

# Cervical Castleman's Disease Associated with Nodular Goiter: A Case Report

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## ABSTRACT

**Background:** Castleman's disease is a rare disorder of the lymphoid system characterized by noncancerous growths that may develop in lymph node tissues throughout the body. It is most common in the neck, mediastinum, and abdominal lymph nodes.

**Case Report:** Here we describe a 40-year-old woman with asymptomatic cervical Castleman's disease in the left thyroid gland, who was found to have nodular goiter and underwent therapeutic surgery to remove the mass.

**Conclusion:** After the occurrence of this case, our clinician should not only master the diagnosis of common diseases, but also have an understanding of the clinical characteristics of rare diseases.

**KEYWORDS:** Castleman disease; Nodular goiter; Lymph node hyperplasia

## INTRODUCTION

Castleman first reported the appearance of giant lymph node hyperplasia in the mediastinum in 1956. Since then, many synonyms have been linked to Castleman disease, including angiofollicular lymph node hyperplasia, giant lymph node hyperplasia, lymph node hamartoma, angiomatous lymphoid hyperplasia, and benign giant lymphoma. The disease can be found in the neck (42%), mediastinum (31%) and abdomen (23%) [1]. The exact etiology remains unclear, but theories include reactive lymphoid hyperplasia, hamartomatous change, benign lymphoid tumor, and inflammatory or infectious reactions of the lymph nodes.

## CASE REPORT

A 40-year-old woman was referred to our department for discovered two mass in her neck accidentally before 1 year ago. She did not have any symptoms. For 1 year, above package of blocks increased gradually, the right flank increased to the walnut size which the quality of material changes not obviously, and the left side increased to the egg size which the quality of material stiffens. On admission to the hospital, physical examination revealed a 3X3cm size softy mass which the boundary was clear and may along with swallow reciprocated while did not have the tenderness in the right flank, but the left side felt a 6X4cm sized

firm mass which the boundary was clear and along with swallows reciprocates unsatisfactory while does not have the tenderness either. And her laboratory data such as virus serology (HHV-8 or HIV), tumour markers, cytokines (IL-6, TNF), TSH and Calcitonin were within normal limits. Computed tomography (CT) scan of the neck showed abnormal nodules in bilateral lobes of the thyroid with clear boundaries and intact membranes. Enhancement showed no enhancement in either bilateral nodules. CT scan of the chest and abdomen showed negative. Ultrasound of the neck showed two-sided thyroid gland tumor and the hollow needle biopsy showed lymphocytic thyroiditis and fiber-hyperplasia (Figure 1). PET/CT was not performed because the patient refused. Our initial diagnosis was nodular goiter. Since the diagnosis was not clear, we decided to perform an operation. The surgery investigates in the thyroid gland right leaf to have a size approximately the 3X3cm nature strong cystic mass, the surface was smooth, can active, the thyroid gland tissue dividing line was clear, the surroundings had not touch the tumescent lymph node. The pathology report of the excision mass showed "the nodular goiter". Subsequently, the left lobe gland was found to be compressed with little change, and a 6X4X3cm firm, propellable mass was seen at the left thyroid gland outflow boundary. The tumor destroys the carotid sheath and enters between the carotid artery and the internal jugular vein. Fortunately, the lesion

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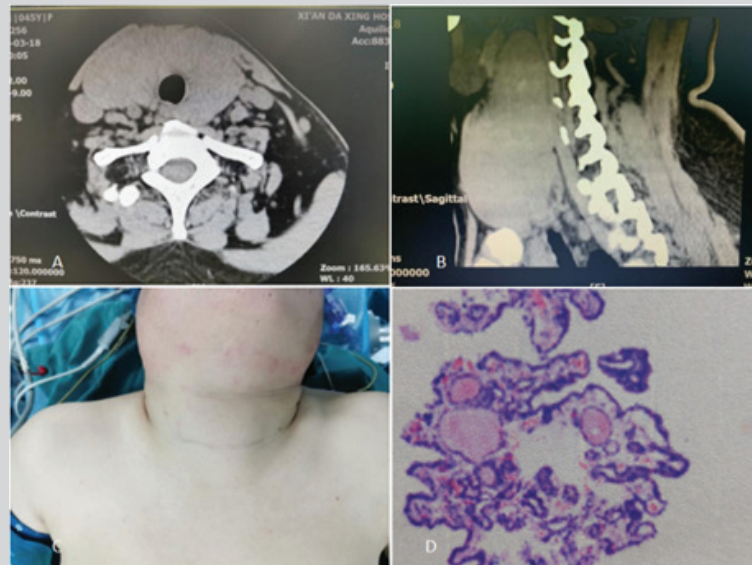
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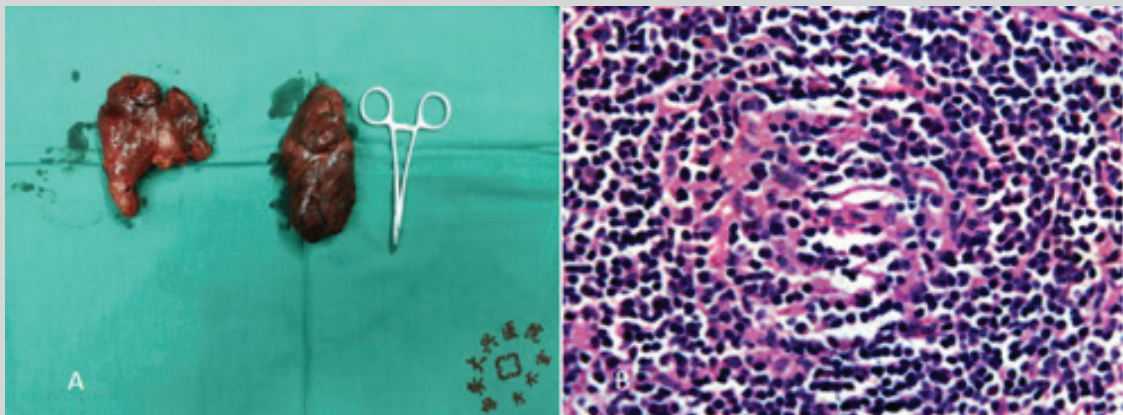
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had an almost intact capsule. Great efforts were made to divide the adhesion between the lesion and the surrounding structure. At the same time, the recurrent laryngeal nerves on both sides of the neck were well explored and protected. The lesion was finally cored out. Pathologic examination showed “the Castleman lymph node swell

(the hyaline vascular type)” (Figure 2). Continued exploration found no enlarged lymph nodes around the thyroid gland. So, we ended the operation. After surgery, we tested the patient for viral serology, tumor markers, and cytokines, but found no positive results, and the patient was discharged as scheduled.



**Figure 1:** Patient case data preoperatively. A: The preoperative CT transversal image showed a well-defined bilateral thyroid gland with significant enlargement; B: Preoperative CT atypia showed a well-defined, markedly enlarged mass at the left side of the thyroid gland; C: Degree III thyroid enlargement was seen on the patient's neck; D: The hollow needle biopsy showed a large number of thyroid follicular cells, and lymphocytic thyroiditis and fibrous hyperplasia were preliminarily considered.



**Figure 2:** Patient case data after surgery. A: The patient's right thyroid gland and left neck mass were excised; B: Postoperative pathology revealed a germinal center of different sizes in the mass. The center was hyaline degenerative, surrounded by small proliferative lymphocytes, with an onion-like skin appearance, and marked vascular hyperplasia between nodular lymphatic vessels.

## DISCUSSION

Castleman's disease is called the blood vessel follicle lymph node proliferation sickness, is one kind of lymph proliferative disease, in 1956 described by Benjamin Castleman. This kind of sickness is quite rare, now Castleman's disease is classified histologically as two types: the hyaline vascular type and the plasma-cell type [2,3]. The case we reported should belong to the clear vascular type. Postoperative pathology revealed multiple germinal centers of varying sizes in the mass. The center is hyaline, surrounded by small proliferative lymphocytes with an onion-like skin appearance and marked vascular hyperplasia between nodular

lymphatic vessels. Most (90%) cases of Castleman disease are of the hyaline vascular type, which are characterized by the presence of small hyaline follicles with interfollicular capillary proliferation [3]. The hyaline vascular type of disease is typically asymptomatic, clinically manifests as a slow-growth mass, and presents as isolated lesions found on routine chest radiographs, just as in this case. The plasma-cell type is characterized by the presence of large follicles with interposed sheets of plasma cells. These tumors, in contrast to the hyaline vascular type, typically present with systemic symptoms of fever and night sweats. The consensus opinion is that the plasma-cell variety represents a more aggressive form of the disease. It was reported that the plasma-cell type is frequently

accompanied by immunodeficiency, infection, Kaposi's sarcoma (13%), non-Hodgkin lymphoma (18%), glomeruloid hemangioma, plasmacytoma, carcinomas of the colon, kidney, and thyroid, mixed connective tissue disease, polyneuropathy, organomegaly, endocrinopathy, monoclonal gammopathy, and skin changes, with the latter five items also known as POEMS syndrome [4,5]. In recent years a multicentric (generalized) type has been shown to exist as generalized lymphadenopathy with the morphologic features of giant lymph node hyperplasia.

This type affects many areas of the body. The symptoms may be similar to those of the plasma-cell type and may include hepatosplenomegaly. Individuals with multicentric Castleman's disease tend to be older (50 to 70 years of age). Considerable interest is currently being focused on the role of human herpesvirus-8 in this lesion [6,7] and lies between element with the white blood cell - 6 (IL-6) to increase related [8,9]. The multicentric form of the disease is quite virulent compared with the typically benign course of classic Castleman's disease. Mortality has been reported to be as high as 50%, most commonly due to overwhelming sepsis.

Diagnosis of Castleman's disease is difficult because of its rare incidence and nonspecific clinical features and results of radiological examination, and definite diagnosis depends on tissue histology. In this case, we gave the patient an initial diagnosis of nodular goiter, but the result at surgery was different from the initial preoperative diagnosis. Our preoperative diagnosis was misdiagnosed because the mass on the left side of the patient's neck was too large and compressed the normal thyroid gland. Although a needle biopsy was performed before surgery, such errors were still not avoided. To avoid misdiagnosis, Castleman's disease should be included in the preoperative differential diagnosis. The computed tomography may reveal a well-defined, localized mass with striking contrast enhancement that is usually not seen in lymphoma or thymoma. Scintigraphy may not be able to differentiate Castleman's disease from malignant tumors, but it is important in excluding the presence of multicentric disease.

Localized Castleman's disease requires surgery, and complete surgical excision allows full recovery in almost all cases [9,10]. When resecting a Castleman tumor of the hyaline vascular type, great care must be taken because of its extreme vascularity. Resection of this variety is often associated with significant intraoperative blood loss. Adjunctive radiotherapy could be an option in patients at high risk of recurrence or unsuitable for surgery [11]. Corticosteroid and anti-neoplastic chemotherapy are suitable for the multicentric type, although research has not identified a standard or best treatment because of its extremely rare incidence. Moreover, Beck, et al [12] reported that treats multicentric type Castleman's disease with the anti-IL-6 immune body, demonstrated the good effect, including general symptom vanishing, the laboratory target restores, to reduce the histology shows the nodulus lymphaticus proliferation, the blood vessel proliferation, the cytoplasmic cell proliferation and so on.

## CONCLUSION

Castleman's disease has no specific clinical manifestations, and in some cases may transform into malignant lymphoma, etc. Many patients cannot benefit from surgery. After hormone therapy, chemotherapy and radiotherapy, only a small number of patients achieve remission, and generally the prognosis is poor. Therefore, the clinician must be familiar with this disease, lymph node pathology and immunohistochemical examination, may be

helpful in the diagnosis and treatment of this disease. In this case, the patient's clinical presentation lacked specificity, and the final diagnosis had to rely on pathology. The patient's lymphatic spread was located in the left thyroid position, the disease had a low incidence, and a number of factors usually associated before led to the patient's technical diagnosis being wrong in the substantive terms. Fortunately, no enlarged lymph nodes were found elsewhere in the patient and there were no clinical symptoms, so the patient had a good prognosis. After the occurrence of this case, our clinician should not only master the diagnosis of common diseases, but also have an understanding of the clinical characteristics of rare diseases. In addition, our clinicians must make a clear diagnosis before surgery, especially adequate imaging examination and laboratory examination before surgery, in order to reduce the risk of surgery.

## CONSENT

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

## AUTHORS CONTRIBUTION

Jinmao Li and Gang Li conceived of the study and participated in paper writing; Zhenzhen Guo and Ning Mao participate in the surgery; Gang Li participated in its design, discussion and coordination. All authors read and approved the final manuscript.

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