Journal of Orthopedic Surgery and traumatology

case reports

Eosinophilic granuloma of the cervical spine in children: presentation of a 2 case and bibliographic review.

Garrido Rojo, R[1], Simon Perez V[2], Deserio Cost J[3], Garcia Chamorro M[4], Forts Lanzuela M[5], Bas Hermida P[6]

1 Reference author 1; Raquel Garrido Rojo, Orthopedic Surgery and Traumatology, University Hospital and La Fe Polytechnic, Valencia; raquelgr3293@gmail.com.

Summary: Introduction; Eosinophilic granuloma is a solitary and benign tumor that forms part of Langherhans cell histiocytosis. It mainly affects the child population and can affect any part of the bone skeleton. At the level of the spine, it is diagnosed because it appears as a "flat vertebra" in radiographic studies. Material and methods; We present case of a 4-year-old girl who presented neck pain and torticollis after a fall two months ago. In the imaging tests carried out, a flat vertebra was observed at the level of C3, compatible with an eosinophilic granuloma . Results; Given the absence of warning signs, it was decided to carry out a conservative treatment using Minerva indiana-type orthosis and periodic X-rays. After two months, a recovery of the vertebral height can be appreciated . Conclusion; It has been shown that eosinophilic granuloma is a benign entity that tends to resolve spontaneously, so the preferred treatment today is observation, with biopsy being necessary in very few cases.

Key words: “eosinophilic granuloma”, “histiocytosis”, “flat vertebra”, “cervical tumor”, “orthosis”, “observation”.

1. Introduction

Eosinophilic granuloma is a disease that is part of the Langerhans histiocytosis (LCH), along with Hans Schuller-Christian disease and Letterer -Siwe disease. It is a clonal proliferation of histiocytes that acts as a benign solitary tumor, constituting 60 to 80% of all LCH cases [1]. At the bone level it can affect any part of the skeleton, although it constitutes less than 28 % of all bone tumors. In 80% of cases it affects children and adolescents [1, 4].

The eosinophilic granuloma presents, in general, a benign behavior. It presents in a solitary form, most frequently affecting the skull, mandible, vertebrae and long bones, especially femur, humerus and clavicle. At the level of the spine, 6.5-25% of bone tumors correspond to eosinophilic granulomas. The most frequently affected is the thoracic spine, followed by the lumbar spine and the cervical spine [2].

Most of these injuries are incidentally identified. Signs and symptoms , if any, will depend on the spinal location of the lesion [3, 4]. At the cervical level, the most common symptoms include neck pain or stiff neck, pain on spinal palpation, and limited range of motion; however instability or neurological symptoms are rare.

Pathologically, there is destruction of bone tissue by proliferation of 42 histiocytes. Laboratory findings are usually nonspecific. Imaging studies 43 typically show an osteolytic area causing complete or incomplete collapse of the 44 vertebral body, known as the vertebra plana [4].

The differential diagnosis should be made with aneurysmal bone cyst, bone infarction, 46 metastasis, fibrous dysplasia, acute or chronic bone infection, and osteosarcoma [3]· 47

Multiple treatments are proposed in the literature such as observation, immobilization , 48 biopsy with or without bone graft, external with local radiotherapy, the use of chemotherapy 49 with methylprednisolone injection, as well as surgery in cases of instability [4, 5]. fifty

The prognosis of solitary eosinophilic granuloma is almost always favorable, since in 51 the majority of cases there is a complete restoration of the vertebral body [5] 52

The objective of this descriptive and retrospective study is to describe the symptoms, 53 diagnosis, and treatment of eosinophilic granuloma through the case of a 4-year-old girl 54 who presented a lesion at the level of the cervical spine. 55

56

2. Materials and methods 57

We present the case of a 4-year-old female patient who reported symptoms of neck pain 58 after a fall about two months earlier, persistent despite analgesic and anti-inflammatory 59 treatment . He did not present systemic or neurological symptoms. 60

On examination, she presented pain on palpation at the upper cervical level, associated 61 discomfort at the level of the paravertebral and trapezius musculature; He showed an attitude of neck extension 62 due to pain. The neurological examination was normal. 63

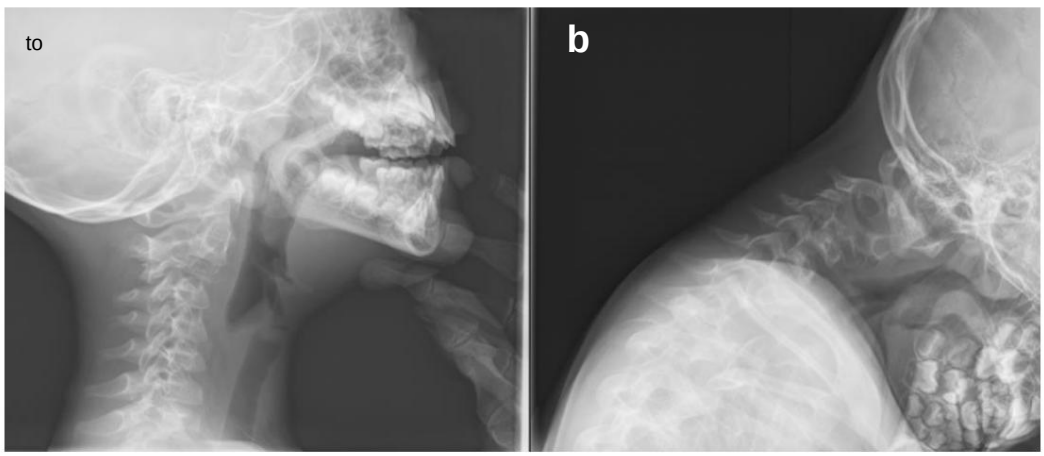
The complementary tests that were performed, including an abdominal ultrasound 64 and a complete blood analysis, were within normal limits. 65

In the simple x-ray of the cervical spine, a notable loss of homogeneous height of C3 ("flat vertebra") is observed , as a typical finding of eosinophilic granuloma. 67 The radiological study was completed with dynamic flexion and extension radiographs, 68 with a CT and an MRI.

69

70

71



72

Figure 1 (a and b). Dynamic lateral radiographs of the cervical spine: a loss of height of the C3 vertebral body is observed, 73

without added instability in flexion and extension. 74

75

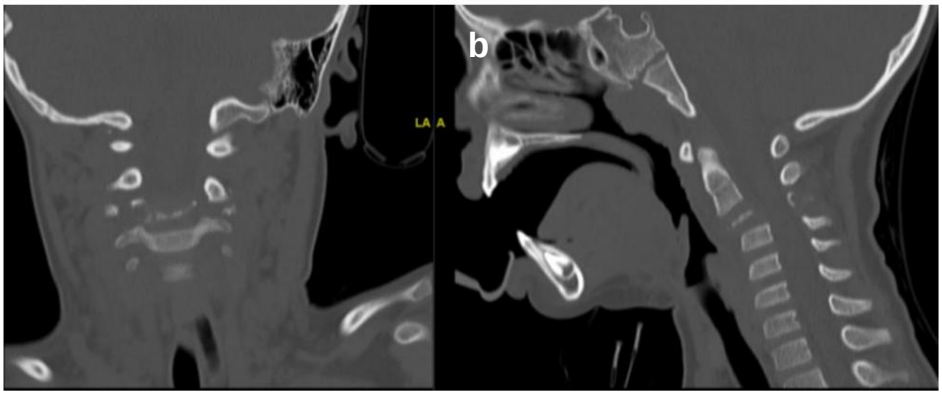


Figure 2 (a and b). CT of the cervical spine showing a lytic lesion of the vertebral body of C3 causing its flattening and collapse.

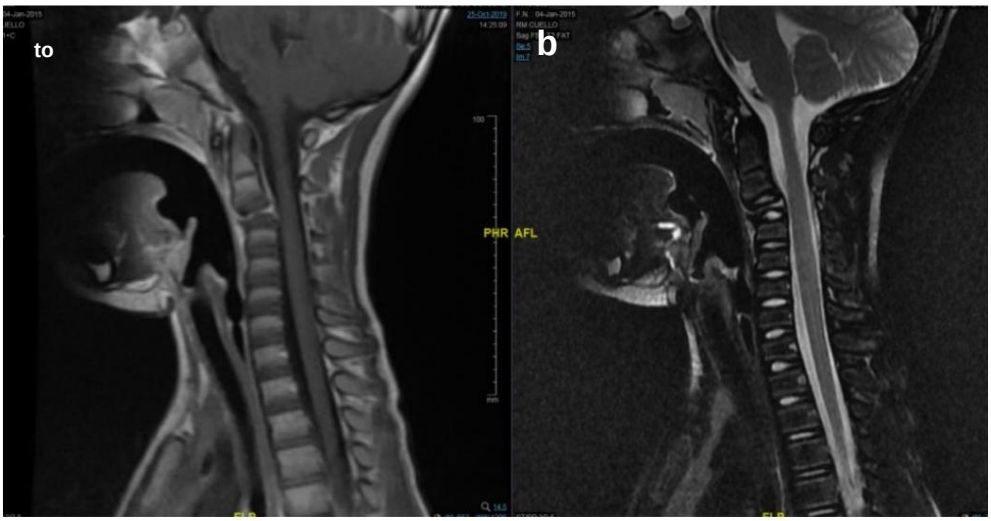


Figure 3 (a and b). T1 and T2-weighted MRI of the cervical spine, sagittal slices, showing flattening of the C3 vertebral body.

3. Results

Given the absence of criteria for spinal instability and the normality of the neurological examination, it was decided to perform a conservative treatment by means of immobilization with an adapted Minerva Indiana-type orthosis.

The control radiographs with the orthosis show the recovery of the physiological lordosis of the cervical segment, as well as a slight but progressive increase in the height of the C3 vertebral body in the radiographic controls of the following months. At all times , the patient remained asymptomatic, with normal neurological examinations.

Figure 4. Clinical images of the patient with the Minerva Indiana adapted orthosis.

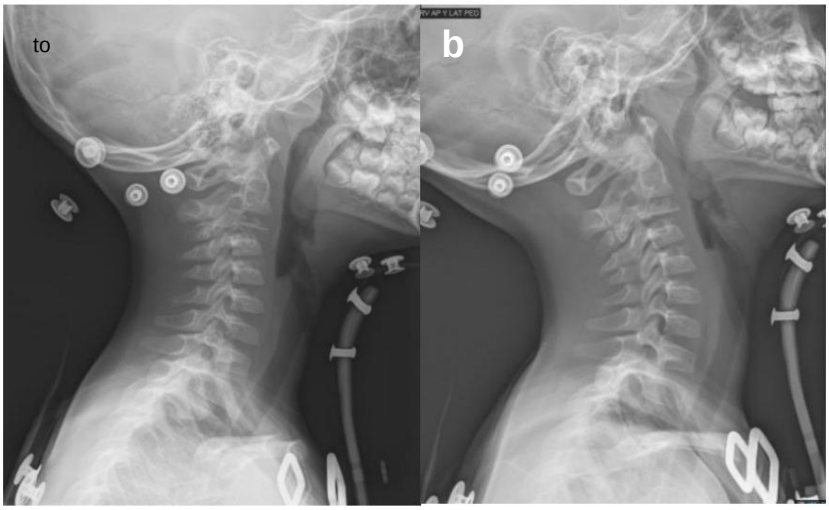


Figure 5. Lateral X-ray of the cervical spine with adapted orthosis (a) and control at two months (b): the restoration of the physiological cervical lordosis and a progressive recovery of the height of the C3 vertebral body can be seen.

4. Conclusion

The diagnosis of eosinophilic granuloma is based on its clinical and radiological characteristics . However, when these are ambiguous or the symptoms are severe, it is necessary to make a histological diagnosis by biopsy.

Multiple treatment options have been described, with satisfactory results and recurrence rates of less than 20%. Given its benign nature, its clinical course, and the fact that it mainly affects children, it is preferable to opt for conservative treatments that imply the least risk of complications. In this sense, since eosinophilic granuloma tends to resolve spontaneously, simple observation is the most recommended therapeutic strategy, although the time required for resolution is unpredictable.

Informed consent: Written informed consent was obtained from the parents of the 119 patient, a minor, for the publication of the case report and accompanying images. 120

Funding: This review did not receive external funding. 121

Conflicts of interest: The authors declare that there are no conflicts of interest. 122

123

124

Bibliography 125

126

1. Dhull AK, Aggarwal S, Kaushal V, Singh S. Into the wild world of eosinophilic granuloma. BMJ Case Rep. 2013; 2013: 127
bcr2013200522. 128
2. Fenoy AJ, Greenlee JD, Menezes AH, Donovan KA, Sato Y, Hitchon PW, et al. Primary bone tumors of the spine in children. J 129 Neurosurg. 2006; 105 (4 Suppl):
252-260. 130
3. Greenlee JD, Fenoy AJ, Donovan KA, Menezes AH. Eosinophilic granuloma in the pediatric spine. Pediatric Neurosurg. 2007; 43: 131 132
285-292.
4. Angelini A, et al. Current concepts for the diagnosis and management of eosinophilic granuloma of bone. J Orthop Traumatol. 133 134
Jun 2017; 18(2): 83–90.
5. Raab P, Hohmann F, Kuhl J, Krauspe R. Vertebral remodeling in eosinophilic granuloma of the spine. A long-term follow-up. 135 136
Spine. 1998; 23: 1351-1354.