly in the hyalinized stroma of the hypocellular area (Fig. 3 A,B). In the hypercellular area, round or polygonal histiocyte-like cells were arranged in sheets between collagen bundles, with large number of foam cells and heavy hemosiderin deposits near the periphery with Prussian blue stain (Fig. 4 A,B,C). The chronic inflammatory cells were minimally infiltrated. No mitotic activity was noted. On immunohistochemical staining with 3-amino-9-ethylcarbazole as substrate for CEA, S-100 protein, desmin and vimentin, using peroxidase anti-peroxidase (PAP) technique, the tumor cells showed weakly cytoplasmic-positive reaction only for vimentin (Fig. 5). The diagnosis of GCTTS was made on the basis of the histologic findings.

No evidence of recurrence was noted during one year of follow-up since the operation.

**DISCUSSION**

GCTTS is known by a variety of entities, including fibrous histiocytoma of tendon sheath, xanthogranuloma, nodular tendosynovitis, myeloxanthoma, benign synovioma and pigmented nodular synovitis. Some regard it as a localized form of pigmented villous synovitis of articular, tendinous
Fig. 4. The findings of hypercellular area.
A: Round or polygonal histiocyte-like cells were densely arranged (H & E, × 200).
B: Foam cells abounded near the periphery (H & E, × 200).
C: Hemosiderin deposits were most commonly seen near the periphery (Prussian blue stain, × 20).

Fig. 5. Positive staining with vimentin was observed in tumor cells (PAP Stain, × 400).

and peritendinous synovial tissue\textsuperscript{15} or a variant of histiocytoma\textsuperscript{12}. Such entities could be associated with the intricate etiopathologic mechanism.

There have been several hypotheses regarding the pathogenesis of the tumor. Suggested origin and differentiation include a reactive inflammatory process to repeated trauma and regenerative hyperplasia, infection, neoplastic process or disturbance in lipid metabolism\textsuperscript{5,12-16}. Among these, at this time, a reactive inflammatory process is more favored\textsuperscript{15}. However, a rare report of malignant form arising in the vicinity of the tendon and with no spontaneous regression represents a true neoplastic process\textsuperscript{11,17}. The exact nature of this tumor still remains uncertain\textsuperscript{12,15,18}.

Most lesions show one or more discrete nodules not attached to the overlying skin, presenting themselves as well-defined, somewhat lobulated and slowly growing masses with firm to rubbery consistency\textsuperscript{5,13-14}. The majority of lesions involve the digits of the hands\textsuperscript{14}, which are the index finger, thumb, and middle finger in descending frequency\textsuperscript{14}. They are mainly reported in the volar aspects of the fingers with no clear localization as to distal or proximal.

<table>
<thead>
<tr>
<th>Case</th>
<th>Sex/Age</th>
<th>Size</th>
<th>Site</th>
<th>Duration</th>
<th>Sign</th>
<th>Authors</th>
</tr>
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<tbody>
<tr>
<td>1</td>
<td>M / 23</td>
<td>Pea-sized</td>
<td>Rt. 4th finger</td>
<td>3yr</td>
<td>Tender nodule</td>
<td>Bun et al (1976)\textsuperscript{9}</td>
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<tr>
<td>2</td>
<td>F / 42</td>
<td>Pea-sized</td>
<td>Rt. 3rd finger</td>
<td>3yr</td>
<td>–</td>
<td>Ahn et al (1990)\textsuperscript{7}</td>
</tr>
<tr>
<td>3</td>
<td>F / 36</td>
<td>1 × 2 cm</td>
<td>Lt. 5th finger</td>
<td>2yr</td>
<td>–</td>
<td>Yi et al (1993)\textsuperscript{8}</td>
</tr>
<tr>
<td>4</td>
<td>F / 45</td>
<td>1 × 1 cm</td>
<td>Lt. 3rd finger</td>
<td>7mo</td>
<td>–</td>
<td>Jun et al (1994)\textsuperscript{9}</td>
</tr>
<tr>
<td>5</td>
<td>M / 36</td>
<td>Bean-sized</td>
<td>Rt. 2nd finger</td>
<td>1yr</td>
<td>–</td>
<td>Kim et al (1994)</td>
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proximal sites. The lesions mostly develop a single occurrence in number, albeit one case with multiple lesions has been shown by Ushijima. The tumors are usually asymptomatic. A small minority may produce pain, digital numbness or loss of motion around the joints. There is a female to male preponderance of 3:2. The tenderness or dull pain occurs in 20% of cases. The duration of lesions ranges from weeks to more than 20 years, with a mean of 2-3 years. They are relatively small in size, ranging from 0.8 to 3.0 cm in diameter with an average of 1.1 cm. In the 5 reported Korean cases including the present case, the age ranged between 23-45 years, with an average of 36.4 years. The ratio of female to male was 3:2. The size of lesions was around a pea to a peanut or 1.0 to 2.0 cm in diameter. All fingers but the thumb, were evenly involved. The duration was about 7 months to 3 years with a mean of 2 years. All cases developed in a single digit, and one case complained of occasional tenderness. The demographic results in the reported Korean cases were in the same category as those reported by several authors. There were no abnormal laboratory findings in all 4 reported cases. However, the present case showed only blood eosinophilia, which should be evaluated in relation to its cause.

An enucleated tumor may be encapsulated, yellow to orange colored, and subdivided into lobules surrounded by thin connective tissue. Microscopically, the cellularity within the tumor varies. Mitoses are focally observed in 20% of cases. The tumors consist of a polymorphic cell population made up of several types of tumor cells including histiocyte-like, fibroblast-like or intermediate, xanthomatosus and multinucleated giant cells. Foam cells are present most often near the periphery of the lesion. The predominant cell type varies with the area, but generally it is a histiocyte-like cell. On electron microscopic study, these cells are all derived from type A (macrophage-like) and type B (fibrocyte-like) cells which are normally present in the human synovial membrane, and they share common functional markers of histiocytes. In this case, the tumor had four lobules. Each lobule had a variable cellularity in the stroma. The majority of cells were histiocyte-like cells, often containing hemosiderin or lipid. In the hypocellular areas, spindle-shaped fibroblasts and foam cells were seen in a fibrous or hyalinized stroma. The multinucleated giant cells, resembling normal osteoclasts, were scattered numerously. The round or polygonal histiocyte-like cells were densely present in the hypercellular areas, with large number of foam cells and heavy hemosiderin deposits near the periphery of it. The hemosiderin deposits might have originated from posttraumatic bleeding. Immunohistochemical staining in tumor cells demonstrated positive only for vimentin, presumably suggestive of the mesenchymal cell origin.

The differential diagnosis is extensive and includes foreign body granuloma, ganglion cyst, epidermal cyst, digital mucous cyst, lipoma, rheumatoid nodule, dermatofibroma, fibroxanthosarcoma, epithelioid sarcoma, embryonal rhabdomyosarcoma, fibroma of tendon sheath, necrobiosis granuloma, xanthoma tendinosum, and acquired fibrokeratoma. The correct clinical diagnosis is made in only 20-30% before the surgery. Histologically, there are enough variabilities to cause occasional confusion with benign, as well as malignant, lesions. GCTTS should be differentiated by its fairly characteristic histopathologic appearances. It is important for both clinical dermatologist and pathologist to be familiar with this lesion, especially if occurring in the fingers.

The optimal treatment is careful and total surgical excision. Radiotherapy is ineffective. After the excision, between 17-48% of lesions recurred. The lobulated nature of the lesions, incorrect initial diagnosis, frequent occult extension into surrounding tissues, and incomplete excision contributed to its high recurrence rate. It is seen that GCTTS requires precise diagnosis and total excision. In the present case histologic diagnosis, coupled with the treatment of surgical enucleation, was made, and no relapse was noted during one year of follow-up.

REFERENCES