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CASE REPORT

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Cervicothoracic Myelopathy Secondary to Mycetoma: Case Report and Literature Review

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ABSTRACT

Mycetoma is a chronic infection of the skin and underlying tissues caused by fungi or bacteria, it presents with multiple fistulous tracts, it is endemic in tropical and subtropical regions 15º to the south and 30º to the north of latitude, this region is known as the mycetoma belt. , the infection is inoculated after trauma, the objective of management is to eradicate the infection and prevent infiltrative cases (for example invasion of bone tissue), the case presented is a 29-year-old female, with secondary vertebral destruction syndrome a dorsal cervical thoracic mycetoma without microbiological isolation, this being a rare entity, which was managed in an interdisciplinary manner with empirical antimicrobials and posterior cervical decompressive surgery. Currently, the patient is five months postoperative with no cervical pain, and adequate neurological status to be able to carry out activities of daily living.

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Introduction

Mycetoma is a chronic granulomatous condition which can be caused by aerobic bacteria or by true fungi which are found in tropical and subtropical regions, at the national level most of the reports are in Michoacán, Oaxaca, Jalisco, Veracruz, Nuevo León, among other states. The most frequent microorganism is Nocardia brasiliensis and Actinomadura madurae in 78-85%, its mechanism of infection is by direct contact on a skin lesion, which allows the invasion of the causal agent. The typical presentation is in male agricultural worker populations at the level of the lower extremities, the cervicothoracic presentation ranges in 1.5-2.5% which, if not treated, can progress and infiltrate the spine and nervous system. Due to the morbidity and mortality caused by this pathology, it is necessary to carry out a timely diagnosis and treatment. The etiological diagnosis is made by means of culture, serology, and histopathology. Among the differentials, tuberculosis, coccidioidomycosis, botryomycosis, sporotrichosis, and neoplasms must be ruled out. The treatment for non-invasive cases is medical and it is recommended to continue it for 6 months after clinical and bacteriological remission, for cases where they present infiltrative lesions to the spine and spinal canal, surgical management with debridement and decompression is recommended in case of neurological compromise [1-3]. Due to the low frequency of this entity, it was decided to carry out this case report and literature review [4-8].

Case

A 29-year-old female, lives in a rural area and works in cattle farming, has a history of non-infiltrating dorsal thoracic cutaneous mycetoma of 2 years of evolution without medical management. She began her condition on July 13, 2021 with a progressive decrease in strength in the lower extremities and ataxia. On July 23, she is referred to our institution for paraplegia and urinary and fecal incontinence.

Presents a 32 x 28 cm lesion in the dorsal cervicothoracic region with multiple fistulous tracts of 3 x 3 mm in diameter with little outflow of straw-colored material (Figure 1), cervical mobility arches without compromise, bilateral strength of C5-C6 normal , C7-T1 2/5 and L2-S1 0/5 bilateral, paresthesias from C7 to distal bilateral, anal tone, and negative bulbocavernosus; severe cervical myelopathy secondary to infiltrating mycetoma is suspected, we applied the Nurick scale (IN) (Table 1) and the modified scale of the Japanese Orthopedic Association (mJOA) (Table 2) where grade 5 was obtained of IN and 6 points of mJOA [9,10].

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Figure 1: Plate of 32 x 28 cm in the dorsal cervicothoracic region with multiple fistulous tracts of 3 x 3 mm in diameter

Tabla 1: Sistema de clasificación de Nurick para la mielopatía cervical degenerativa.

Grado	
0	No hay síntomas de la raíz o de la médula
I	Signos o síntomas de las raíces. No hay evidencia de afectación de la médula
II	Signos de afectación de la médula. Marcha normal
III	Anomalía de la marcha. Capacidad para trabajar
IV	La anomalía de la marcha impide el empleo
V	Puede deambular sólo con ayuda
VI	En silla de ruedas o postrado en cama

Table 1: Nurick scale

Tabla 2: La escala modificada de la Asociación de Ortopedia Japonesa.

Valoración de la disfunción motora de la extremidad superior
0 – Incapacidad de mover las manos
1 – Incapacidad para comer con una cuchara, pero capaz de mover las manos
2 – Incapacidad para abotonarse la camisa, pero puede comer con una cuchara
3 – Es capaz de abotonarse la camisa con gran dificultad
4 – Es capaz de abotonarse la camisa con una ligera dificultad
5 – Sin disfunción
Valoración de la disfunción motora de la extremidad inferior
0 – Pérdida total de la función motora y sensorial
1 – Preservación sensorial sin capacidad de mover las piernas
2 – Puede mover las piernas, pero no puede caminar
3 – Capaz de caminar en un piso plano con una ayuda para caminar (bastón o muleta)
4 – Puede subir y/o bajar escaleras con barandal
5 – Falta de estabilidad de moderada a significativa, pero es capaz de subir y/o bajar escaleras sin barandal
6 – Leve falta de estabilidad, pero camina con un balanceo suave sin ayuda
7 – Sin disfunción
Valoración de la disfunción sensorial de las extremidades superiores
0 – Pérdida completa de la sensibilidad de la mano
1 – Pérdida sensorial severa o dolor
2 – Pérdida sensorial leve
3 – Sin pérdida sensorial
Valoración de la disfunción de los esfínteres
0 – Incapacidad de orinar voluntariamente
1 – Dificultad marcada con la micción
2 – Dificultad leve a moderada con la micción
3 – Micción normal

Table 2: Modified Scale of the Japanese Orthopedic Association

We request simple radiographs, CT and simple and contrasted magnetic resonance imaging of the cervicothoracic spine: AP and LATERAL CERVICAL SPINE: Decreased density of C6-T1 bodies.

CT of the Cervicothoracic Spine and Thorax: 7 cm tumor in the left pulmonary apex which infiltrates the epidural space from C6-T2 through left junctional foramens.

MRI of the Cervicothoracic Spine: Osteolysis of the body of T1 is observed, epidural collection of C6-T2 which lateralize the dural sac and spinal cord to the right, in addition to generating bilateral root compression and multiple paravertebral collections (Figure 2)

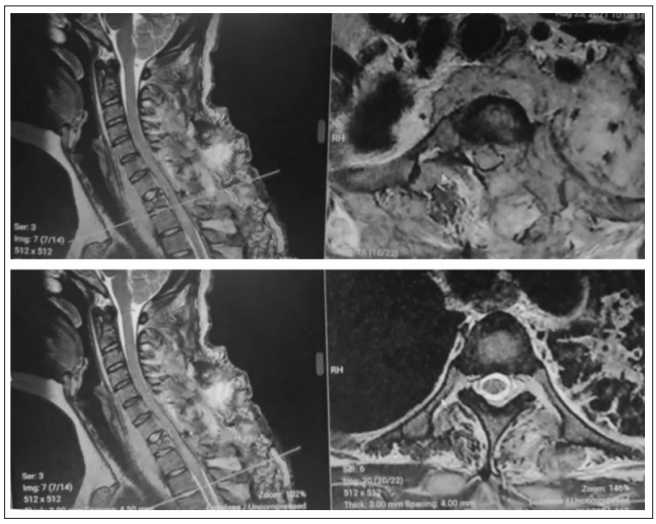

Figure 2: Pathological fracture of the body of T1, epidural collection of C6-T2

Due to the history of non-infiltrating dorsal thoracic cutaneous mycetoma without medical management and the clinical and radiological findings, we suspected a syndrome of vertebral destruction in T1 of infectious origin with severe cervical myelopathy, for which multidisciplinary management was carried out by dermatology, infectology, internal medicine and orthopedics; On the part of dermatology and infectology, dapsone, trimethoprim and sulfamethoxazole were started orally, our department scheduled C6-T1 left laminectomies, debridement of the epidural and paravertebral space, a rigid collar was placed for 3 months (Figure 3), the microbiology reports were negative and histopathology reports concluded leukocytoclastic vasculitis and chronic inflammation, in his last physical examination (5 months postoperative) he presented a posterior cervical scar without complications, remission of the fistulous tracts in 100%, bilateral strength of C5-T1 4 /5 and L2-L5 4/5, S1 3/5, Normosthetic from C7 to bilateral distal, without compromise of sphincters.

Figure 3: Posterior cervical approach with left C6-T1 laminectomies

Discussion

The first case of mycetoma was described in India in 1714 by French missionaries who called it "fourmilière des pieds" which means foot anthill in French, in 1860 Henry Van Carter introduced the term mycetoma which referred to an infection caused by fungi, a term that is still valid but that is currently known to be caused by fungi or bacteria, the epidemiology of this entity is not well known because the reports are based on small series, at the national level in the largest series found reports 70 cases/year, of which 76% affect the male gender and the age of greatest affection is 20-40 years, however, cases have been reported from 3 to 80 years.

The microbiological agent isolated in the highest percentage in Mexico is Nocardia brasiliensis in 86.6% and Actinomadura madurae in 10.2%, in contrast to publications from Africa and India where eumycetes predominate. The incubation period is unknown, but it is known that it is always due to traumatic inoculation and this period will be affected by the microorganism, the patient's nutritional status and the immune response. The diagnosis of mycetoma requires microbiological identification because the clinical lesion is the same. If it is actinomycetes or eumycetes, to achieve this there are multiple techniques (Gram, cultures, PCR, biochemical tests, histopathology) with which the specific treatment (antibiotics or antimycotics) can be determined.

Treatment regimens are not well established due to the scarcity of studies with a high degree of scientific evidence, which is why they currently vary according to the microbiological agent, pharmacological combination and expert opinion, however for the combination of dapsone and trimethoprim-sulfamethoxazole (TMS) it is recommended to continue for 6 months after remission of the skin lesions and bacteriological negativization, for the combination of TMS with aminoglycosides, 15 weeks are recommended with serial renal and audiological controls due to its adverse effects, in some series they have stopped TMS for life [11-13]. In cases where neurological deficits due to compression occur, surgery is recommended based on the principles of spinal infections not associated with the surgical site [4,5,14].

Conclusion

Cervico-thoracic mycetomas are a rare presentation of a neglected tropical disease as the WHO stipulates, in the largest series an incidence of approximately 2.5% is reported and the morbidity-mortality increases for which it must receive a specific treatment based on 3 key points: Result of microbiology and histopathology. Neurological compromise. spinal stability [15-18].

Having a result of microbiology, a specific regimen can be established, in case of neurological deficit, the compromised site (anterior, posterior or combined) is assessed to offer a decompressive procedure and in case of instability, offer some fixation technique (anterior, posterior or combined) however we currently do not find publications that address this point (vertebral stability-mycetoma).

The key treatment in this pathology are antimicrobials for which we must have microbiological isolation and in cases where microorganisms are not isolated, it is recommended to carry out empirical management based on regional epidemiology and guide us with the clinical and radiological response, all the life due to the risk of recurrent lesions, especially in cases such as the one we present (size of the lesion, extrapedial site, time of evolution, among other risk factors) [19-20].

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