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Research Article

SURGICAL MANAGEMENT OF INTRATHORACIC MASS WITH AN UNCOMMON DIAGNOSIS

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ABSTRACT

Thoracic cavity masses present with wide variety. Sometimes rare tumours are difficult to diagnose even after clinical and radiological correlation. Intrathoracic Castleman's disease is one of such rare entity and is difficult to diagnose despite radiological and clinical evaluation. Here we present a successful surgical management of unicentric intrathoracic castleman disease presenting as mass

INTRODUCTION

Thoracic cavity masses has wide variety of presentation. These masses may present across all ages and gender. These masses can be classified on the basis of their location/origin and may be subdivided into parenchymal, pleuralor mediastinal. Histopathologically these masses are divided into benign and malignant. Rare tumours are difficult to diagnose and require clinical and radiological correlation. Intrathoracic Castleman's disease is one of such rare entity and is difficult to diagnose despite radiological evaluation. Here we present a successful surgical management of intrathoracic castleman disease presenting as mass.

Case Report

A 36-yr old female presented with a history of breathlessness on exertion for 1 month. She was evaluated with chest x-ray (Figure1), which showed right sided massive pleural effusion. The biochemical analysis of Pleural fluid showed elevated ADA levels and cytology for malignant cells was negative. She was started empirically on ATT in view of increased ADA levels. In view of persistence of symptoms she underwent Computed tomographic scan of the chest which revealed a 7.3 x 7.2 x 6.1 cm mass in the right anterior pericardiophrenic recess with massive pleural effusion (figure 2, 3). A differential diagnosis of Solitary fibrous tumour, Lung Sequestration was suspected. In view of massive pleural effusion with significant breathlessness a tube thoracostomy was performed and slow decompression of 2 litres of pleural fluid was done. The cytology of pleural fluid was negative for malignant cells. The collection was exudative and ADA was within normal limit. Her past medical and family history was non-contributory. Physical examination was unremarkable except for reduced air entry on right side basal area anteriorly. Haematological analysis revealed anaemia with hypoalbuminemia on biochemical analysis.

Surgical resection was planned via a right postero-lateral Thoracotomy approach. A large encapsulated tumour with intact capsule and firm consistency arising from the diaphragmatic pleura in the right cardiophrenic angler anteriorly. There was minimal pleural effusion. The tumour was adherent to right lower lobe, mediastinal pleural and to diaphragm. Large calibre feeding vessels were seen arising from the right internal mammary artery. These were dissected and divided between ligatures and clips. The tumour was densely adherent to diaphragmatic surface so part of diaphragm was resected en bloc along with a wedge of adherent right lobe. Her post-operative recovery was uneventful and she was discharged from hospital on day 4. Histopathological analysis revealed a hyaline variant of localized Castleman's disease (figure 4). All resected margin were free of any tumour. She

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was referred to a haematologist for further work up of Castleman's disease. HIV and HHV 8 were done, both of which were negative. Her CECT abdomen was done to rule out multicentre disease which was negative. The patient remained asymptomatic in her post-operative period with no further recurrence of symptoms till one year of follow up.

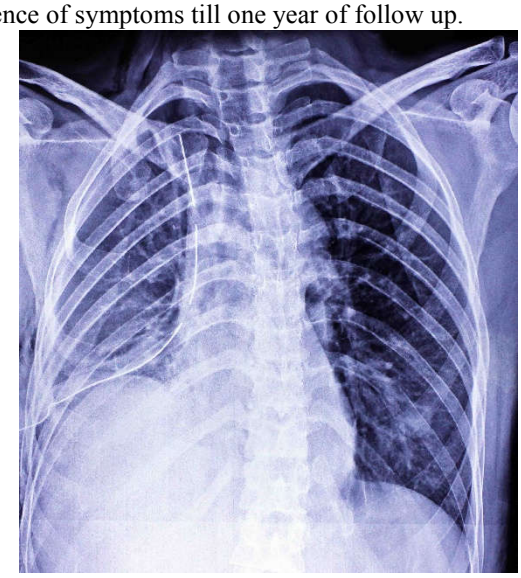

Figure 1

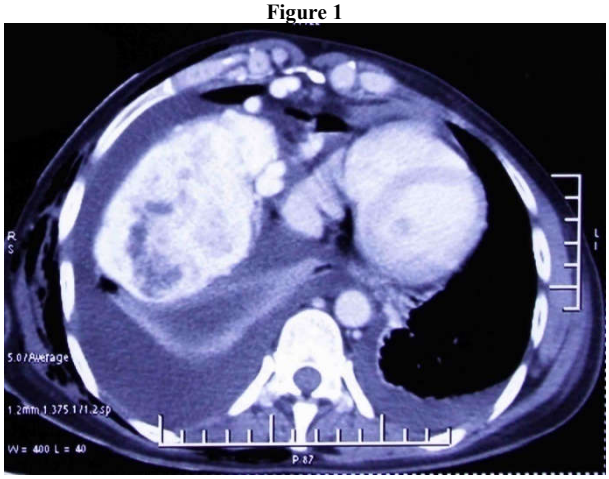

Figure 2

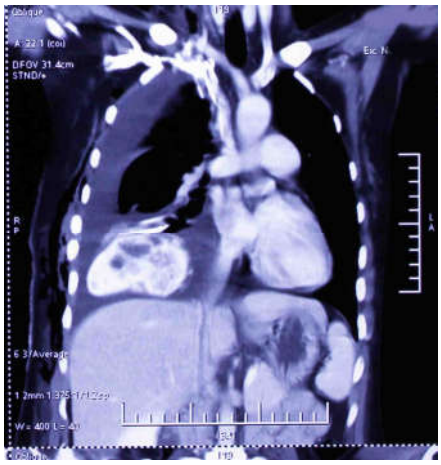

Figure 3

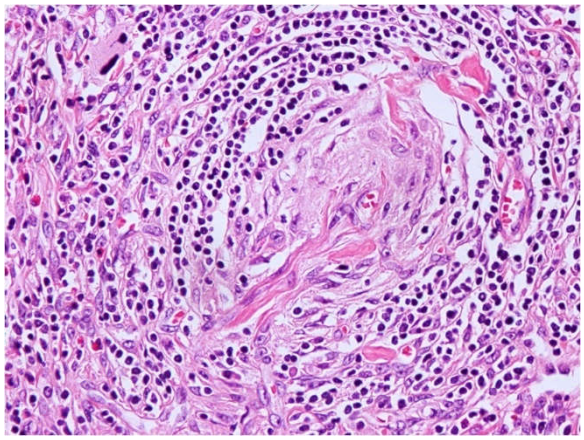

Figure 4

DISCUSSION

Castleman disease (CD) is a heterogenous group of benign lymphoproliferative disorder described by Dr. Benjamin Castleman in the 1950s (Castleman *et al*, 1956). Though the insight into its pathogenesis has improved with time it is still far from complete. Castleman's disease can be Unicentric (UC) or Multicentric (MCD). Pathologically it is classified as hyaline vascular type, plasma cell type and mixed type (Cronin *et al*, 2009). MC Castleman's is a systemic disease usually associated with plasma cell type variant and presents with variety of clinical symptoms and signs which include night sweats, fever, generalized peripheral lymphadenopathy and hepatosplenomegaly (Peterson *et al*, 1993). It may be associated with HIV and HHV-8 .MC disease has also been associated with malignancies like Kaposi's sarcoma and lymphomas. It is managed with multimodality approach including surgery, radiation, steroids, antiviral agents, specific antibodies, inhibitors of cytokines activity, and chemotherapy (Casper *et al*, 1995]. Unicentric castleman's disease usually occurs in young age and is mostly asymptomatic. It is a slow growing disease with no gender or race predominance that is mostly diagnosed incidentally on radiological imaging. Only few lesions may produce symptoms due to pressure effects of the mass. Most commonly reported site is in the mediastinum, but it may also present as intra-abdominal masses or as involvement of cervical, axillary, and inguinal nodes (Keller *et al*, 1972). In the mediastinum it may mimic a lymphoma, thymoma, germ cell tumour, Tubercular mediastinal lymphadenitis, HIV related lymphadenitis, follicular hyperplasia *or Toxoplasma* lymphadenitis. Laboratory reports may show anaemia, hypoalbuminemia and eleveated Erythrocyte sedimentation rate (ESR). Serologies for hepatitis B, HHV-8, and HIV may be positive, Serum levels of IL-6, vascular endothelial growth factor (VEGF; uncommon), lactate dehydrogenase, and CRP are usually high. Biopsy of lesion which may be attempted under ultrasonographic or computed tomographic guidance, but is usually inconclusive. Excisional biopsy is usually recommended. Once the diagnosis is confirmed the treatment depends on type of disease. Surgery is curative if resection is possible in unicentric disease. Many case report and retrospective series report excellent rates of cure after surgery (McCarty *et al*, 1995). Surgery should be offered even with the possibility of partial resection which has nevertheless shown to increase the survival of patient (Olscamp *et al*, 1980).In cases where surgery is not possible neo-adjuvant

therapy with rituximab / steroids with or without Cytoxan, IL-6(R) monoclonal Antibody and other novel agents (e.g., bortezomib, lenalidomide) have been tried. Embolization and local radiotherapy may also be considered (Chronowski *et al*, 2000).

CONCLUSION

Castleman's Disease continues to pose a diagnostic challenge and may mimic many pathological conditions particularly in case of unicentric involvement in the thorax where it commonly mimics mediastinal masses like thymoma. Though it is rare it should be considered in the differential diagnosis of intrathoracic nodules/mass lesion. A meticulous diagnostic approach based on detailed history and physical examination, imaging studies and histopathological evaluation by experienced pathologist is required for a correct diagnosis. A careful search should be done for lesions elsewhere in the body by appropriate imaging studies to rule out Multicentric disease. Awareness of CD is important amongst clinicians because of the progressive clinical course with increased risk for the development of lymphoma in cases of multicentric involvement.

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