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LETTER TO THE EDITOR

A case of spermatic cord cyst with nodular histiocytic/mesothelial hyperplasia

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Dear Editor,

Nodular histiocytic/mesothelial hyperplasia (NHMH) is a rare and benign tumor-like lesion and easily misdiagnosed clinically and pathologically. We report a case of right spermatic cord cyst with NHMH.

A 4-year-old patient presented with a right spermatic cord cyst, which developed 4 years ago. He had no history of trauma, inflammation, urinary tract infection, or surgery. Transillumination of scrotum was positive, revealing a cyst measuring about 3 cm \times 2 cm \times 3 cm. Excision of the cyst was performed under general anesthesia. Macroscopically, a mural nodule with a diameter of 6 mm was found in the cyst. Histologically, a solid-appearing cell cluster was detected in the mural nodule. Morphology included relatively single, cohesive polygonal or oval cells, arranged in solid sheets or nests, with ovoid or deeply grooved nuclei and a moderate amount of pale pink cytoplasm in the nodular area. The nuclei had delicate chromatin and no obvious atypia and mitosis (Figure 1). Immunohistochemically, the histiocytic cells were strongly positive for the histiocytic marker vimentin, CD68 (Figure 2), and negative for S-100, cytokeratin, calretinin, pan-cytokeratin (pan-CK), desmin, and smooth muscle actin (SMA). The pathological diagnosis was spermatic cord cyst with NHMH.

The concept of NHMH was first presented by Chan *et al.*¹ in 1997. Their study demonstrated nodular lesions composed of mostly diffuse CD68-positive cells admixed with a few mesothelial cells. We searched for NHMH at PubMed using the following keywords: nodular histiocytic/mesothelial hyperplasia, nodular histiocytic hyperplasia, and nodular mesothelial hyperplasia. A total of less than fifty cases of NHMH were found in different parts of the body, including lung, pleura, peritoneum, endometrium, pericardium, omentum majus, bladder, inguinal hernia, and spermatic cord cyst.²⁻⁵

The etiology of NHMH is still unknown. Studies suggest that it may be a reactive lesion caused by inflammation, trauma, or tumors. Suarez-Vilela and Lzquierdo-Garcia suggested that NHMH was a process mediated by adhesion molecules and their respective ligands, after mesothelial injury and cytokine stimulation. §

Morphology of NHMH resembles tumor growth. NHMH is predominantly composed of histiocytes with scattered mesothelial cells. 9,10 Histiocytic proliferation may be confused with primary

mesothelial lesions or neoplasms such as granulosa cell tumor, eosinophilic granuloma, chronic myelogenous leukemia, and carcinoma.8

NHMH is easily misdiagnosed as malignancy when cells of the mural nodule exhibit atypia and active mitosis, which may lead to overtreatment in clinical practice. Choi and Song suggested that cytological examination should be combined with

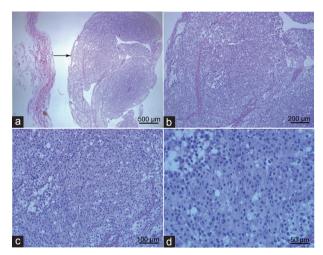


Figure 1: (a) The nodular appearance of the lesion is displayed. The arrow shows nodular area. Scale bar = 500 μ m. (b) The lesion area presents nodular and lobulated. Scale bar = 200 μ m. (c) The cell morphology is relatively single, cohesive polygonal or oval cells. Scale bar = 100 μ m. (d) The aggregates of cells show abundant cytoplasm and ovoid or twisted nuclei. Scale bar = 50 μ m.

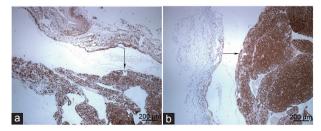


Figure 2: (a) Vimentin(+), the nodular clusters (arrow) of lesional cells are positive for vimentin by immunohistochemistry. (b) CD68(+), the nodular clusters (arrow) of lesional cells are positive for CD68 by immunohistochemistry. Scale bars = $200 \mu m$.

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immunohistochemistry to facilitate accurate diagnosis and avoid invasive procedures or unnecessary therapies.¹⁰

The purpose of this article is to delineate the clinicopathologic features of NHMH and spread awareness of the benign nature of the disease, to prevent a diagnosis of malignancy and associated radical management.

AUTHOR CONTRIBUTIONS

HJC and JZ took care of the patient and collected clinical information; HJC drafted the manuscript; and DHL performed the pathology. All authors read and approved the final manuscript.

COMPETING INTERESTS

The authors declare no competing interests.

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