Inflammatory pseudotumors of the paratesticular area are rare, and are often reported in the literature by various terms, e.g., proliferative funiculitis, inflammatory myofibroblastic tumor, pseudosarcomatous myofibroblastic proliferation and fibrous pseudotumor. This is one of the most common lesions of that region, and typically presents as a longstanding, painless scrotal mass. Here, we describe a 34 year-old man who has had a palpable scrotal mass for the past 10 years. The excised mass was composed of multiple conglomerated nodules, which had homogeneous rubbery cut surfaces. Histologically, each was a well circumscribed, but unencapsulated mass of hyalinized collagenous tissue interspersed with lymphoplasmacytic cells and lymphoid follicle formation. A small fraction of paucicellular spindle cells was positive for vimentin, smooth muscle actin and CD68. Ultrastructurally, abundant collagen fibrils were mixed with paucicellular spindle cells and inflammatory cells. These spindle cells had abundant rough endoplasmic reticula and myofilaments with focal densities, indicating myofibroblastic differentiation.

Key Words: Inflammatory pseudotumor-Paratesticular
Paratesticular Inflammatory Pseudotumor

1:100 dilution, Fig. 3A) and CD68 (Zymed, 1:50 dilution), but not desmin (D33; Zymed, 1:50 dilution), cytokeratin (AE1/AE3; Zymed, 1:50 dilution), Ki-67 (polyclonal, Dako, 1:250 dilution) or ALK-1 (p80; DAKO, 1:40 dilution). The histological and immunohistochemical results are summarized in Table 1. Electron microscopy revealed abundant extracellular collagen fibrils, with rarely-found spindle cells (Fig. 3). The spindle cells had thin peripherally-located myofilaments, and abundant rough endoplasmic reticula with focal densities and were found to be mixed with inflammatory cells including plasma cells and mast cells.

DISCUSSION

Variable terms such as inflammatory pseudotumor, inflammatory myofibroblastic tumor, proliferative funiculitis (pseudosarcomatous myofibroblastic proliferation of spermatic cord) and fibrous pseudotumor (periorchitis) have been used to describe lesions like the reported lesion. Currently, the term inflammatory myofibroblastic tumor has been generally accepted, encompassing all such lesions. This is essentially a tumor which exhibits cellular, fascicular fibroblastic/myofibroblastic proliferations, accompanied by a prominent infiltrate of chronic inflammatory cells, particularly plasma cells and mast cells. The spindle cell component typically has plump, variably atypical nuclei. The mitotic rate is variable. Immunohistochemically, histiocytes or bizarre spindle cells are generally smooth muscle actin-positive, and overexpress ALK-1 protein (p80), which stains positively in 40% of inflammatory myofibroblastic tumors.

The present case is considered to be located at the extreme pole of more cellular inflammatory pseudotumors, i.e. typical histology of inflammatory myofibroblastic tumor. Because the present case showed sparse cellularity, we think that more cellular cases of inflammatory myofibroblastic tumors should be distinguished from inflammatory leiomyosarcoma, and even malignant lymphoma. Cigar-shaped, centrally-located blunt nuclei are regarded as a characteristic of leiomyosarcoma, but this is frequently obscured by infiltrating inflammatory cells, especially in inflammatory leiomyosarcoma, which leads to the misdiagnosis of inflammatory pseudotumors, as the above malignant tumors. In such circumstances, immunohistochemistry is often helpful in ascertaining the nature of the spindle cells.

![Fig. 1. The external surface of the mass shows gray tan multinodular appearance.](image)

**Table 1. Review of the previously reported Korean cases including the present case**

<table>
<thead>
<tr>
<th>Author &amp; year</th>
<th>Cases</th>
<th>Age (yr)</th>
<th>Presenting symptom</th>
<th>Symptom duration</th>
<th>Location</th>
<th>Gross finding</th>
<th>Calcification on H-E</th>
<th>Immunohistochemistry</th>
<th>Electron microscopy</th>
</tr>
</thead>
<tbody>
<tr>
<td>Yim, et al. (1994)</td>
<td>1</td>
<td>59</td>
<td>Slow growing, nontender mass</td>
<td>Incidentally found mass</td>
<td>Testicular tunics, proximal spermatic cord</td>
<td>Two separate nodules</td>
<td>Absent</td>
<td>+: vimentin, actin</td>
<td>Not done</td>
</tr>
<tr>
<td>Paik, et al. (1995)</td>
<td>2</td>
<td>28</td>
<td>Slow growing, nontender mass</td>
<td>6 months</td>
<td>Testicular tunics</td>
<td>Multinodular appearance</td>
<td>Absent</td>
<td>+: vimentin, actin, S-100 protein (weak)</td>
<td>Not done</td>
</tr>
<tr>
<td></td>
<td>3</td>
<td>30</td>
<td>Slow growing, nontender mass</td>
<td>5-6 yr</td>
<td>Testicular tumics</td>
<td>Multinodular appearance</td>
<td>Present</td>
<td>+: vimentin, actin</td>
<td>Not done</td>
</tr>
<tr>
<td>Yoo, et al. (2000)</td>
<td>4</td>
<td>52</td>
<td>Slow growing, nontender mass</td>
<td>10 yr</td>
<td>Testicular tumics</td>
<td>Multinodular appearance</td>
<td>Absent</td>
<td>+: vimentin - desmin, S-100 protein (weak)</td>
<td>Not done</td>
</tr>
<tr>
<td></td>
<td>5</td>
<td>38</td>
<td>Slow growing, nontender mass</td>
<td>14 months</td>
<td>Testicular tumics</td>
<td>Multinodular appearance</td>
<td>Absent</td>
<td>+: vimentin - desmin, S-100 protein (weak)</td>
<td>Not done</td>
</tr>
<tr>
<td>Kim, et al.* (2004)</td>
<td>6</td>
<td>32</td>
<td>Slow growing, nontender mass</td>
<td>10 yr</td>
<td>Peripectidymal area</td>
<td>Multinodular appearance</td>
<td>Absent</td>
<td>+: vimentin, actin - desmin, S-100</td>
<td>Fibroblasts (stage III), abundant Collagen fibrils</td>
</tr>
</tbody>
</table>

yr, years; +, positive immunoreactivity; -, negative immunoreactivity; H-E, hematoxylin and eosin stain; *, the present case.
for correct diagnosis. In most inflammatory leiomyosarcomas, the muscle markers such as smooth muscle actin or desmin, are positive in the fascicles, whereas ALK-1 (p80) does not stain in the spindle cells. However, the present case differs in that the spindle cells in the sparsely cellular area showed immunonegativity for smooth muscle actin and ALK-1, but minimal desmin immunoreactivity. Ultrastructurally, the presence of the fibronexus junction is critical for diagnosing myofibroblastic differentiation in inflammatory myofibroblastic tumor including fibrous pseudotumors. The fibronexus junction is a cell-to-matrix specialization composed of myofilaments, and fibronectin fibrils.

It is a shelf-like region of the cell surface, where convergence of intracellular smooth muscle myofilaments and extracellular fibronectin filaments are present. It is regarded as an important, but not entirely specific, ultrastructural marker for fully differentiated stage 4 myofibroblasts. The present case showed abundant rough endoplasmic reticula, and peripheral myofilaments, indicating stage 3 fibroblasts (“myoid” fibroblasts). This observation supports the conclusion that the extremely fibrous inflammatory pseudotumor is located at the extreme pole of inflammatory myofibroblastic tumors.

After reviewing the previous reported Korean cases of parater-

![Image](image_url)
Paratesticular Inflammatory Pseudotumor

all the cases typically presented with a slowly growing, non-tender lesion, occurring first in middle to late adulthood. The mean age was 40 years, and the mean duration of the symptomatic period was 4.6 years. All the cases yielded good outcomes without recurrences. These clinicopathologic findings are summarized in Table 1.

In summary, the authors emphasize slowly growing inflammatory pseudotumor in the paratesticular area, which seems to be located at the extreme pole of the more cellular type of inflammatory myofibroblastic tumors.

REFERENCES