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CASE REPORT

A rare presentation of unicentric Castleman's disease in the thigh: A case report and review of literature

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Abstract

BACKGROUND

Castleman's disease (CD) is a rare lymphoproliferative, emulating both benign and malignant diseases. The diagnosis of CD is formulated upon the combination of clinical and laboratory criteria and ultimately confirmed by histopathological assessment. Due to its rarity, CD presents a challenge in treatment selection, with available options encompassing surgery, chemotherapy, and autologous stem cell transplantation. However, studies suggest that surgical resection of the lesion is the most effective treatment modality, especially for unicentric CD (UCD).

CASE SUMMARY

Here, we describe the case of a 25-year-old woman who presented with painless left thigh swelling for 10 wk. She had been following a low-fat diet to lose weight and had normal laboratory results. Magnetic resonance imaging revealed a well-circumscribed, demarcated cystic lesion located in the left inguinal region with eccentrically positioned signal void vascular structures, measuring 4.3 cm × 3 cm × 3.2 cm, likely of lymphoid origin. The patient underwent surgical resection, and the final histopathology showed a vascular proliferation and hyalinization of the vessel walls, along with atretic germinal centers traversed by penetrating vessels, consistent with CD. The patient was discharged home one day after the procedure in good condition, with a follow-up appointment scheduled in our outpatient clinic.

CONCLUSION
Although surgical resection is the mainstay for UCD, a multidisciplinary approach is needed due the lack of specific diagnostic features and treatments.

Key Words: Castleman's disease; Lymph nodes; Surgical resection; Lymphoproliferative disorder; Case report

Core Tip: Castleman's disease (CD) is a rare disorder primarily affecting lymph nodes and associated tissues. Clinical characteristics and survival vary significantly among the three histological subtypes of CD. Diagnosis of CD primarily relies on histopathological examination, supported by imaging modalities such as computed tomography scan, magnetic resonance imaging, and ultrasound. Histopathological examination is crucial for diagnosing both unicentric-CD (UCD) and multicentric-CD, especially after ruling out other disorders, including infections, malignancies, and autoimmune conditions. Despite its rarity, CD presents a range of treatment options. However, studies consistently highlight surgical resection as the optimal treatment modality, particularly for UCD.

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INTRODUCTION

Castleman's disease (CD), alternatively referred to as giant lymph node hyperplasia or angiofollicular lymph node hyperplasia, encompasses a spectrum of rare diseases within the family of lymphoproliferative disorders. This non-malignant proliferation manifests in two distinct forms: Unicentric CD (UCD) and multicentric CD (MCD)[1].

MCD constitutes 15% of the total CD cases and has an incidence of approximately 3.4 *per* million *per* year in the United States[2,3]. MCD type can be further classified into two subtypes including human herpesvirus 8 virus (HHV)-8-positive and HHV-8-negative (or idiopathic) MCD. Different from UCD, MCD is a systemic, progressive, and frequently fatal disorder affecting multiple nodes and accompanied by generalized signs and symptoms. These include lymphadenopathy, hepatosplenomegaly, fever, fatigue, and weight loss, with more severe cases complicated by thromboembolic disorders, anemia, and hypoalbuminemia. These events are triggered by the increased production of proinflammatory markers such as interleukin 6 (IL-6) and fibrinogen. MCD treatment often requires a combination of corticosteroids, immunomodulatory agents, and chemotherapy[4].

UCD is the most common type of CD, with an approximate incidence of 16-19 *per* million *per* year in the United States [2,3]. UCD typically manifests in the third to fourth decade of life, with no apparent gender preference[3,5], although a mild preponderance among females was reported[6]. To date, the cause of the disease remains elusive, though several hypotheses have been postulated[1]. UCD typically affects one or multiple lymph nodes in a single location, more frequently the mediastinum, abdominal cavity, retroperitoneum, pelvis, neck, and less commonly, in the axillary or inguinal regions[5,7]. As UCD usually involves the slow growth of lymph nodes at a single anatomical site, clinical symptoms predominantly result from the local mass effect[4].

Histopathologically, CD can be distinguished in three subtypes: Hyaline vascular (HV), plasma cell (PC), and mixed. The majority of UCD cases exhibit a HV-type histology (74%–91%), although cases with PC-type histology or mixed features have also been documented (9%–26%)[8]. Considering its nonmalignant nature and isolated location, surgical resection is often curative and thus the preferred approach[5].

Here we present a case study concerning UCD located in the inguinal region in a young 25-year-old female.

CASE PRESENTATION

Chief complaints

A 25-year-old woman presented to our clinic with a painless left thigh swelling persisting for 10 wk following a low-fat diet for weight loss.

History of present illness

10 wk earlier, the patient noticed painless swelling in her left thigh after starting a low-fat diet for weight loss. She visited our hospital to seek further evaluation and assessment.

History of past illness

The patient denied any history of sweating, fatigue, anorexia, or jaundice. Additionally, she had no chronic conditions such as vascular diseases, connective tissue disorders, infections, malignancies, autoimmune diseases, viral illnesses like human immunodeficiency virus (HIV) or HHV-8, diabetes, trauma, interventions, or concurrent medication use.

Personal and family history

The patient married at an appropriate age, had two daughters, a healthy spouse and parents, and no family history of connective tissue disorders, vascular diseases, infections, malignancies, or autoimmune diseases. She also denied a history of smoking or alcohol consumption.

Physical examination

On examination, a mobile, non-tender subcutaneous mass measuring approximately 5 cm × 3 cm was noted in the left inguinal region. Vascular and neurological examinations of the lower limb were unremarkable. Chest and abdominal examinations revealed no tenderness or palpable masses. A comprehensive lymph node examination was within normal limits.

Laboratory examinations

Preoperative complete blood count and coagulation profile showed: Haemoglobin: 13.1 g/dL; leukocyte count: 8.70 × 10^9/L; hematocrit: 35.5%; platelet count: 359 × 10^9/L; prothrombin time 11.5 s; partial thromboplastin time: 27.9 s; international normalized ratio: 0.98. Liver and renal function tests, erythrocyte sedimentation rate (ESR), and C-reactive protein were all within the normal range. Systemic levels of IgG (0.8 g/L), IgM (0.6 g/L), and IL-6 (10 pg/mL) were also normal. Viral screening for HHV-8 and HIV was negative.

Imaging examinations

The initial workup included an ultrasound of the left thigh, which revealed an ovoid, intensely hypervascular mass lesion located at the left inner thigh, approximately 1 cm beneath the skin surface, measuring 3.5 cm × 1.6 cm × 5.2 cm. Magnetic resonance imaging (MRI) demonstrated a well-circumscribed, oval-shaped cystic lesion with markedly high-signal intensity on short tau inversion recovery sequences and low signal intensity on T1-weighted images, projected in the left inguinal region. Eccentrically positioned signal void vascular structures were observed, indicative of vascular involvement. The lesion measured 4.3 cm × 3 cm × 3.2 cm, likely of lymphoid origin (Figure 1).

Histological findings

The final histopathological examination revealed vascular proliferation, hyalinization of vessel walls, and atretic germinal centers traversed by penetrating vessels. The mantle zones exhibited thickening with lymphocytes arranged in layers (Figure 2).

FINAL DIAGNOSIS

UCD.

TREATMENT

The patient underwent surgical resection of the mass.

OUTCOME AND FOLLOW-UP

The patient was discharged home one day after the procedure in good condition, and a follow-up appointment was scheduled in our outpatient clinic. At 1 year, a computed tomography (CT) scan of the chest, abdomen, and pelvis did not reveal any involvement of other lymph nodes. Viral screening for HHV-8 and HIV remained negative.

DISCUSSION

CD represents a rare lymphoproliferative disorder encompassing a spectrum of diverse conditions, including UCD and MCD, with or without association with HHV8[1,4,5]. This case highlights a unique presentation of UCD in the inguinal region, contributing to the limited literature on this uncommon manifestation. Various aspects of this clinical case offer avenues for discussion. The risk factors for UCD are not clear, and its etiology remains largely unknown, mirroring the complexity of CD as a whole.

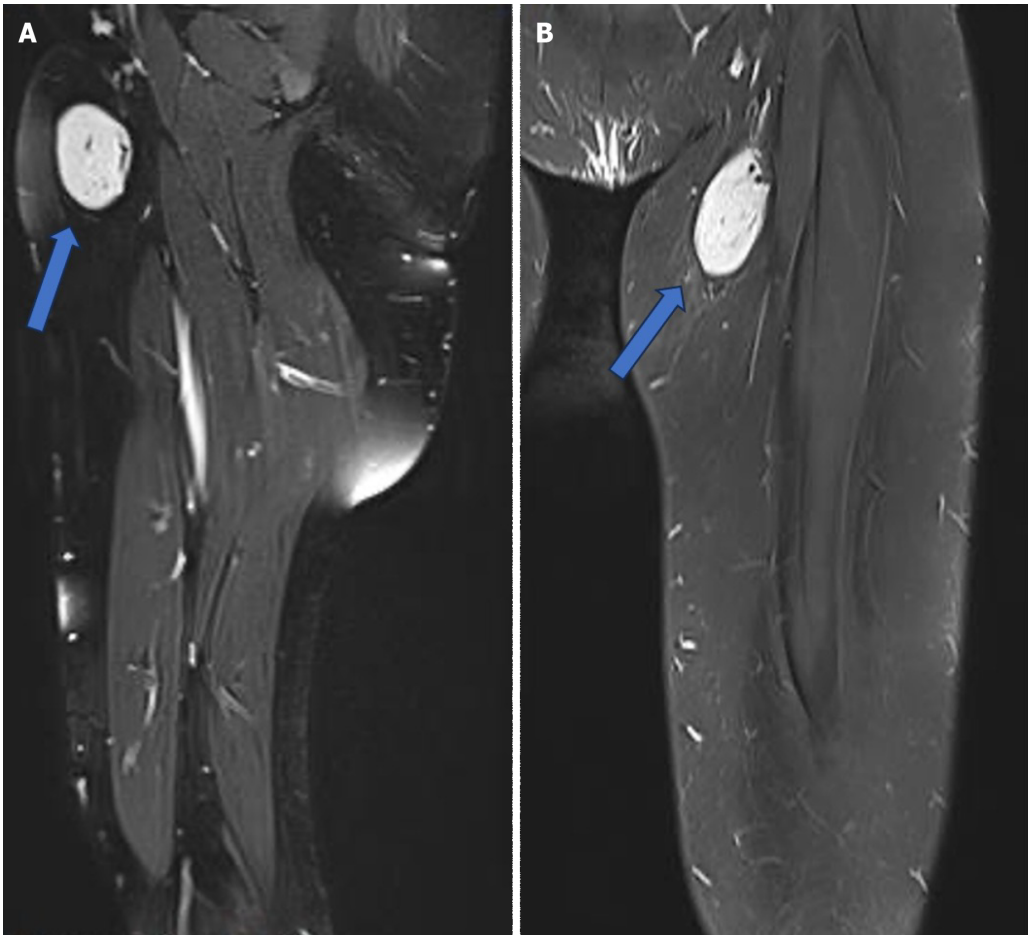

Figure 1 Magnetic resonance imaging showed a well-circumscribed and demarcated cystic lesion projected in the left inguinal region with eccentrically positioned signal void vascular, likely lymphoid in nature (arrow). A: Sagittal view. B: Coronal view.

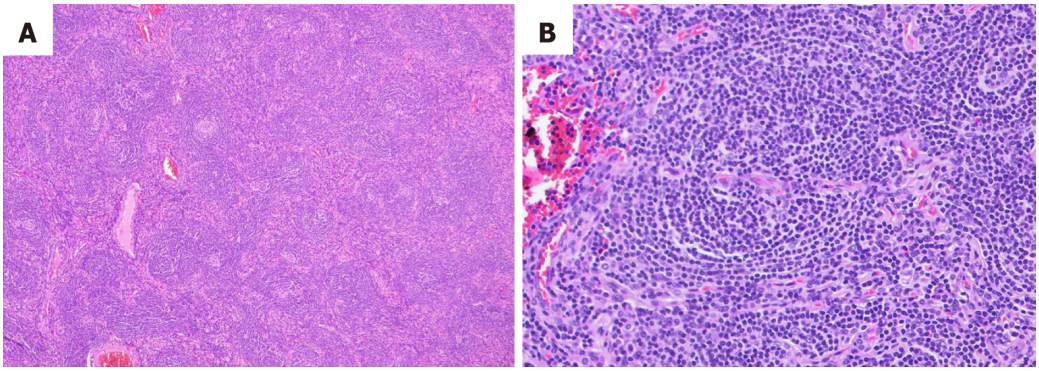

Figure 2 Hematoxylin and eosin staining showing. A: Regressed follicles with increased endothelial venules; B: Higher magnification of one of the regressed follicles showing a prominent mantle zone (onion skin appearance) and penetrated by vessel.

In MCD, the relationship with HHV-8 is not consistently identified, while other MCD cases are associated with the so-called polyneuropathy, organomegaly, endocrinopathy, monoclonal PC disorder, skin changes syndrome, also known as Crow-Fukase syndrome[9]. This syndrome is considered a variant of plasma cell dyscrasia, typically involving the overproduction of abnormal PC in the bone marrow[10].

Regarding UCD, while various hypotheses have been proposed, including the involvement of infectious agents and autoimmune processes, no definitive cause has been identified. This underscores the complexity of the disease and emphasizes the need for further research to elucidate its underlying mechanisms. Evidence suggests that UCD is likely a clonal neoplastic condition, with the stromal cells, particularly follicular dendritic cells (FDCs), being the most probable cell of origin[11]. Given this hypothesis, an investigation employing next-generation sequencing of UCD lymph node tissue revealed somatic mutations in the platelet-derived growth factor receptor β gene in nearly 20% of cases. These mutations were predominantly found in CD45− cells, likely representing stromal cells[12]. Notably, the authors

performed in vitro studies and further confirmed that this mutation leads to a gain of function, providing proliferative and survival advantages[13]. It was also noted that there were no documented monoclonal immunoglobulin and T-cell receptor gene rearrangements[14]. Nonetheless, the presence of dysplastic FDCs may explain the increased propensity of these individuals to develop FDC sarcoma[4,15].

From a clinical perspective, UCD frequently presents as a painless mass confined to a solitary anatomical site, consistent with the presentation observed in our patient. Compression on contiguous anatomical structures can lead to various symptoms; for example, nodules in the chest can cause dyspnea and coughing[4]. In the PC subtype of UCD, patients often present with symptoms such as fever and fatigue, significantly impacting their quality of life. These symptoms may persist over time and vary in intensity. Additionally, laboratory abnormalities are commonly observed in this subtype, including elevated IL-6 levels[4]. Polyneuropathy, Hodgkin disease, non-Hodgkin lymphoma, and FDC sarcoma can be found in up to 18% of UCD cases[16].

Although occurrences in regions such as the inguinal area are less frequent compared to sites like the mediastinum or neck, UCD can indeed affect these less common areas. Therefore, conducting a thorough differential diagnosis to distinguish UCD from other lymph node masses becomes crucial in clinical practice. In this context, imaging modalities, including ultrasound, CT, and MRI (Figure 1), play crucial roles in the diagnosis and characterization of UCD lesions, aiding in treatment planning. Given the specific anatomical site of the lesion in the inguinal region, we opted for MRI over CT to provide better visualization of the surrounding soft tissues and vascular structures. MRI offers superior soft tissue contrast resolution, making it particularly useful for delineating the anatomy of the inguinal region and identifying any associated vascular abnormalities. Additionally, MRI is capable of demonstrating flow void structures, which are indicative of vascular channels and can help identify feeding vessels supplying the lesion. This capability is especially relevant in cases where vascular involvement or tumor vascularity needs to be assessed[15].

In numerous cases, the presence of an enlarged lymph node may prompt suspicion of lymphomas. Consequently, histopathological examination remains the gold standard for diagnosing UCD. The histopathological aspects vary, with subtypes including HV, PC (found especially in HHV8+ and idiopathic MCD cases), or mixed[17]. However, in unicentric forms, the HV subtype predominates. In our case, the investigations facilitated a straightforward diagnosis, showing characteristic findings such as vascular proliferation, hyalinization of vessel walls, and atretic germinal centers (Figure 2)[8]. Although histopathology was conclusive for a typical HV UCD form, given the concern for additional unrecognized lesions, we conducted viral investigations. Our patient's negative viral screening results for HHV-8 and HIV suggested that UCD in this case was not associated with these viral infections, which are commonly implicated in the multicentric forms of the disease. This underscores the importance of comprehensive diagnostic evaluation, including viral screening, to differentiate between different subtypes of CD and guide appropriate management strategies[18].

Generally, the treatment of CD depends on the disease subtype, with curative surgery being the gold standard for UCD and monoclonal antibody-based immunotherapy being the standard of care for MCD[4].

Surgical excision of the affected lymph node in UCD provides both diagnostic confirmation and therapeutic benefit[5,19], with an overall survival > 90% at 5 years and prompt resolution of local and systemic symptoms (if present) following resection[4]. The role of radiotherapy is controversial, although this approach can be considered in non-resectable cases[20]. According to a recent international evidence-based consensus, unresectable asymptomatic UCD may be monitored, whereas symptomatic unresectable UCD usually requires treatment with rituximab combined or not with steroids, or anti-IL6 antibody therapy, followed by surgical excision[4]. Strategies for embolization have been also proposed[21].

On the other hand, the treatment of MCD is primarily based on immunomodulatory agents. Due to the central role of IL-6 in MCD pathophysiology, the use of anti-IL-6 therapies (*e.g.*, rituximab, tocilizumab, siltuximab) is recommended as the first-line treatment in all patients with idiopathic MCD, with the addition of corticosteroids in case of severe symptoms. In case of inadequate response, cytotoxic chemotherapy as per lymphoma or myeloma regimens can also be considered[4].

In our patient's case, excisional biopsy successfully removed the lesion, resulting in the resolution of symptoms. However, follow-up is essential to monitor for any (rare) recurrence or development of complications[22]. The main characteristics of UCD are outlined in Table 1.

CONCLUSION

CD is a lymphoproliferative disorder causing an overgrowth of cells in the lymph nodes throughout the body. The diagnosis is primarily made through CT scan, MRI, ultrasound, and histopathologic examination. Treatment varies depending on the subtype. However, since CD involves the enlargement of lymph nodes, complete surgical resection of the tumors is the standard approach across all subtypes. CD necessitates a multidisciplinary approach in evaluating and treating patients with this rare pathology, mainly due to the condition's lack of specific features and limited treatment modalities.

Table 1 Main features of unicentric Castleman disease

	Features	Ref.
Epidemiology	Incidence (United States): 16–19 cases per million person-years. No gender predominance	[2,3]
Cause	Probable clonal neoplastic condition: Benign neoplasm of FDCs	[4,10-14]
Risk factors	Not known	[10]
Clinical presentation	Single lymph node mass (non-tender lymphadenopathy): Asymptomatic or manifesting as pain, sensation of heaviness, compression of organs, and anatomic structures (*e.g.*, chest lesions). In the plasma cell subtype: Fever, fatigue, and laboratory abnormalities (IL-6 secretion: Elevated ESR, elevated CRP, anemia)	[1,4-7,9, 15,16,20, 21]
Disorders associated	Polyneuropathy, Hodgkin disease, non-Hodgkin lymphoma, and follicular dendritic cell sarcoma	[16,20,21]
Site	Mediastinum, neck, abdominal cavity, retroperitoneum, and pelvis. Less frequently in axillary or inguinal regions	[5,7]
Imaging	Useful for differential diagnosis: Ultrasonography, CT, MRI	[15,20]
Differential diagnosis	Lymphomas; cancer (*e.g.*, lymph node metastasis); benign proliferative lesions (*e.g.*, thymoma); inflammatory processes (*e.g.*, sarcoidosis); infections (*e.g.*, tuberculosis)	[17,18]
Histopathology	Confirm the diagnosis: Hyaline vascular or plasma cell-type aspects	[8,20]
Virology	HHV-8 negative	[18]
Therapy	Surgical excision is preferred. Radiotherapy, embolization, or systemic therapy for non-resectable symptomatic lesions	[5,19-21]
Outcome	Long-term follow-up to monitor for potential recurrences	[22]

FDCs: Follicular dendritic cells; ESR: Erythrocyte sedimentation rate; CRP: C-reactive protein; CT: Computed tomography; MRI: Magnetic resonance imagining; HHV-8: Herpesvirus/human herpesvirus-8.

FOOTNOTES

Author contributions: AlSheikh S, Altoijry A and Al-Mubarak H designed the research study; Alanezi T completed the first draft of this manuscript and performed the experiments and data collection; Alsallum OD and Alanezi T was involved in data collection; Alakeel F provided pathological findings; AlSheikh S, Altoijry A, Al-Mubarak H, Alsallum OD, lakeel F and Alanezi T revised the manuscript. All authors have read and approved the final version of the manuscript.

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