Cervical Osteochondroma and Myelopathy in Children: A Case Report and Literature Review

Santiago Aguer, Javier Dal Lago, Matías L. Cullari, Facundo Aguirre, Eduardo Levy, Ruy Lloyd

Spinal Pathology Department, Hospital Británico de Buenos Aires, Autonomous City of Buenos Aires, Argentina.

ABSTRACT

Osteochondroma is the most common benign skeletal tumor, with the cervical spine being the most frequent site for spinal osteochondromas. Spinal exostoses typically arise from the lamina of cervical vertebrae. Although rare, cervical myelopathy can occur and may lead to significant neurological consequences. Surgical intervention is often considered for asymptomatic lesions to prevent potential neurological deterioration. We report the case of an 11-year-old patient with an incidentally discovered cervical osteochondroma originating from the left lamina of C4, causing spinal cord compression. Magnetic resonance imaging (MRI) of the cervical spine revealed a lesion protruding into the canal, leading to a reduction in canal diameter. The patient underwent surgical resection of the C4 posterior arch with decompression, performed without fusion. At two years post-surgery, no local recurrence or neurological symptoms were observed.

Keywords: Osteochondroma; spine; hereditary multiple exostoses; myelopathy; children.

Level of Evidence: IV

Osteocondroma cervical y mielopatía en niños: reporte de un caso y revisión de la bibliografía

RESUMEN

El osteocondroma es el tumor benigno más común del esqueleto. La columna cervical es la ubicación más frecuente para el osteocondroma espinal. Las exostosis espinales surgen, con más frecuencia, de las vértebras cervicales, aparecen típicamente en la lámina. Aunque la mielopatía cervical es un cuadro raro, puede tener serias consecuencias neurológicas. Si se descubren lesiones asintomáticas, se puede considerar la intervención quirúrgica para prevenir el deterioro neurológico. Se detectó incidentalmente un osteocondroma cervical en una paciente de 11 años. Este se originaba en la lámina de C4 y comprimía la médula espinal. La resonancia magnética de columna cervical mostró el osteocondroma en la lámina izquierda de C4 que sobresalía en el canal, lo que reducía su diámetro. La paciente fue sometida a una resección del arco posterior de C4 y a descompresión sin fusión. A los 2 años de la cirugía, no había evidencia de recurrencia local ni síntomas neurológicos. Palabras clave: Osteocondroma; columna vertebral; exostosis múltiple hereditaria; mielopatía: niños.

Nivel de Evidencia: IV

INTRODUCTION

Osteochondroma is the most common benign tumor of the skeletal system. It can occur as a solitary lesion or as part of an autosomal dominant hereditary syndrome known as hereditary multiple exostoses (HME). This condition primarily affects the growth plate, typically presenting during adolescence, and its progression halts upon reaching skeletal maturity. Generally, osteochondromas are asymptomatic and rarely undergo malignant transformation. They predominantly occur in the metaphyses of long bones, with spinal involvement being uncommon (1–4%). However, in cases of HME, spinal involvement increases to approximately 7–9%.¹ Among spinal osteochondromas, the cervical spine is most frequently affected, comprising 50-58% of cases, with 64% involving the posterior elements of the spine. Although rare, 0.5–1% of patients may experience progressive

Received on June 4th, 2024. Accepted after evaluation on July 27th, 2024 • Dr. MATÍAS L. CULLARI • matiaslcullari@gmail.com (D) https://orcid.org/0000-0002-6058-6686

How to cite this article: Aguer S, Dal Lago J, Cullari ML, Aguirre F, Levy E, Lloyd R. Cervical Osteochondroma and Myelopathy in Children: A Case Report and Literature Review. Rev Asoc Asoc Argent Ortop Traumatol 2024;89(6):651-656. https://doi.org/10.15417/issn.1852-7434.2024.89.6.1979

symptoms of myelopathy or radiculopathy, which, if left undiagnosed and untreated, can lead to severe complications.²

This report presents the case of an 11-year-old female patient with HME and cervical osteochondroma, who underwent surgery due to spinal cord compression. A literature review on this topic is also provided.

CLINICAL CASE

An 11-year-old girl diagnosed with HME had been under care at the Children's Orthopedic Service since 2010, presenting with multiple lesions on both arms and legs (Figure 1).



Figure 1. Radiographs of both shoulders, AP view (**A and B**), right ankle, AP view (**C**) and both knees, AP view (**D**). Multiple osteochondromatous lesions.

In May 2020, she was referred to the Spine Pathology Service after an incidental finding of a cervical osteochondroma on a routine computed tomography (CT) scan. At the time, the patient had no cervical symptoms, and neurological examination revealed normal findings, including present reflexes and negative Hoffman, Clonus, and Babinski signs. CT imaging showed an osteochondroma in the left lamina of C4, extending into the spinal canal (Figure 2). Additionally, a pedunculated osteochondroma in the posterior wall of T1 and an osteochondroma in the spinous process of C2 were identified, both extending into the spinal canal. Magnetic resonance imaging (MRI) confirmed that the C4 lesion reduced canal diameter and demonstrated a hyperintense signal on T2 sequences (Figure 3).

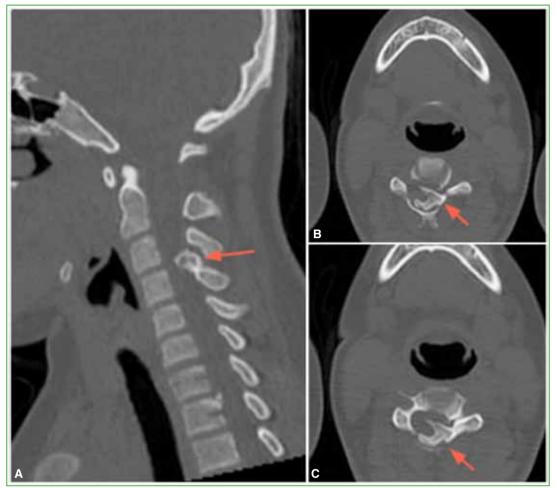


Figure 2. Computed tomography of the cervical spine, sagittal (**A**) and axial (**B and C**) sections. Osteochondroma of the left lamina of C4 with narrowing of the spinal canal (red arrow).

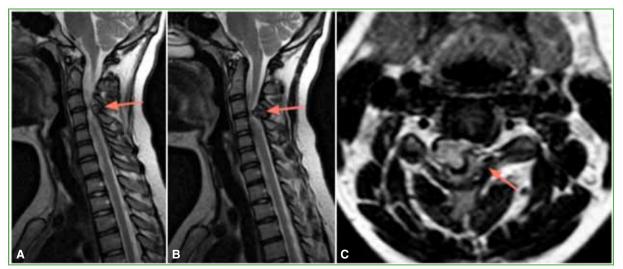


Figure 3. Magnetic resonance of cervical spine, sagittal (A and B) and axial (C) slices. Osteochondroma of the left lamina of C4 with narrowing of the spinal canal and hyperintense signal of the spinal cord at that level (red arrow).

S. Aguer et al.

The T1 and C2 osteochondromas did not exhibit spinal cord involvement on MRI. Subsensory evoked potentials from all four extremities indicated decreased amplitude. Given these findings, posterior cervical decompression and resection of the posterior arch of C4 were performed without instrumentation (Figure 4).

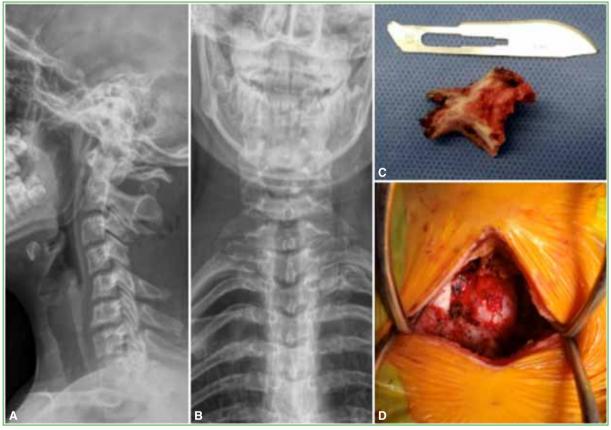


Figure 4. Anteroposterior and lateral radiographs of the cervical region in the immediate postoperative period (A and B) and image of the resection of the posterior arch of C4 (C and D).

The patient experienced no neurological complications and was discharged on postoperative day three. A rigid cervical collar was used for two months. Pathological examination confirmed the diagnosis of osteochondroma. Three months after surgery, the patient developed local kyphosis at the C4–C5 segment, which is currently being monitored. At two years postoperatively, there was no evidence of local recurrence on CT or MRI (Figure 5).

Both the T1 pedunculated osteochondroma and the finding in the spinous process of C2 remained stable, and conservative treatment was continued.

Currently, the patient is pain-free and actively participates in sports.

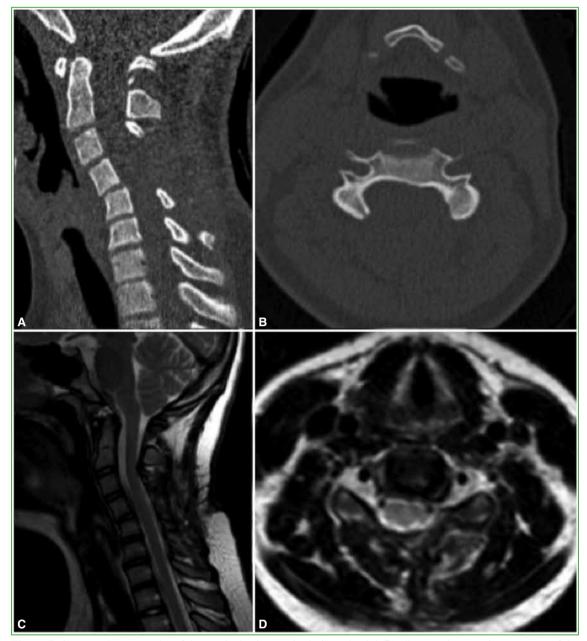


Figure 5. Computed tomography of the cervical spine, sagittal (A) and axial (B) slices, and magnetic resonance of the cervical spine, sagittal, T2 (C) and axial T2 (D) slices. Postoperative control at 24 months.

DISCUSSION

The first documented case of myelopathy associated with HME was published by Reid in 1843.³ In 1907, Ochsner and Rothstein described the first case of cervical myelopathy secondary to HME treated with decompression.⁴ Spinal osteochondromas most commonly affect the cervical region, followed by the thoracic and lumbar spine. Within the cervical spine, C2 (29%) and C5 (24%) are the most commonly affected levels, while C4 involvement, as seen in our patient, is less frequent (17%).^{1,5} These lesions often originate from the lamina and extend outward, rarely causing myelopathy. The incidence of myelopathy in such cases is low (0.5–1%) and

typically manifests in the second or third decade of life. Myelopathy in pediatric patients with HME is uncommon. Roach et al. evaluated 44 asymptomatic HME patients using MRI or CT and found that 38% had spinal osteochondromas, with 27% showing canal involvement.⁶ This highlights the potential underreporting of spinal lesions in HME. Based on these findings, annual spinal MRI is recommended for patients with HME, especially during puberty. In this case, the patient's cervical osteochondroma was incidentally discovered on a CT scan, as spinal monitoring had not been performed previously. For asymptomatic lesions, prophylactic surgery may be considered to prevent neurologic deterioration. Surgical options include laminoplasty, isolated laminectomy, or laminectomy with arthrodesis.⁷ During surgery, lesion location, resection and residual stability are critical considerations. Complete resection, including the cartilage cap, is essential to minimize recurrence risk associated with incomplete excision.⁸ Laminectomy is often the preferred approach, with the decision to include posterior arthrodesis made on a case-by-case basis to prevent deformities such as cervical kyphosis.^{9,10}

CONCLUSIONS

Although cervical myelopathy due to osteochondroma is rare, it can result in significant neurological complications in patients with HME. Annual spinal MRI is recommended for early detection of lesions. If surgery is required, laminectomy, with or without posterior fusion, is the preferred treatment option.

Conflict of interest: The authors declare no conflicts of interest.

S. Aguer ORCID ID: <u>https://orcid.org/0000-0001-9079-1966</u> J. Dal Lago ORCID ID: <u>https://orcid.org/0000-0002-0868-5505</u>

F. Aguirre ORCID ID: https://orcid.org/0009-0008-7148-9575

E. Levy ORCID ID: <u>https://orcid.org/0000-0002-9158-8617</u> R. Lloyd ORCID ID: <u>https://orcid.org/0000-0003-3897-3628</u>

REFERENCES

- Kamiya Y, Horii E, Sakai Y, Inoue H. Cervical cord compression in pediatrics with hereditary multiple exostoses: a report of two cases and review of the literature. *J Pediatr Orthop B* 2016;25(3):267-70. https://doi.org/10.1097/BPB.0000000000220
- Giudicissi-Filho M, de Holanda CV, Borba LA, Rassi-Neto A, Ribeiro CA, de Oliveira JG. Cervical spinal cord compression due to an osteochondroma in hereditary multiple exostosis: case report and review of the literature. *Surg Neurol* 2006;66 Suppl 3:S7-S11. https://doi.org/10.1016/j.surneu.2006.05.057
- 3. Reid J. Case of disease of the spinal cord from an exostosis of the second cervical vertebra. *Lond Edinb Mon J Med Sci* 1843;3:194-8.
- Ochsner EH, Rothstein T. XII. Multiple exostoses, including an exostosis within the spinal canal with surgical and neurological observations. *Ann Surg* 1907;46(4):608-16. https://doi.org/10.1097/00000658-190710000-00012
- 5. Burki V, So A, Aubry-Rozier B. Cervical myelopathy in hereditary multiple exostoses. *Joint Bone Spine* 2011;78(4):412-4. https://doi.org/10.1016/j.jbspin.2011.02.021
- Roach JW, Klatt JW, Faulkner ND. Involvement of the spine in patients with multiple hereditary exostoses. J Bone Joint Surg Am 2009;91(8):1942-8. https://doi.org/10.2106/JBJS.H.00762
- Fukushi R, Emori M, Iesato N, Kano M, Yamashita T. Osteochondroma causing cervical spinal cord compression. Skeletal Radiol 2017;46(8):1125-30. https://doi.org/10.1007/s00256-017-2633-6
- Fowler J, Takayanagi A, Siddiqi I, Ghanchi H, Siddiqi J, Veeravagu A, et al. Cervical osteochondroma: surgical planning. *Spinal Cord Ser Cases* 2020;6(1):44. https://doi.org/10.1038/s41394-020-0292-7
- Akhaddar A, Zyani M, Rharrassi I. Multiple hereditary exostoses with tetraparesis due to cervical spine osteochondroma. World Neurosurg 2018;116:247-8. https://doi.org/10.1016/j.wneu.2018.05.078
- Veeravagu A, Li A, Shuer LM, Desai AM. Cervical Osteochondroma causing myelopathy in adults: Management considerations and literature review. *World Neurosurg* 2017;97:752.e5-752.e13. https://doi.org/10.1016/j.wneu.2016.10.061