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Case Report

Retroperitoneal Unicentric Castleman's disease, a case report

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ABSTRACT

Introduction: Castleman's disease (CD) is an unusual nonmalignant lymphoproliferative disorder which presented with enlarged hyperplastic lymphoid tissue and had three subtypes, hyaline-vascular, plasma cell, and mixed form according to histologic findings.
Case report: We report a 31-year-old woman who presented with abdominal pain from 3 months ago. Imaging studies showed a well-defined mass on back of pancreas without any invasion. The histopathological examination revealed lymphoid tissue with prominent vascular proliferation and hyalinization of the vessel walls compatible with Hyaline vascular CD. So, pathological assessment is essential for the diagnosis.
Conclusion: Unicentric CD must be considered as differential diagnosis in a solid solitary abdominal mass. In patients with abdominal mass of an ambiguous nature, surgical resection is necessary as diagnostic tool and the first treatment approach.

1. Introduction

Castleman's disease (CD) was described in 1954 at first time and known as an unusual nonmalignant lymphoproliferative disease [1]. The reason is unidentified, and it is considered by the growth of neoplastic masses of lymphoid organ. Clinical subtypes including unicentric and multicentric and histological subtypes including hyaline vascular (HV), plasma cell (PC), and mixed are described [2]. CD mostly happens in young people (15–35 years) with no gender tendency for incidence the disease [3]. The etiology of CD is uncertain with different clinical manifestations that cause to many diagnostic and treatment difficulties [4]. Unicentric CD has a good prognosis and needs only surgery with no additional treatment. The patients usually persist symptomless afterward [5]. So, certain diagnosis is essential to avoid unnecessary treatment. The diagnosis of CD is challengeable for clinicians and pathology evaluation is requirement to affordable treatment [6]. The clinical symptoms and signs are various and the diagnosis is problematic and optimal managing is still indefinite. Our study reports a 31-year-old woman as an infrequent case of unicentric CD, who presented with a retroperitoneal mass around of pancreas. This case report has been reported in line with the SCARE Criteria [7].

2. Case presentation

A 31-year-old woman was referred to the general surgery department from emergency room with abdominal pain lasting 3 months without weight loss or lack of appetite. The patient received only analgesic for abdominal pain. Family and past medical histories were unremarkable. Routine laboratory data were within normal limits. Human immunodeficiency virus (HIV) and other viral tests were negative. Tumor markers analysis were negative. The patient had mild tenderness on right upper quadrant on physical examination. Radiologic studies showed a hypoechoic mass at back of pancreas on sonography. Also, abdominopelvic computed tomography (CT) scan revealed an enhanced retroperitoneum mass back of duodenum and pancreas (Fig. 1). Endoscopic ultrasound (EUS) report showed a hypoechoic lesion at head and uncinate process of pancreas measuring 38 × 35mm. The first diagnosis was malignant tumors including adenocarcinoma or lymphoma. But mass EUS guided fine needle aspiration examination was negative for malignancy. Because no definitive pre-surgery diagnosis recognized, surgical resection was necessary. The patient underwent mid-line laparotomy and suspected solitary mass was resected (Fig. 2). Sent specimen for pathology evaluation contains a lobulated tan mass measuring 4 × 3.5 × 3cm. On cut section homogeneous whitish surface is seen. The pathology evaluation revealed lymphoid tissue with prominent vascular proliferation and hyalinization of the vessel walls and mantle zones are

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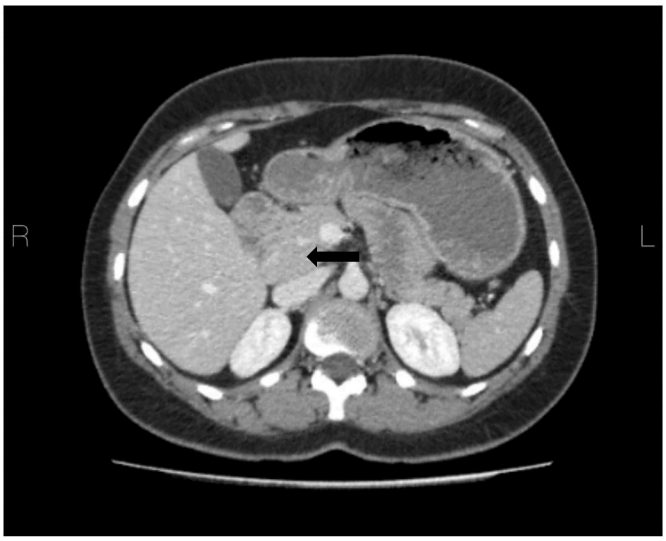

Fig. 1. CT scan showed an enhanced mass on retroperitoneum back of pancreas without any invasion.

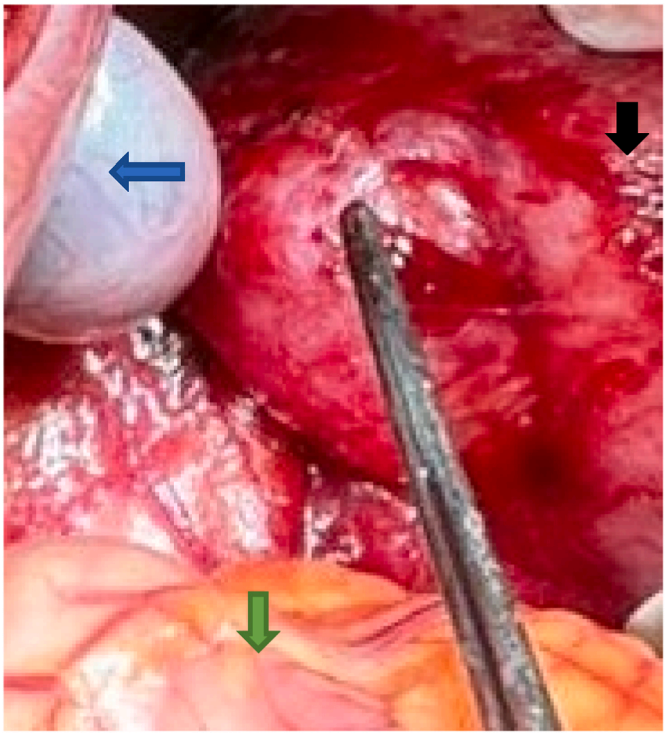

Fig. 2. On operation revealed a round solid solitary mass (gallbladder: blue arrow, tip of forceps: mass, green arrow: transvers colon, black arrow: pancreas).

thickened with lymphocytes arranged in layers - onion skin appearance compatible with Hyaline vascular CD (Fig. 3). Also, immunohistochemistry (IHC) staining for CD 21 and CD23 were done to rule out concomitant follicular dendritic sarcoma which showed negative results and diagnosis was established. She was discharged in appropriate condition after surgery and received no supplementary therapy after surgery. In follow up examination, she is still asymptomatic after 6 months from surgery.

3. Discussion

CD is an infrequent lymphoproliferative disorder that is considered by lymphoid tissue proliferation and enlargement [8]. The most usual site of the neoplasm is in the mediastinum. Abdominal location is uncommon [9]. CD most usually displays as unicentric CD with an enlarged mass and is in differential diagnosis of lymphoma, metastatic lymphadenopathy, and other inflammatory lymphadenopathy [10]. The clinical exhibitions of CD differ seriously among the unicentric and multicentric subtypes. Unicentric disorder is generally asymptomatic or might existing with lymph node enlargement. Unicentric subtype frequently has a good outcome and is cured by surgical resection [11] which is similar our case. Furthermore, HIV-related CD is considered to be connected to human herpes virus-8 (HHV-8) [10]. Our case had HIV test with negative result [3]. Retroperitoneal unicentric CD in around of pancreas is regularly obscured and hard to identify. Also, there is no definite exhibition to discriminate it from a neuroendocrine tumor or lymphoma. Imaging presentations of CD are extremely problematic to differentiate from other diseases, and the pre-surgery imaging diagnosis does not generally compatible with histopathological diagnosis after surgery [12] which in our case, no definite diagnosis was present before surgery and surgical removal was essential. Unicentric CD should be noticed in solid abdominal or retroperitoneal masses. An improved information of this disorder and its distinctive would assistance surgeon to evade unreasonably wide resection for this nonmalignant disease when encountering with abdominal or retroperitoneal masses [13]. While the best management for CD is still unidentified, surgical resection of the unicentric subtype of the mass is gold standard for treatment. But multicentric subtype of CD has a poor outcome and typically managed with a combination of corticosteroids and chemoradiotherapy. So, in our patient with unicentric CD, a complete surgical resection was done; and there is no evidence of relapse up to now [14]. Wilbur B. et al., evaluated 16 patients on their cross-sectional study and showed 100% of patients cured after surgery after 5 years follow up [15]. Bracale et al. evaluated 53 patients on case series article with CD and showed the best choice for Unicentric CD treatment unrelatedly of histological subtype (HV or PC), is a complete surgical removal, which is a therapeutic method in nearly all patients without relapse after 20 years follow up after time of surgery [16]. Also, its infrequency, absence of definite indicators, and conclusive diagnostic radiologic presentations, pre-surgery diagnosis is problematic and surgeons might confront trouble in operation due to its hypervascular nature [17].

4. Conclusion

In the existence of a solitary solid retroperitoneal mass, the probable diagnosis of unicentric CD should be noticed. In suspected mass for unicentric subtype of CD, complete surgical removal is adequate with avoidance of unnecessary extensive surgery.

Ethical approval

Ethical approval is not needed due to the fact that the treatment of the patient was based on approved options and was not controversial.

Sources of funding

None.

Author contribution

Seyed Amir Miratashi Yazdi: surgeon performing the operation, major contribution of the idea, study design, revise the paper, and follow up. Elham Nazar: pathologist, data collection, and writing the paper.

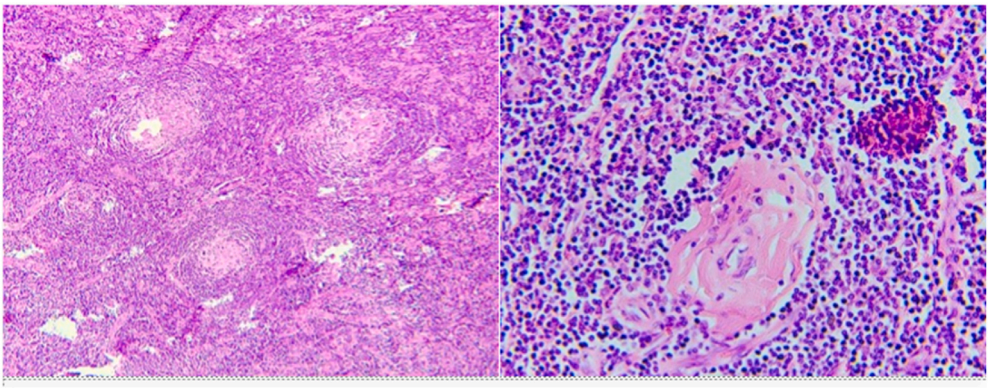

Fig. 3. Histopathologic examination showed lymphoid tissue with hyalinized vascular component (H&E, X100 and X400).

Consent

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

Registration of research studies

1. Name of the registry:
2. Unique Identifying number or registration ID:
3. Hyperlink to your specific registration (must be publicly accessible and will be checked):

Guarantor

Elham Nazar.

Provenance and peer review

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Declaration of competing interest

None.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2022.104109.

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