A tale about an unusual cause of pygalgia in ankylosing spondyloarthritis

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August 30, 2022

Abstract

Neurinomas are tumors of Schwann cells of the peripheral nerve sheath. Sacral location is rarely reported especially in spondyloarthritis patients. Herein, we report a case of uncommon pygalgia in a 25-year-old man with history of a non-radiographic axial spondyloarthritis and in whom the diagnosis of sacral neurinoma was established.

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- **Prof Imene Gharsallah** : Professor, head of Rheumatology department , Military Hospital of Tunis, Tunisia**Email** : imengharsallah@hotmail.fr
- **Prof Bassem Louzir :** Professor, head of internal medicine department , Military Hospital of Tunis, Tunisia**Email:**louzir.bassem@yahoo.fr**Declarations section:**

- **Consent for publication:** Patient was informed and fully consented to publication.
- **Availability of data and material:** The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.
- **Competing interests : None to declare**
- **Funding : None**
- **Pharmaceutical / industry support: None****ABSTRACT:** Neurinomas are tumors of Schwann cells of the peripheral nerve sheath. Sacral location is rarely reported especially in spondyloarthritis patients. Herein, we report a case of uncommon pygalgia in a 25-year-old man with history of a non-radiographic axial spondyloarthritis and in whom the diagnosis of sacral neurinoma was established.**KEY WORDS:** Neurinoma; Schwannoma; spondyloarthritis; pygalgia**KEY CLINICAL MESSAGE** :

- Neurinoma of peripheral nerve roots is a rare entity.
- Neurinoma and spondyloarthritis is an unusual association
- Differential diagnosis of sacroiliac joint pain is made with S1 nerve roots tumors such as Neurinoma.**PRESENTATION:**A 25-year-old man with history of a non-radiographic axial spondyloarthritis admitted for persistent right buttock pain. The sacroiliac radiographs showed a slight enlargement of the first right sacral foramen (Figure A). An MRI of the lumbosacral spine and the sacroiliacs was done in the background to objectify a sacroiliitis in flare. However, the MRI had shown a soft tissue mass lateral to the left S1/S2 foramina. This was consistent with a neurinoma measuring 18mm * 22 mm. The patient was operated on with total resection of the neuroma. Histological findings have confirmed the diagnosis (Figure F). The evolution was good with disappearance of pygalgias.**DISCUSSION:**The incidence of spinal schwannoma, according to recent studies, varies between 3 and 4 cases / 1,000 000 people per year [1]. MRI is the gold standard for the diagnosis of spinal schwannoma, it allows structural and spatial analysis of the lesion [2]. The average age of the appearance of neurinomas is between 30 and 50 years old. In our observation, the age was younger than usual. The most clinical signs are the pain of both the spine and root system reflecting the lesion syndrome. Surgery is the gold standard of treatment for spinal schwannoma [3].**REFERENCES:**[1] Emel E, Abdallah A, Sofuoglu OE, Ofluoglu AE, Gunes M, Guler B, Bilgic B. Long-term Surgical Outcomes of Spinal Schwannomas: Retrospective Analysis of 49 Consecutive Cases. Turk Neurosurg. 2017;27(2):217-225. doi: 10.5137/1019-5149.JTN.15678-15.1. [2] Amezyane T, Pouit B, Bassou D, Lecoules S, Desramé J, Blade JS, Béchade D, Algayres JP. Une cause rare de lombosciatique [A rare cause of sciatica]. Rev Med Interne. 2006 ;27(6):494-6. doi: 10.1016/j.revmed.2005.10.001. [3] Chang, Ung-Kyu, et al. Radiosurgery Using the Cyberknife for Benign Spinal Tumors: Korea Cancer Center Hospital Experience. Journal of Neuro-oncology, vol. 101, no. 1, 2011, pp. 91-9.**FIGURE'S LEGEND:**Figure A: Enlargement of the first right sacral foramen on a pelvic x ray. Figure B, C: Frontal T1 and T2 sacroiliac sequences showing targeted appearance of the mass consistent with neurinoma' diagnosis. Figure D, E: Sagittal lumbar spine T1, T2 and STIR weighted sequences showing high signal soft tissue mass on the right S1 nerve root on T2 and STIR weighted sequences and low signal on T1 weighted sequences. Figure F: Spindle cell tumor proliferation showing characteristic nuclear palisades.