

Adult Eosinophilic Granuloma of Thoracic Spine

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Abstract

A rare case report of adult eosinophilic granuloma of upper thoracic spine involving the vertebral body. A 48-year-old male with chronic pain over lower neck without neurological involvement. MR imaging shows lytic lesion over D2 vertebral body. Open biopsy done; it is consistent with EG. Immunohistochemistry markers positive for CD1a, S100. Adult Eosinophilic Granuloma is a self-limiting and spontaneous resolution condition in adult. It should be in the list of differential diagnosis for a solitary lytic lesion of spine in adults.

Keywords: Adult eosinophilic granuloma, Solitary lytic lesion, Langerhans cell histiocytosis

Introduction

Eosinophilic granuloma (EG) is a benign disorder that occurs mostly in children and adolescents but rarely in the adult population. It is an osteolytic condition due to the proliferation of reticulohistiocytes. Although it occurs in the spine the incidence of EG is 1 in 1,500,000. It is part of the spectrum of diseases known as M. Letterer – sive M. Hand Schuller Christian [1]. It was initially described by Ortlani and Erlich and further studied by Lichtenstein and Jaffe [2]. Recently it has been classified as Langerhans cell histiocytosis since the origin of the cell is an abnormal proliferation of Langerhans cells due to the inflammatory process. EG are mostly solitary lesions affecting the spine, the other bones include the skull, ribs, pelvis, and long bones of extremities. About the spine, it most commonly is an isolated solitary lesion affecting the thoracic spine and least in the cervical spine.

Case Description

A 48-year-old adult male presented with chronic lower neck pain over the nape of the neck, aggravated by traveling. There was no radiating pain or other associated symptoms. He doesn't have other constitutional symptoms. He has tenderness on deep percussion over the lower cervical and upper thoracic region more on the right side. The neurological evaluation was normal. The laboratory parameters were favoring of inflammatory or infectious causes with an elevated erythrocyte sedimentation rate (ESR) and C reactive protein (CRP). The radiological workup was initiated. Magnetic resonance imaging both plain and contrast studies were carried out. The findings include a solitary lytic region over the right lateral aspect of the D2 vertebral body with adjacent marrow edema (Fig.

1). The radiological diagnosis was infective or inflammatory etiology. He has been posted for an open biopsy under general anesthesia. The biopsy was done by posterior approach to the upper thoracic spine D2 vertebra, the lesion was curetted through the D2 pedicle. The sample was sent for the frozen section, it was consistent with inflammatory cells. The samples were also tested for routine microbial culture and sensitivity, gene xpert, mycobacterium tuberculosis culture, fungal culture, and histopathological examination. The gene xpert shows the TB was not detected. The microbiological culture and sensitivity show no growth of organisms till seven days. The HPE finding was granulation tissue, bony fragments, and fibro-collagenous tissue with dense inflammatory cell infiltrate rich in eosinophils and vascular proliferation (eosinophil-rich lesion) (Fig. 2). The immunohistochemical markers were positive for CD1a, Langerin, and S100 consistent for EG (Fig. 3). The patient has been treated symptomatically.

Discussion

EG is one of the forms of Langerhans cell histiocytosis. It is an uncommon benign bone lesion comprising less than 1% of all bone tumors [3]. The involvement of the spine is 7 to 15%, most common in the cervical spine in adults [4]. It is a disease of childhood or adolescence and is very rare in adults. The female sex has preponderance in both age groups. The clinical presentation of thoracic and lumbar spine involvement will be pain and neurological deficits whereas in the cervical spine, it causes stiffness or torticollis. In adults, cervical spine involvement is 47% followed by thoracic (33%) and lumbar (20%) [4]. The lytic lesion involves often involves posterior elements in adults and the vertebral body in

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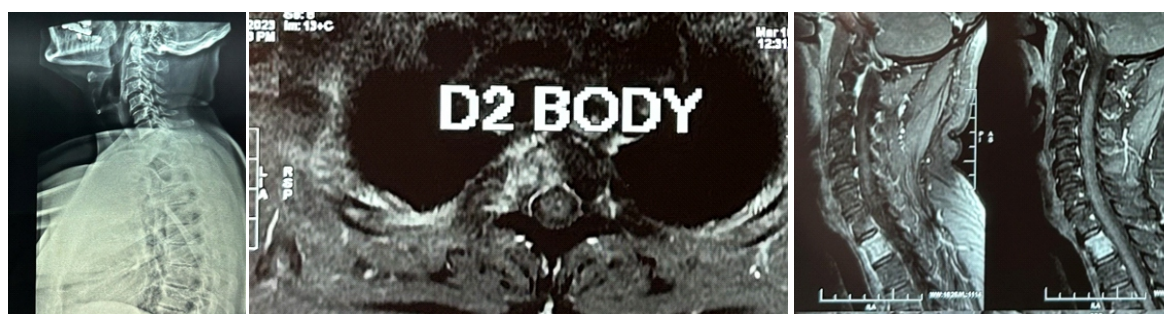


Figure 1(a-c): a, radiograph of cervicothoracic spine with no obvious lytic lesion, b. Axial T2 image shows solitary lytic lesion over right lateral aspect of D2 vertebral body, c. Sagittal contrast T1 shows hyperintense lesion of D2 body.

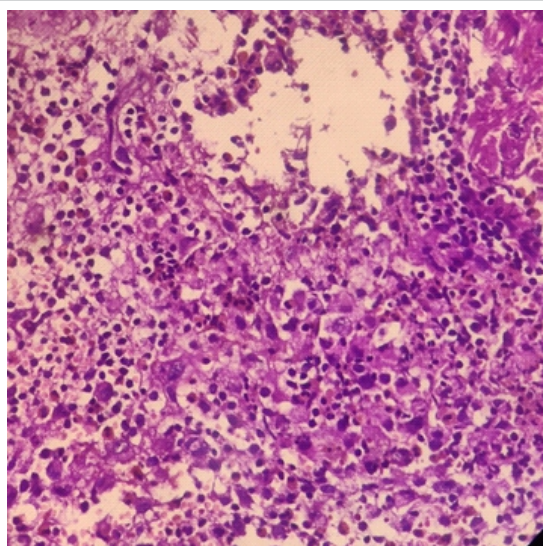


Figure 2: HPE – section shows inflammatory cell infiltrate with predominance of eosinophils admixed with lymphocytes. Few polygonal cells with oval or kidney bean shaped nuclei with fine chromatin and some with grooves noted.

children. In children, the classic xray feature is the collapse of the vertebral body known as vertebra plana which were coined by Buchmann [5].

In adults, EG is usually asymptomatic even if the x-ray shows a lytic lesion. MR imaging shows a rim enhancement lesion of the involved spine. Even then the correlation of symptoms and radiological

findings ways the diagnosis is not specific unless it confirms by biopsy and HPE [6]. The differentials to be considered on the stage of biopsy and HPE includes tumors such as myeloma, metastatic, aneurysmal bone cyst, and infection such as osteomyelitis. The immunohistochemistry markers such as CD1a, S100, and Langerin are very important in making the diagnosis. EG is a self-limiting disease. The treatment options for EG vary from observation, radiotherapy, chemotherapy, and surgery. The choice of surgical option should be considered in case of neurological involvement where decompression with or without reconstructive procedure, and curettage of the lesion should be carried out. When comparing the treatment modalities for EG there was no statistical difference in treatment options [7]. Since EG is self-limiting and has a tendency for spontaneous resolution, observation, and conservative treatment have an important role [8]. The main aim of any treatment is to maintain the architecture of the involved skeleton.

Conclusion

Adult EG is a self-limiting benign solitary lytic lesion involves most common in the cervical spine. Biopsy and HPE are very important in arriving at the diagnosis. IHC such as CD1a, and S100 should order in suspected solitary lytic lesions in the adult population. Observation and conservative treatment play a role in treating this condition. It should be considered in the list of differential diagnoses for solitary lytic lesions of the spine in adults.

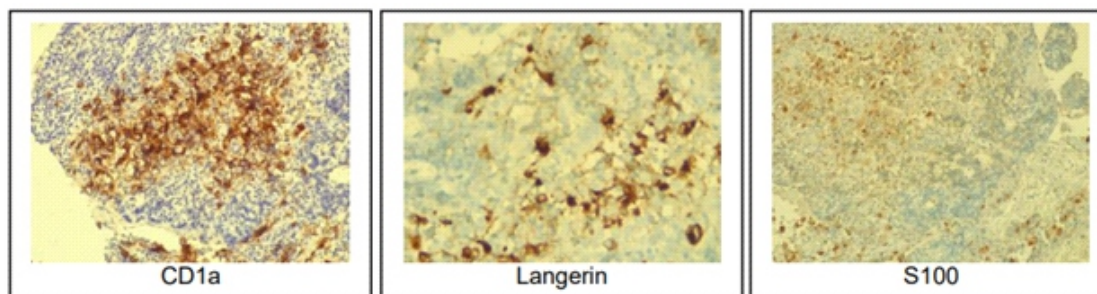


Figure 3: shows immunohistochemistry markers positive for CD1a, Langerin, S100

Declaration of patient consent : The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his/her consent for his/her images and other clinical information to be reported in the Journal. The patient understands that his/her name and initials will not be published, and due efforts will be made to conceal his/her identity, but anonymity cannot be guaranteed.

Conflict of Interest: None; **Source of Support:** None

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