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Parenchymal Sparing Surgery with Minimally Invasive Approach in Castleman's Disease: Case Report

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I. Introduction

Castleman's disease, which is also known as the angiofollicular lymph node hyperplasia, is a rare lymphoproliferative disorder. Regarding the clinical appearance, the disease has two types, namely the unicentrically and multicentrically placed. The unicentric placement (UCD) is typically localised and generally asymptomatic. The multicentric placement (MCD), on the other hand, is often a systemic disease seen in presence of an immune system defect, such as the HIV. The unicentric pattern is generally localised in mediastinum and pulmonary hilum and it is the most frequent variation.

The conventional surgical approach is thoracotomy for the Castleman's disease located in the thorax. Having been performed more often in recent years, thoracoscopic surgery has been seen to be a safe and efficient approach for the treatment of mediastinal tumors (1, 2), though a limited amount of evidence and few cases have been reported regarding the VATS application in the UCD. The parenchymal lung involvement of the disease is rarely encountered. We have treated our subject with the videothorascopic approach, which is rarely reported in the literature, leaving the parenchyma intact.

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II. Case Presentation

Our patient was a 45 year-old male. Following the incidental identification of an anomaly during a medical inspection in his work place, he was sent to our hospital for further investigation and treatment. A non-smoker, his chest radiography did not show any significant lesions, except for a left hilar enlargement (Fig. 1A). His thoracic CT scan indicated that there was a 3*2 cm mass stretching from the non-enhancing left hilar area to the parenchyma (Fig. 1B). The FDG involvement value of the lesion was measured to be SUVmax 1.6 in PET CT (Fig. 1C). The physical examination and the blood chart were normal. The patient did not have any immune system defects and no diagnostic findings were encountered in his bronchoscopy examination. There were also no diagnostic information in his family background and he did not have any additional diseases.

The patient, who was decided to undergo a surgery for diagnosis and treatment, was intubated with a double-lumen endotracheal tube under general anaesthesia. A 3 cm mass lesion was identified to be sitting on the interfissural area in the hilar vicinity via the single-lung ventilation three-port VATS approach in the right lateral decubital position (Fig. 1D and E). Severely vascularised, the mass was removed from the thorax as an en bloc by dissecting it from the lingula segment parenchyma with an endostapler and by sparing the interlobar arteries and veins (Fig. 1F). Following hemostasis, a single chest tube was placed inside the thorax. On the third post-operative day, the tube was removed and the patient was dispatched. The follow-ups of our patient still continue in the twelfth post-operative month.

In the histopathological examination, an image of a typical "lollipop folicule" was seen inside the vascularised hyalinising germinal centre (Fig. 2A and B). The simultaneous haematological check-ups of our case, which is regarded the hyalinovascular type Castleman's disease, are still on-going.

III. Discussion

Castleman's disease has three histopathological subtypes, namely the hyalinovascular (90% of all cases), the plasma cell (8-9% of all cases),

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and the intermediary (1-2% of all cases) (3). The result in our case turned out to be the most frequently encountered one – the hyalinovascular-type.

Although the disease does not have a high-specificity method of diagnosis, the lesions can be identified easily by a CT scan. The median SUVmax value of the UCD in the literature is 5.1 (offset values are 2.2-10) (4). The SUVmax value in our case was seen to be 1.6. PET CT can be used to reduce the differential diagnosis.

Castleman's disease can be diagnosed through a histopathological verification. Especially in unicentrically placed cases, surgical en bloc resection also provides a curative treatment. The purpose of a UCD surgery is to completely remove the lesion. For the UCD patients who received incomplete resection or were not able to be resected, response to radiotherapy was reported to be 70% (5). Expected results may not be acquired from surgery or radiotherapy with some asymptomatic local patients. With these patients, the systemic chemotherapy which is applied for the MCD is a good option.

IV. Conclusion

VATS is a significant alternative in the diagnosis and treatment of Castleman's disease placed in the thorax. Even though the conventional surgery is performed commonly, a curative treatment can be provided with a minimally invasive approach. Complete resectability is possible for lesions whose edges are clearly identified.

Declarations

Funding: This study wasn't funded.

Conflict of interest: Eylem Yentürk, Hüseyin Melek, Hülya Öztürk Nazlıoğlu and Ahmet Sami Bayram declare that they have no conflict of interest.

Ethics approval: Studies of human interest were conducted according to the ethical standards of the responsible committee or, for all subsequent revisions, the 1964 Declaration of Helsinki. Since it was a retrospective study, an ethics committee approval was not required.

Consent to participate: Written informed consent was obtained from the patient for publication. A copy of the written consent is available for review by the Editor-in–Chief of this journal.

Consent for publication: Author made consent from individuals to publish their data prior to submitting paper to journal.

Availability of data and material: Suitable.

Code availability: Suitable.

Authors' contributions: We hereby certify that the work submitted is in full accordance criteria for authorship.

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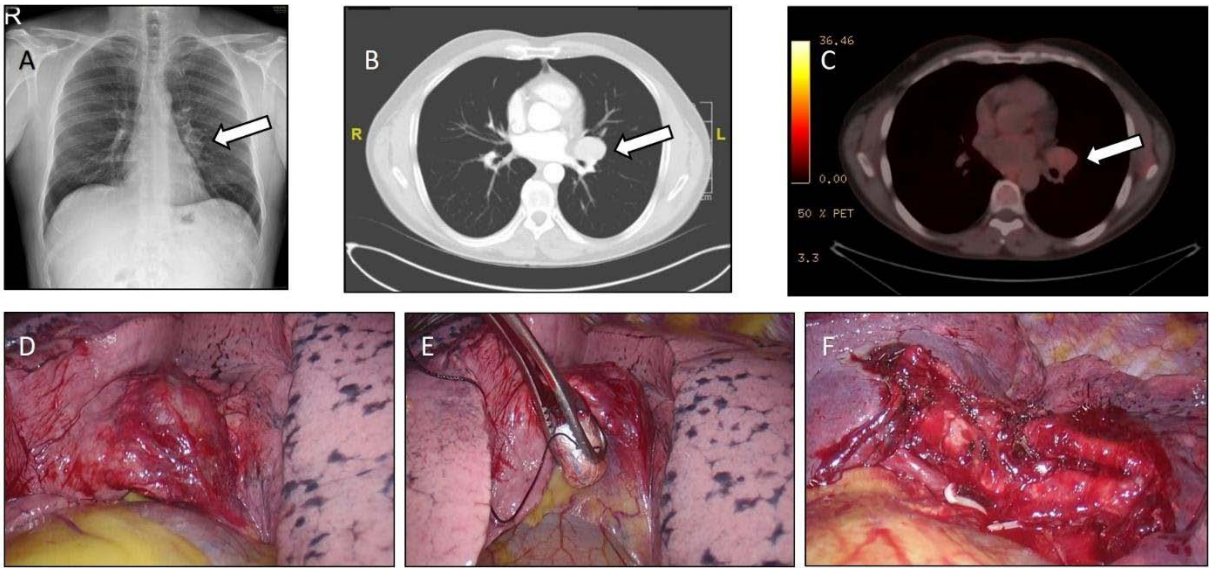


Figure 1: (A) PA chest radiography; (B) Thorax CT (C) PET CT; (D and E) Mass lesion settled in the interfissural area; (F) Image after the parenchymal protecting complete resection

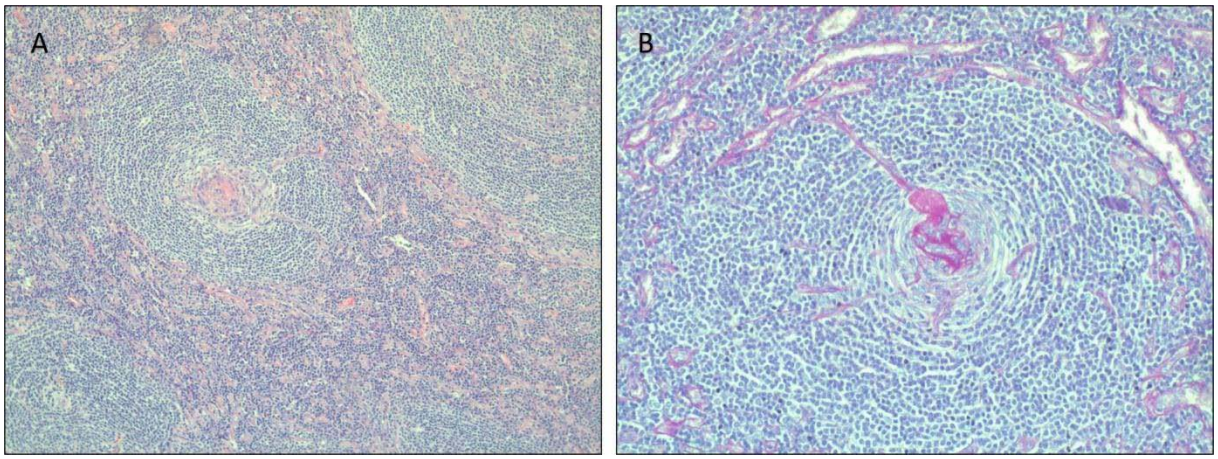


Figure 2: (A) Hyalinised germinal centre involving vascularisation (H&E, 25X); (B) Image of lollipop folicule (PAS, 200X)

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