

Axillary Castleman disease

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ABSTRACT

The Castleman Disease (CD) is a rare benign lymphoproliferative disease. This disease typically presents with lymphadenopathy in a particular region, and its underlying causes are not completely understood. The most prevalent sites of occurrence are the mediastinum, neck, abdomen, and retroperitoneum, with only a small proportion (2%) of instances documented in the axillary area. This case report discusses a 60-year-old female patient with unicentric axillary CD in light of the current literature. During the physical examination, a mobile, painless, well-defined, and soft mass was found in the left axilla. The mass was around 8×6 cm. The patient's axillary ultrasonography revealed a 6.5 x 3 cm hypoechoic, heterogeneous solid lesion with prominent central vascularity in the left axillary tail. The patient's serological testing for HBC, HIV, and HCV were all negative. The patient was scheduled to have a diagnostic excisional biopsy. The histological study revealed the presence of hyaline vascular variant Castleman lymphadenopathy. CD is a rare benign condition characterized by abnormal growth of lymphatic tissue. It is most commonly found in the mediastinum and least commonly in the axilla. The diagnosis is established through the integration of clinical, radiographic, and pathological evidence. It is frequently discovered as an incidental finding while making a differential diagnosis of long-term lymphadenopathy. The treatment approach differs based on the specific histologic subtype, the extent of lymph node involvement, and any associated comorbidities. The hyaline vascular subtype's prognosis is more favorable than the plasma cell subtype. Surgical excision is regarded as a therapeutic intervention for individuals who have a unicentric disease and do not have additional complications. In this case report, CD was discovered incidentally during the surgical removal of axillary lymph nodes, which was done for differential diagnosis.

Keywords: Castleman, lymphoproliferative, lymphadenopathy, axilla

INTRODUCTION

Castleman Disease (CD) was initially described by Benjamin Castleman in 1956 [1]. The CD is a rare benign lymphoproliferative disease. This disease typically presents with lymphadenopathy in a specific location, and its exact cause is not completely understood [2]. It is frequently observed in the early years of life, and the proportion of males to females is equal [3]. The most frequent sites of occurrence are the mediastinum, neck, abdomen, and retroperitoneum, with just 2% of cases documented in the axillary area [4]. As a result, the CD that is specifically located in the axilla is much more uncommon. While the exact cause is not fully understood, studies have indicated that the development of this condition may be attributed to the clonal proliferation of tumor stromal cells and the acquired gene mutations in individuals with unicentric localization. According to reports, multicentric instances arise from immunological dysfunction and an excessive rise in cytokines caused by factors such as IL-6, human herpes virus-8 (HHV-8), and human immunodeficiency virus (HIV) [1].

This case report discusses a 60-year-old female patient with unicentric axillary CD in the light of the literature.

CASE REPORT

This case report discusses a 60-year-old female patient who was identified with CD after undergoing an excisional lymph node biopsy to diagnose axillary lymphadenopathy (LAP). The patient's medical record indicated the presence of swelling in the left axillary region for approximately six months, along with intermittent episodes of high fever and chills. On physical examination, a mobile, painless, well-circumscribed, and soft LAP measuring approximately 8×6 cm was observed in the left axillary region. The patient's axillary ultrasound showed a solid lesion in the left axillary tail measuring 6.5 x 3 cm. The lesion appeared hypoechoic and heterogeneous, with a prominent blood supply in its center. The patient's serological testing for hepatitis B and C viruses (HBC and HCV), as well as human immunodeficiency virus (HIV), were all negative. The USG-guided thick needle biopsy on the left axillary region



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did not yield a histological diagnosis. An excisional biopsy was scheduled for the patient to obtain a tissue sample for diagnosis. Preoperative laboratory tests were normal. The patient underwent axillary lymph node excision under general anesthesia. During the intraoperative investigation, a single lymph node measuring 8×5 cm was identified and surgically removed while ensuring the preservation of the capsule's integrity. Figure 1 displays the image of the surgical specimen.



Figure 1. Image of the surgical specimen

In the histological investigation, the specimen was 7.5×6×2.5 cm and had a cream-brown color. It was soft and had a uniform appearance with a creamy white surface when examined in cross-section. The presence of hyaline vascular variant Castleman lymphadenopathy was noted during microscopic examination. Immunohistochemically, the markers CD10, CD5, CD20, CD23, Bcl-2, Ki67, IgM, IgD, and Kappa+Lambda revealed negative results, while CD3 and Bcl-6 revealed positive results. The histopathological image was shown in Figure 2.

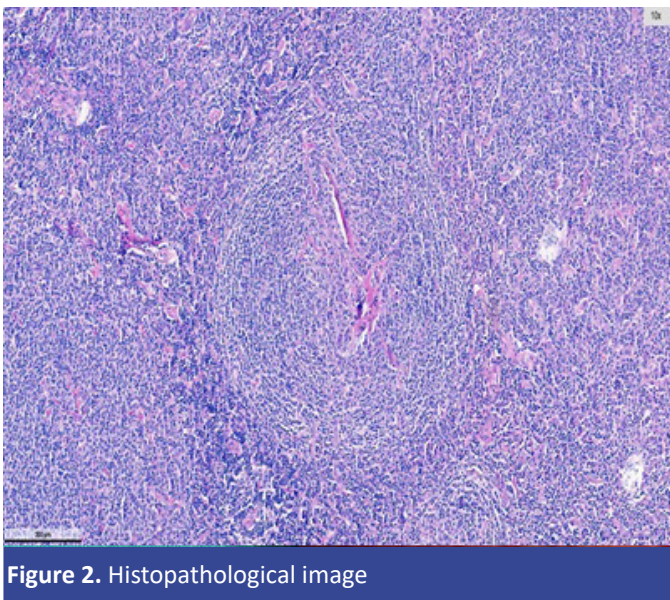


Figure 2. Histopathological image

DISCUSSION

The CD is a rare lymphoproliferative disease that should be present in differential diagnoses of prolonged lymph node enlargement [1]. In this case report, a histological examination of the patient revealed the presence of CD after surgical excision of a lymph node in the axilla that did not exhibit any signs of clinical regression for around six months. CD is categorized as unicentric or multicentric based on the number of lymph nodes affected. It is further divided histopathologically into hyaline vascular, plasma cell variants, and mixed type [5]. The patient in this case report was found to have unicentric hyaline vascular type CD. The plasma cell variant presents clinically with systemic inflammation, fever, anemia, widespread lymphadenopathy, cytopenia, and severe multi-organ failure [6]. The plasma cell variant has a more unfavorable prognosis than the hyaline vascular type. Dual infection with HHV-8 and HIV is commonly observed in patients with multicentric CD [5]. Unicentric CD patients are usually asymptomatic; laboratory findings are generally normal and often detected incidentally [7]. CD may be accompanied by various complications, such as paraneoplastic pemphigus (PNP), bronchiolitis obliterans (BO), kidney injury, and idiopathic thrombocytopenic purpura (ITP). Among these, BO and PNP are regarded as indicators of poor prognosis [1]. The patient in this case study did not exhibit any complications.

The diagnosis of CD can be made through the combination of clinical, radiographic, and pathological evidence [8]. A definitive diagnosis can be achieved through the microscopic analysis of the surgical specimen. Due to the frequent lack of diagnostic results from core needle biopsy, it is necessary to perform surgical excision and histological evaluation [3]. The patient in this case report underwent a sonography-guided thick needle biopsy on a suspicious lesion in the axilla, which showed clinical and radiological signs of malignancy. However, a surgical excision was carried out due to the inconclusive nature of the biopsy result.

The treatment involves surgical excision, radiation, chemotherapy, immunotherapy, and antiviral medications. The specific treatment method depends on the histologic subtype, stage, and extent of the disease. Surgical excision is regarded as an effective therapy approach for the treatment of unicentric CD. Nevertheless, in cases when autoimmune sequelae, including immune-mediated cytopenia and bullous pemphigus, coexist with unicentric CD patients, a combination of systemic treatment and surgical excision may be required. These patients exhibit a more severe and rapidly progressing clinical course [7, 8]. Corticosteroids, rituximab, thalidomide, lenalidomide, bortezomib, cyclosporine, sirolimus, and interferon are possible options for the treatment of multicentric CD [5]. In this case report, only axillary LN excision was performed because it was unicentric and had no accompanying complications.

CONCLUSION

CD is a rare benign condition characterized by abnormal growth of lymphatic tissue. It is most commonly found in the mediastinum and least likely in the axilla. The diagnosis is established through the integration of clinical, radiographic, and pathological evidence. It is frequently discovered incidentally while differentiating the causes of prolonged lymphadenopathy. The treatment approach differs based on the specific histologic subtype, the extent of lymph node involvement, and any associated comorbidities. The hyaline vascular subtype's prognosis is more favorable than the plasma cell subtype. Surgical excision is regarded as a definitive treatment for patients who have unicentric disease and do not have any additional problems. In this case report, the presence of CD was incidentally discovered after the surgical removal of axillary lymph nodes, which was done to determine the cause of the lymphadenopathy.

Limitation of this study: Inaccessibility of USG image and limited to a single patient.

Availability of data and materials: The data that support the findings of this study are available from the corresponding author, upon reasonable request.

Ethics Approval and Consent to Participate: Signed informed consent was obtained from the patient included in this case report and is available upon request. The Declaration of Ethics, which requires only the patient's informed consent because it is retrospective and complies with CARE rules, has been exempted by **Saglik Bilimleri University, Prof. Dr. Cemil Tascioglu City Hospital**. The study was conducted in accordance with the Declaration of Helsinki.

Scientific Responsibility: All authors analysis and interpretation of data. All authors read and approved the final manuscript. All authors have participated sufficiently in the work and agreed to be accountable for all aspects of the work.

Ethics

Informed Consent: A written informed consent has been granted from the patient.

AI statement: No Artificial Intelligence (AI) application was used in the preparation of this study.

Footnotes

Authorship Contributions

O.Y.: Literature review, data collections, study design; M.G.D.: Data Collections, Literature Review, English Editing; A.E.: Data Collections, Literature Review; M.A.: Data Collections, Literature Review; O.Y.: Analysis, Critical Revision of the Article, and Literature Review; U.K.: Study design, analysis, and interpretation, writing the article, critical revision of the article, and literature review.

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