



A Rare Case of Cervicalgia

Un caso raro de cervicalgia

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Abstract

Objective To underline the importance of a detailed diagnosis of pain and perform self-criticism regarding the delay in diagnosis of a case of mechanical cervicalgia.

Material and methods We present the case of a patient with sudden neck pain, with normal X-rays. The magnetic resonance imaging (MRI) scan was apparently unremarkable, and the condition was treated as mechanical cervicalgia. After several consultations in the Emergency Department, a new X-ray was performed, which showed an anomaly between the first vertebrae and is accompanied by a computed tomography (CT) scan that revealed a fracture of the odontoid apophysis and destruction of the second cervical vertebra of metastatic origin.

Results The patient was treated with radiotherapy plus cervical collar and several cycles of chemotherapy, and died two years later.

Discussion The cancers that most frequently metastasize to the spine are those of the breast, lung and prostate, with the most frequent location being thoracic (70%), lumbar (20%), and, finally, cervical (10%). When the tumor is discovered as a metastasis, as in our case, these patients have a high mortality rate. To our knowledge, few cases of neoplasms discovered as pathological fractures of the odontoid apophysis have been described in the literature.

Conclusion Non-mechanical cervicalgia due to metastasis of the odontoid apophysis as a result of an undiagnosed lung neoplasm is a rare case in the literature and a complex diagnosis, in which a detailed clinical history of the evolution of pain and the

Keywords

- ▶ cervicalgia
- ▶ metastasis
- ▶ odontoid apophysis
- ▶ delay in diagnosis

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presence of red flags are of vital importance for its suspicion and rapid diagnosis, through techniques such as MRI.

Resumen

Objetivo Subrayar la importancia del diagnóstico detallado del dolor y realizar autocrítica por la tardanza diagnóstica de una cervicalgia mecánica.

Material y Métodos Se presenta una paciente con dolor súbito en el cuello, con radiografías y exploración normales. La resonancia magnética (RM) resultó aparentemente anodina y se trató como cervicalgia mecánica. Tras varias consultas con el Servicio de Urgencias, se realizó una radiografía que informó de anomalía entre las primeras vértebras y se amplió con una tomografía computarizada (TC) que reveló fractura de odontoides y destrucción de la segunda cervical de origen metastásico.

Resultados La paciente fue tratada con radioterapia más collarín cervical y varios ciclos de quimioterapia, y falleció a los dos años.

Discusión Los cánceres que más frecuentemente metastatizan en la columna vertebral son los de mama, pulmón y próstata, siendo las localizaciones más frecuentes la torácica (70%), la lumbar (20%) y, por último, la cervical (10%). Cuando el tumor se descubre como metástasis, como en nuestro caso, estos pacientes tienen una alta tasa de mortalidad. Según nuestro conocimiento, en la literatura hay pocos casos descritos de neoplasias descubiertas como fractura patológica de odontoides.

Conclusión El dolor cervical no mecánico debido a una metástasis en odontoides, a consecuencia de una neoplasia de pulmón no diagnosticada, es un caso raro en la literatura y de complejo diagnóstico, en que una historia clínica detallada de la evolución del dolor y la presencia de signos de alarma son de vital importancia para su sospecha y rápido diagnóstico, mediante técnicas como la RM.

Palabras Clave

- ▶ cervicalgia
- ▶ metástasis
- ▶ odontoides
- ▶ retraso diagnóstico

Introduction

The spine is the most common location for bone metastases, since 5% to 10% of all cancer patients have spinal metastases.¹ These are more frequent in patients with breast, lung and prostate cancer, and have a male predominance. They are not the most frequent in terms of general numbers, though. The most frequent symptomatic spinal metastases are thoracic (70%), followed by lumbar (20%), and, finally, cervical (10%).^{1,2}

We herein present a cause of non-mechanical cervicalgia due to a C2 metastasis, as a result of an undiagnosed lung neoplasm. We want to underline the complexity of the diagnostic imaging and the importance of a detailed clinical history of the evolution of pain and the presence of and clinical warning signs and symptoms, in addition to perform self-criticism for the delay in diagnosing this complicated case of cervicalgia, hoping that it will serve for our improvement in future cases.

Material and methods

We herein present a clinical case of rare incidence which occurred in our hospital that we report for its disclosure.

A 59-year-old woman presented to the Emergency Department with sudden neck pain, without weight loss, compartment syndrome or associated radiculopathy, and

without the presentation of masses, abnormal enlargements or adenopathies. The patient did not have a personal history of interest, was not a smoker, did not report any known drug allergies, and was afebrile and with stable vital signs. The physical examination was normal, with no pain on palpation of the cervical spinous processes or paravertebral muscles, preserved bilateral reflexes, and no changes in strength or sensitivity. After these findings, she was diagnosed with mechanical cervicalgia, so we decided on the conservative treatment with home-based analgesia.

A week later, the patient returned to the Emergency Department due to the non-remission of the symptoms, reporting on this occasion that the cervical pain increased when standing and was relieved when lying down. Given the persistence of the pain, an imaging study was indicated, and the radiographs of the cervical spine performed showed a decrease in the C5-C6 disc space, but no additional alterations. On this occasion, the treatment was increased by going up a step in the analgesic scale, and tramadol was started.

The patient went to the Emergency Department a third time, and was referred to the Traumatology Department. The diagnostic imaging study was extended, with the performance of magnetic resonance imaging (MRI) scans and an electroneurogram (ENG). The MRI revealed the existence of impingement of C5-C6 with retrolisthesis, but without involvement of the spinal canal, and no axonal repercussions of



Fig. 1 Magnetic resonance imaging scan showing no findings suggestive of pathology in the C1-C2 vertebrae.

the C5-C6 myotomes were found on the ENG. Given the apparent conventional cervicalgia, the patient was referred to the Rehabilitation Service, which occurred eight months after the onset of symptoms (► **Figure 1**).

After the first sessions of rehabilitative treatment and given that the symptoms did not improve, we decided to repeat the X-rays, which reported an anomaly between the first and second cervical vertebrae that had not been previously observed (► **Figure 2A**). Given the findings, a computed tomography (CT) scan was requested, which revealed a fracture of the odontoid apophysis with impaction on the clivus, a fracture of the anterior arch of the body of C2, a soft-tissue mass, and moth-eaten pattern of bone destruction (► **Figure 2B**).

Given this finding, an extension study was carried out, showing a pulmonary mass suggestive of primary pulmonary neoplasia and metastatic involvement of the right hepatic lobe, both adrenal glands, and the D2 and D3 vertebral bodies. The biopsy confirmed the diagnosis of poorly-differentiated carcinoma with a mutation of the epidermal growth factor receptor (EGFR).

The patient began oncological treatment with erlotinib and cervical mass radiotherapy, in addition to zoledronic acid and pain management, with good clinical and radiological response at the pulmonary level. One year later, she presented brain metastases; we ruled out radiotherapy and decided to start therapy with bevacizumab, which was not favorable and, unfortunately, she died 21 months after the diagnosis.

Discussion

In the spine, the least frequent vertebrae regarding the appearance of metastases are the cervical ones, which only

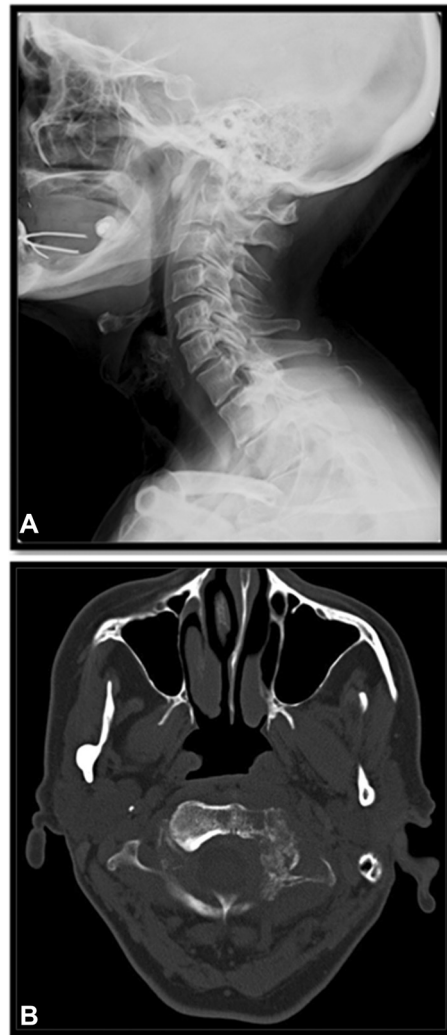


Fig. 2 (A) Simple radiograph taken six months after the previous radiographs, in which alterations in the first cervical vertebrae were reported for the first time. (B) Diagnostic CT scan performed at the same time as the second radiographs.

account to up to 10% of the cases.² The lesions can be osteoblastic or osteolytic and, in order to identify them on a plain radiograph study, between 30% and 50% of the vertebra must be affected.³ Some authors⁴ state that, in 1 out of every 10 cases of spinal metastases, the patient had not yet been diagnosed with cancer, becoming a patient with high mortality because of the advanced state of the disease in which they are due to the delay in diagnosis. A total of 60% of cervical spine metastases are caused by a breast, lung or prostate tumor.⁵ Despite the percentage that metastasizes to the cervical spine, little importance has been given to odontoid apophysis metastases;^{6,7} to our knowledge, there are few cases reported in the literature,¹ and the diagnosis is challenging.

Cervicalgia is pain in the cervical region that can spread to the neck, head or upper extremity, which limits movement and can be accompanied by neurological dysfunction (1% of cases). It is a very frequent pain, generally a consequence of muscular overload or other traumatic neuromuscular injuries, such as “cervical whiplash”. Back pain in general is

Table 1 Differential diagnosis of mechanical and inflammatory pain¹²

Cervicalgia	Mechanical	Non-mechanical
Characteristic	<ul style="list-style-type: none"> • Uni- or bilateral. • Accompanied by headaches. • Non-constant pain predominantly in the evening. • Worsens with exercise and improves with rest. 	<ul style="list-style-type: none"> • Uni- or bilateral. • Accompanied by instability. • Constant pain predominantly in the morning. • Worsens with exercise, but does not improve with rest. Affects with night rest.
Causes	<ul style="list-style-type: none"> • Structural alterations (bone, ligamentous and soft-tissue injuries), degenerative pathology, rheumatological alterations... 	<ul style="list-style-type: none"> • Vertebral tumor invasion or destruction of the vertebra. • Pathological fracture.
Beginning	<ul style="list-style-type: none"> • Insidious onset with slow worsening. • Acute onset related to major trauma. 	<ul style="list-style-type: none"> • Insidious onset with rapidly progressive worsening. • Acute onset after minimal trauma.
Age	<ul style="list-style-type: none"> • Elderly patients in relation to degenerative changes. 	<ul style="list-style-type: none"> • Patients of any age presenting red flags.
Radiological diagnosis	<ul style="list-style-type: none"> • X-ray after 6-8 weeks of conservative treatment without improvement. • Computed tomography/Magnetic resonance imaging scans only if there is emergence of neurological dysfunction or fracture. 	<ul style="list-style-type: none"> • X-ray at the onset of symptoms, ± magnetic resonance imaging scan. • Complete neurological examination.

estimated to affect four out of five adults. Although the most frequent is that of the lower back, cervical pain has an exceptional prevalence between 10% and 13%, appearing at some point in life in up to 70% of the population and causing between 11% and 14% of cases of sick leave.⁸ It generally responds well to the conservative treatment, but if it persists throughout time or worsens, it may be a sign of something more serious.^{9,10}

After reaching the age of 40, it is normal to begin to notice some stiffness in the cervical spine, caused by osteoarthritis and its consequent muscle tension. In fact, the facets and interapophyseal joint capsules are the cause of 25% to 75% of the cases of idiopathic cervical pain. In addition to these, in the cervical spine there are multiple structures that can result in cervical pain, such as ligaments, nerve roots, paravertebral muscles and the intervertebral disc, the latter causing pain indirectly due to irritation of nerve structures

or as a result of the instability associated with its herniation.⁹⁻¹²

Isolated or simple cervicalgia is usually located in the posterior paramedian region of the cervical musculature, radiating towards the occiput, shoulder or periscapular region, with insidious onset, without metameric distribution and associated with heat, tingling and even headache. There is no metameric distribution, and cervical rotation may be severely limited. When the pain extends through the territory of a peripheral nerve, it is a radiculopathy, and patients generally report acute pain and a tingling and burning sensation, and there may also be changes in strength and sensitivity.

It is important to know that cervicalgia can be mechanical (80%) or inflammatory (the remaining 20%), as a result of tumoral, inflammatory or infectious processes (→ **Table 1**; → **Table 2**).

Table 2 Causes of cervicalgia^{10,11}

Mechanical pain	Irradiated	Non-segmental
		Segmental: radiculopathies
Inflammatory pain	Rheumatic diseases: RA, AS and other spondyloarthropathies, juvenile chronic osteoarthritis, polymyalgia rheumatica, polymyositis, Forestier-Rotes Querol disease, juvenile ankylosing hyperostosis	
	Tumors (primary or metastatic): of the prostate, breast, lung, thyroid...	
	Infections: discitis (<i>Staphylococcus aureus</i> , <i>Mycobacterium</i> ...), osteomyelitis, meningitis, herpes zoster, Lyme disease...	
Referred pain	Abdominal and diaphragmatic diseases (gallbladder, subphrenic abscess, pancreas, hiatal hernia, peptic ulcer), aortic aneurysm, ischemic heart disease, vertex lung tumors, vertebral basilar insufficiency, acromioclavicular pathology, temporomandibular pathology, thoracic outlet syndrome.	

Table 3 Nurick classification of the disability caused by cervical myelopathy¹⁵

Grade I	No difficulty in walking.
Grade II	Gait difficulties that do not affect occupational activities.
Grade III	Gait difficulties that limit occupational activities.
Grade IV	Walking tolerated with assistance.
Grade V	Unable to walk. Bedridden.

Depending on the duration, cervicalgia can be acute (less than seven days), subacute (seven days to seven weeks), and chronic (more than seven weeks). One of the most frequent causes of acute pain is cervical sprain and the posttraumatic cervicalgia secondary to a traffic accident known as “cervical whiplash”.¹⁰ Most patients recover before 6 weeks, but between 10% and 15% of the cases become chronic, the latter being more frequent in middle-aged women and related to highly-demanding jobs. Certain symptoms that may indicate a more serious injury are the appearance of pain disproportionate to the trauma that occurred, signs of neurological involvement or loss of strength/sensitivity, and night pain or pain associated with chest pressure. No clear correlation has

been found between the degree of cervicoarthrosis and the intensity of the pain, but it has been observed that neurotic personality and the presence of depressive symptoms are factors that increase the probability of recurrence of cervical pain.^{13,14}

As in everything in medicine, it is essential to carry out a correct history and physical examination to help in the primary classification of patients. It is vitally important to take a good history, investigate personal history (including age, profession and sports, in addition to inquiring about possible mental disorders) and family history, and ask about the characteristics of the pain (acute or latent onset, acute evolution, chronic or recurrent, inflammatory or mechanical, or if there is root involvement). We should ask in the same way about the accompanying symptoms and signs, such as headaches, visual disturbances, dizziness and/or tinnitus, syncope caused by movements, fever or constitutional symptoms. The degree of functional limitation is determined by the Nurick classification according to the gait disturbance and its occupational consequences.¹⁵ (► **Table 3**)

Red flags are clinical signs and symptoms that are considered risk indicators for serious underlying disease. Many clinical guidelines recommend the use of red flags as a screening method to detect severe spinal pathology, since up to 4% of the cases of cervicalgia observed in primary care

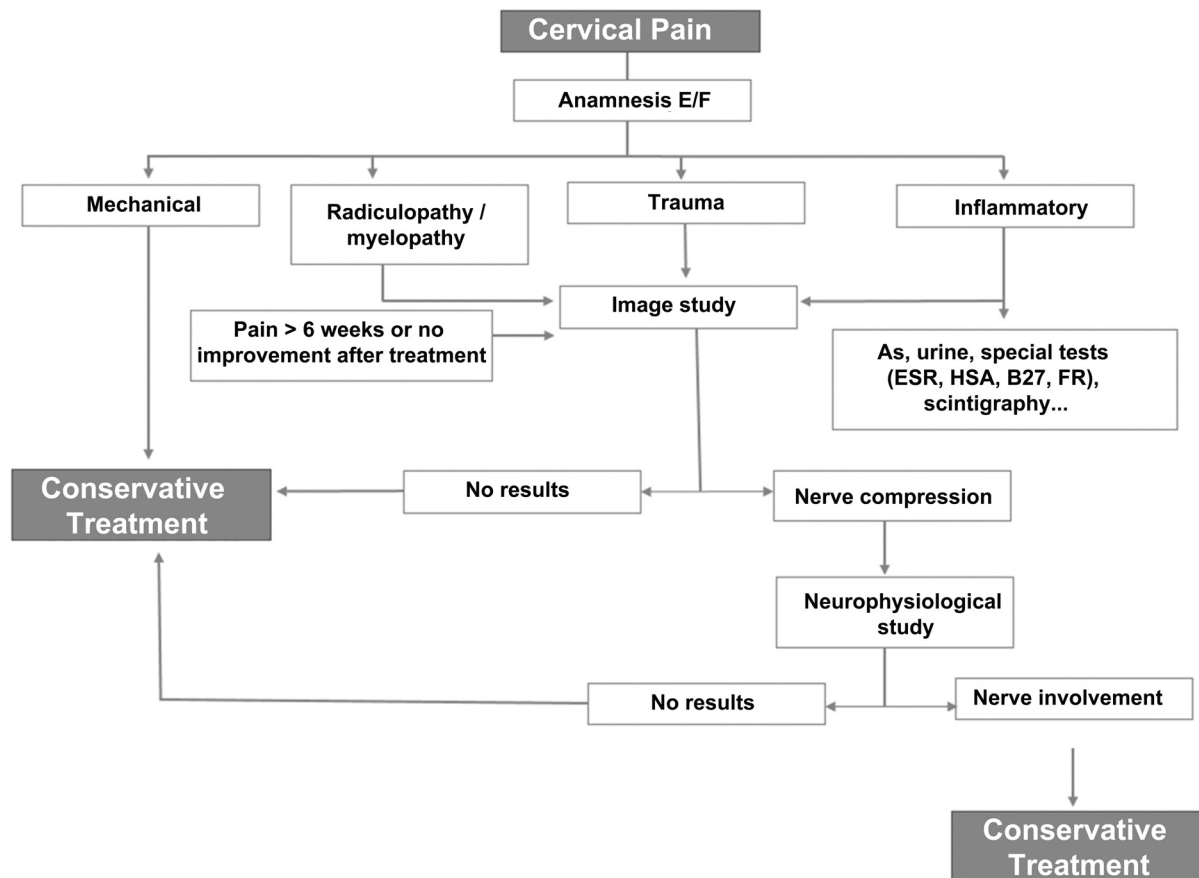


Diagram 1. Algorithm on how to act against cervicalgia¹⁰⁻¹².

and up to almost 6% of those observed in the Emergency Department result in important pathology.¹⁶ In a study by Shaw et al.,¹⁶ a widely variable sensitivity was observed regarding the red flags studied, from 62% of patients who presented poor general condition, 38% who had flank pain or the 35.8% who experienced urinary symptoms, up to the 1.1% of cases herpes zoster or unexplained weight loss. On the contrary, all the red flags studied by them showed a specificity, higher than 90%.¹⁶ Despite the fact that the study¹⁶ did show a higher incidence of serious pathology in the presence of red flags, it must be said that their usefulness is still somewhat uncertain, given their current poor definition and non-specificity.

The basic complementary test with which we begin our diagnostic study is the simple radiograph of the cervical spine in two projections (anteroposterior and lateral). If the pain begins associated with trauma or it is pain with inflammatory characteristics, the radiographic study is performed from the first moment. In the absence of these, the indications to perform radiographs are the presence of the aforementioned red flags, such as:^{10,16}

- Age over 70 years, onset of symptoms before 20 years or after 55 years, poor general condition, fever, involuntary weight loss, insidious onset, impaired night rest.
- Personal history of tuberculosis, abdominal aortic aneurysm, nephrolithiasis, recent bacterial infection, immunosuppression, pregnancy, cancer, parenteral drug abuse (PDA), rheumatoid arthritis (RA), osteoporosis or pathological fracture, spinal Paget disease, herpes zoster, major trauma, recent history of spinal intervention, carriage of spinal instrumentation.
- Iatrogenic: prolonged use of corticosteroids, anticoagulation.
- Physical examination revealing intractable pain, constant and progressive non-mechanical pain, morning stiffness for more than 30 minutes, saddle anesthesia, progressive changes in strength or gait, progressive changes in sensation, anal sphincter incontinence, and improvement in pain after exercise.
- Associated symptoms: urinary symptoms, urinary retention or incontinence, chest or abdominal pain, skin changes, and colitis.

In the presence of red flags, the indication is to start directly with an imaging study complementary to the physical examination. Sometimes, when serious pathology is suspected, the recommendation is to start directly with a more exhaustive imaging study, such as MRI, as indicated by Childress and Stuek¹⁷ in a recent article, since plain radiography has a low sensitivity for these types of clinical presentations.

The presence of neurological symptoms makes it necessary to request an MRI, since it is the technique that best visualizes the soft tissues and intraspinal pathology. Protrusions or extrusions of the intervertebral disc can be detected in up to 30% of asymptomatic subjects. Computed tomography is useful to extend the radiographic study, because it

enables a better evaluation of the bone and adjacent soft tissues. The ENG is used to locate the affected roots or nerves.

As a summary and in an orderly manner, an algorithm on how to act against cervicalgia¹⁰ is presented in ►**Diagram 1**.

Conclusion

Despite the lower percentage of cervical metastases compared to the rest of the spine, we must not forget about them or underestimate this axial location. The pain pattern ranges from the typical mechanical to the inflammatory one, and an exhaustive and well-detailed clinical history is essential for a correct and rapid diagnosis. The presence of red flags at the beginning of the picture is an indication to perform an MRI, which continues to be the gold standard for the detection of bone metastases. As a personal criticism, we recognize that in the first MRI, signs of malignant pathology were already present, but this was only observed after the images had been studied in greater detail.

Conflict of interests

The authors have no conflict of interests to declare.

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