

Inflammatory Pseudotumor of Lymph Node – A Case Report

Hye Ra Jung¹, M.D., Sun Young Kwon¹, M.D., Mi Sun Choe¹, M.D.,
Yu Na Kang¹, M.D., Sang Pyo Kim¹, M.D., Kun Young Kwon¹, M.D.,
Sang Sook Lee¹, M.D., Chang Ki Yeo², M.D.

*Department of Pathology, Keimyung University School of Medicine¹,
Department of Otorhinolaryngology, Keimyung University School of Medicine²,
Daegu, Korea*

Abstract : Inflammatory pseudotumor of lymph node is a rare disease with a chronic course. It is usually spontaneously recovered. Perinodal soft tissues are involved, thus clinically malignant condition is suspected. We report a case of inflammatory pseudotumor of cervical lymph node occurred in a 37-years old woman. We performed light microscopy, and immunohistochemical staining. Histologically, the component cells are mainly spindle shaped and form bundles and whorls, often giving rise to a storiform pattern. Variable inflammatory cells and Reed–Sternberg like cells are noted. The spindle cells show strong reactivity for vimentin and CD68. They do not express CD3, CD20, and CD21. Inflammatory pseudotumor of lymph node is a rare spindle cell lesion and sometimes is confused as other malignant conditions.

Key Words : Inflammatory pseudotumor, lymph node, neck

Introduction

Inflammatory pseudotumor of lymph node is a rare benign disease. Perinodal soft tissues are usually involved and malignant condition is clinically suspected. Differential

diagnosis of inflammatory pseudotumor from other infectious or malignant condition is important because it usually shows spontaneous remission. We herein report a case of inflammatory pseudotumor of cervical lymph node.

Case

A 37-years old woman visited our hospital due to a palpable mass at left neck for three weeks. On physical examination, focal edema or tenderness was absent. On computed tomography (CT), there were multiple enlarged heterogeneously enhancing lymph nodes with perinodal infiltrations in the left level II, III, IV, VA, and VB (Fig. 1). Fine needle aspiration of the mass was done after 1 week. Variablesized lymphoid cells and histiocytes were noted on the smears. The possibility of reactive hyperplasia was suggested. Since clinically malignancy was suspected based on radiologic findings, excision biopsy was performed. On operation, the mass was adhered to surrounding fat tissue and strap muscles. Microscopically, capsular and subcapsular fibrosis was severe and extended to hilar region and trabeculae (Fig. 2). Proliferation of spindle cells and infiltrates of various inflammatory cells among spindle cells were noted. Several fibrotic nodules with whorling pattern were found

(Fig. 3). Immunohistochemical stains for CD3, CD20, and CD21 were negative in spindle cells (Fig. 4). Nested PCR for *Mycobacterium tuberculosis* was negative. PAS, GMS, AFB, and Warthin starry stains showed no specific findings. The lesion was diagnosed as inflammatory pseudotumor. After removal, size of the remaining lymph nodes was decreased. Six months later, the left level II lymph nodes were increased in size again. Fine needle aspiration cytology result was performed. The smears showed fibrovascular tissue and lymphocytes without definite malignancy. The possibility of inflammatory pseudotumor could not be excluded. Close follow-up was recommended to her. Until now she is well.

Discussion

Inflammatory pseudotumor is a benign reactive disease and rarely involves lymph nodes. Inflammatory pseudotumor arising from the lymph node was firstly reported in

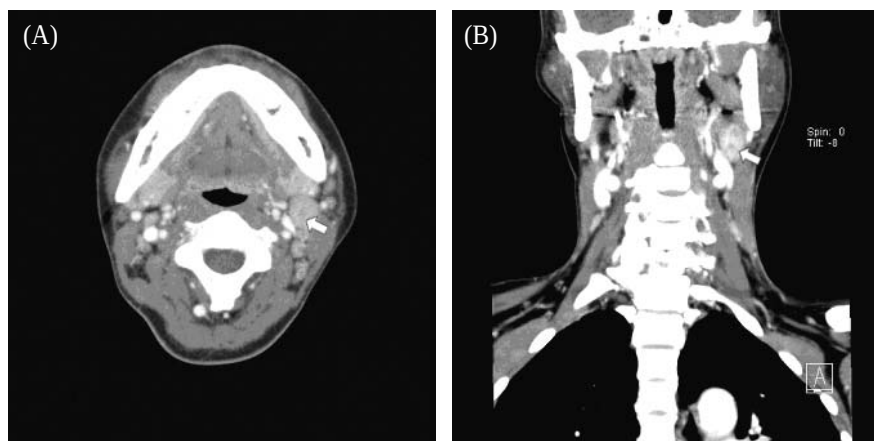


Fig. 1. Axial (A) and coronal (B) computed tomography show multiple enlarged heterogeneously enhancing lymph nodes with perinodal infiltrations (arrow) at the left level II, III, IV, VA, and VB.

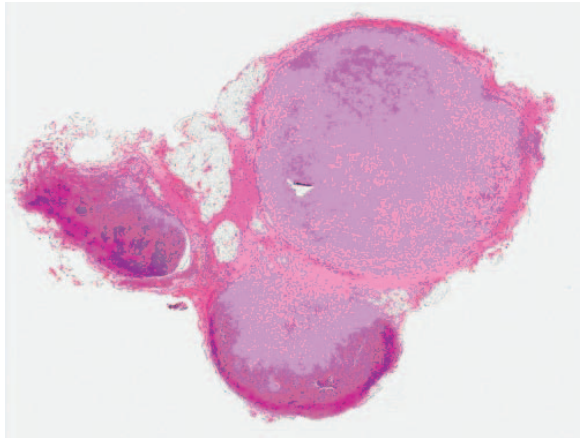


Fig. 2. Low power microscopic finding of the specimen shows marked fibrosis involving capsule, subcapsular area and trabecules (Hematoxylin & eosin stain, $\times 3$).

1988 [1]. Microscopic findings are surprisingly characteristic. At low magnification, a proliferation of small blood vessels associated with spindle cells and inflammatory cells is present. The process

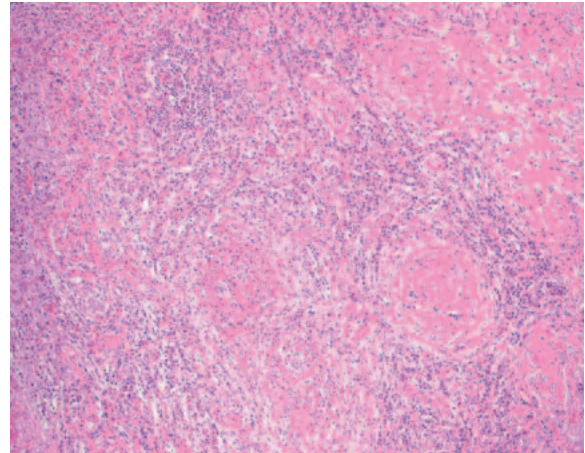


Fig. 3. Light microscopy of the lymph nodes show that fibrotic nodules with whorling pattern and storiform pattern of spindle cells. Variable inflammatory cells are admixed with spindle cells which are arranged in whorling pattern (Hematoxylin & eosin stain, $\times 100$).

involves the capsule, trabeculae, and hilum of the lymph nodes, and frequently forms

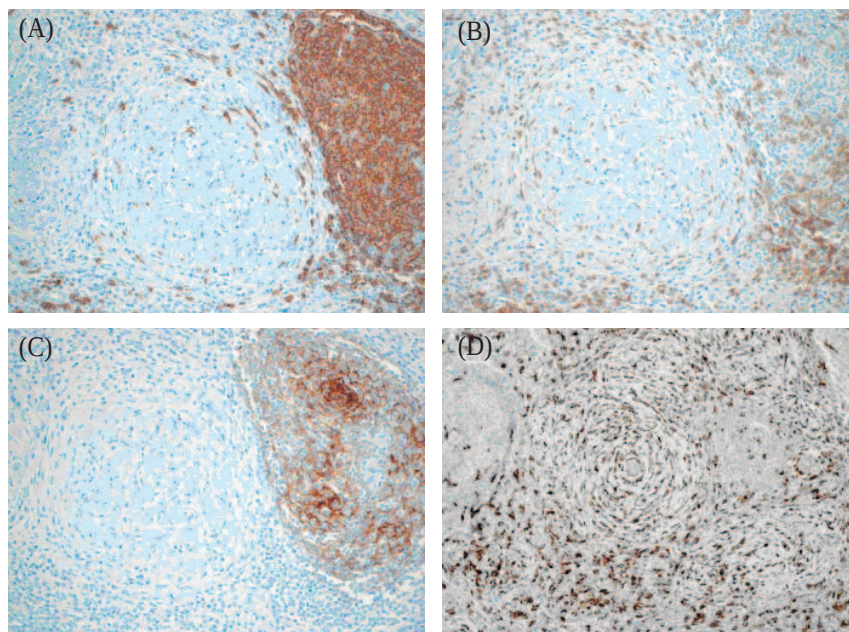


Fig. 4. Immunohistounemical stains show that the fibrotic nodule with whorling pattern is negative for CD20 ($\times 200$) (A), CD3 ($\times 200$) (B), and CD21 ($\times 200$) (C). The nodule is positive for CD68 ($\times 200$) (D).

storiform pattern. Because this fibrotic lesion extends into the perinodal fat tissue, the lymph node adheres to surrounding structures and clinically malignant conditions are suspected. Moran *et al.* reported a histopathologic spectrum of changes and divided into three stages [2]. Although the microscopic features are characteristic, there are many diseases to be differentiated, such as malignant lymphoma, dendritic and interdigitating reticulum cell sarcomas, tuberculosis, Kikuchi's disease, and malignant fibrous histiocytoma [3–5]. Because the inflammatory pseudotumor of lymph node is often spontaneously remitted, differentiation this lesion from other infectious or malignant disease is very important and excision of the lymph node is essential for diagnosis.

According to Kojima *et al.* [5], about half of the patients had localized disease, but the remaining half had systemic disease. Perrone *et al.* [1] reported the patients presented with fever and laboratory abnormalities such as elevated erythrocyte sedimentation rate, anemia, or hypergammaglobulinemia. The etiology of inflammatory pseudotumor is not well studied. Epstein–Barr virus may have a role in splenic or hepatic inflammatory pseudotumor, but it is not likely that EBV is involved in the pathogenesis of inflammatory pseudotumor in lymph node [6]. Most of clinical and pathologic features can be related to the production of interleukin–1 [1]. Resolution occurred after various treatments including surgical resection, antibiotics, steroid therapy, combination chemotherapy for malignant lymphoma, radiotherapy, and spontaneously. Relapse was reported in 15% of the cases [5].

In conclusion, the inflammatory pseudotumor

of lymph node is a benign disease which could be confused with malignant lymphoma or other infectious disease. Excision of the lymph node is mandatory for accurate diagnosis. Also, the lesion can relapse and regular follow up is required.

Summary

The inflammatory pseudotumor of lymph node is a rare benign disease. Because of perinodal involvement, malignant lymphoma is clinically suspected. Histologic findings of the lesion are very characteristic. So, excision biopsy is essential to diagnose these lesion. Although, most of the cases were cured after excision, recurrence was reported like as present case. Therefore, regular follow up is required.

References

1. Perrone T, De Wolf-Peeters C, Frizzera G. Inflammatory pseudotumor of lymph nodes. A distinctive pattern of nodal reaction. *Am J Surg Pathol* 1988;**12**:351-61.
2. Moran CA, Suster S, Abbondanzo SL. Inflammatory pseudotumor of lymph nodes: a study of 25 cases with emphasis on morphological heterogeneity. *Hum Pathol* 1997;**28**:332-8.
3. Lombardi S, Olivieri O, Morelli L, Corrocher R. Systemic inflammatory pseudotumor, an unusual cause of fever of unknown origin mimicking a malignant lymphomatous process: case-report and review of the literature. *Haematologica* 2000;**85**:539-43.
4. Davis RE, Warnke RA, Dorfman RF. Inflammatory pseudotumor of lymph nodes. Additional observations

- and evidence for an inflammatory etiology. *Am J Surg Pathol* 1991;**15**:744-56.
5. Kojima M, Nakamura S, Shimizu K, Hosomura Y, Ohno Y, Itoh H, *et al.* Inflammatory pseudotumor of lymph nodes: clinicopathologic and immunohistological study of 11 Japanese cases. *Int J Surg Pathol* 2001;**9**:207-14.
6. Arber DA, Kamel OW, van de Rijn M, Davis RE, Medeiros LJ, Jaffe ES, *et al.* Frequent presence of the Epstein-Barr virus in inflammatory pseudotumor. *Hum Pathol* 1995;**26**:1093-8.