Clinical note

Clinical-scintigraphy discordance in a case of malignant external otitis

T. Morales-Avellaneda, A.I. González-Ramírez, B. Rodríguez-Alfonso, J. Mucientes-Rasilla, J.R. García-Berrocal, B. De Diego

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A B S T R A C T

We report the case of an immunocompromised female patient with external otitis, who developed a picture of external otitis with poor response to conventional treatment and subsequent isolation of Pseudomonas aeruginosa in the ear secretion culture. The final diagnosis was malignant external otitis. Our objective is to describe the scintigraphic findings as well as to analyze their correlation with the clinical pattern and to evaluate the possible causes of the discordance found.

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Discordancia clínica-gammagráfica en un caso de otitis externa maligna

R E S U M E N

Presentamos el caso de una paciente inmunodeprimida, que desarrolló un cuadro de otitis externa con pobre respuesta al tratamiento convencional y posterior aislamiento de Pseudomonas Aeruginosa en el cultivo de secreción ótica. El diagnóstico definitivo fue de otitis externa maligna. Nuestro objetivo es describir los hallazgos gammagráficos y analizar su correlación con los hallazgos clínico-quirúrgicos y valorar las posibles causas de la discordancia encontrada.

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Introduction

Malignant external otitis (MEO) is a relatively rare complication of external otitis which generally occurs in immunosuppressed and diabetics patients. This disease presents as a necrotizing external otitis affecting only soft and cartilaginous tissues, evolving to osteomyelitis at the base of the cranium and affecting the temporal bone or the base of the cranium. However, many authors do not distinguish between these two manifestations and consider them as synonymous. Pseudomonas aeruginosa is the pathogen most frequently isolated in secretion cultures followed by opportunistic Gram-negative bacilli and, less frequently, by fungi (Aspergillus fumigatus).

Scintigraphy with 99mTc-HDP and 67Ga provides valuable information related to extension, prognosis, therapeutic planning and monitoring and disease recurrence.

At present the mortality has diminished from 30–40% to 20% thanks to early diagnosis and adequate, opportune treatment. Recurrence may appear up to one year after finalization of treatment and thus, prolonged, regular follow up is necessary in these patients.

Clinical case

We present the case of a 53-year-old woman with a history of well differentiated lymphocytic lymphoma receiving the first cycle of treatment with R-DAHOX (dexamethasone: 40 mg d1–4, oxaliplatin 130 mg/m² d1, ARA-C 2 g/m² d2–3). She was attended in the Emergency Department for facial paralysis, otalgia and right otorrhea, fever, deterioration in the general state, intolerance to oral intake and dizziness. The patient was diagnosed with MEO and initiated treatment with amoxicillin/clavulanate with poor clinical response. She was admitted to hospital and Pseudomonas aeruginosa was isolated from the ear exudate, and intravenous antibiotic therapy was initiated according to the antibiogram test.

Following 5 days of treatment with ceftazidime associated with 3 days with amikacin, a scintigraphy was requested to evaluate bone involvement. After performing planar images (Fig. 1) and SPECT-CT with 99mTc-HDP (Fig. 2) following the standard protocol, pathological uptake was observed in the right mastoid and petrous bone without passing the midline and with probable extension to the ipsilateral temporal–mandibular articulation, being thereafter classified as level II (according to the Alfred Hospital classification).

Taking the scintigraphic findings into account and the bad clinical evolution of the patient, tympanoplasty was performed with mastoidectomy showing hypertrophic mucosa in the antrum, clear

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Fig. 1

Fig. 2

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otorrhea and thinning of the mastoid and tympanic tegmens and no apparent involvement of the auditory bones or the Eustachian tube.

The patient showed progressive improvement after surgery and 4 weeks of combined antibiotic treatment, remaining afebrile, stable and without pain.

**Discussion**

Although the initial diagnosis of MEO is clearly clinical, imaging studies play a very relevant role in determining the extension of the disease in both soft tissue and the bones of the base of the cranium, thereby facilitating therapeutic planning and patient follow up.

Computerized tomography (CT) is useful to confirm the diagnosis in advanced forms. Nonetheless, it is not a specific test and provides scarce information in the follow up. MR is useful to define the involvement of the soft tissue, particularly all the infratemporal region, but it also has limited value in the follow up of these patients.3

Bone scan and SPECT-CT with ⁹⁹mTc-HPD is very useful for early diagnosis of the disease with a high sensitivity (100%), albeit with a low specificity, remaining positive after pathological resolution. It is therefore of little interest in the short term follow-up. Scintigraphy with ⁶⁷Ga shows lesser spatial resolution. However, it is considered of great utility for the control and follow-up of these patients because of its high specificity in active inflammatory processes. Normalization of the scintigraphy with ⁶⁷Ga is an indicator of cure of the disease.3,4

In the present case the patient presented a positive bone scintigraphy which did not correlate with the surgical findings in which there were no evident signs of bone extension. This discordance may be attributed to the presence of superficial bone erosion by contiguity (osteitis) with scarce macroscopic but clear scintigraphic evidence or early response to treatment. In the latter case we consider the performance of scintigraphy with ⁶⁷Ga to have been useful. Nonetheless, the urgency of the clinical situation of the patient required an immediate therapeutic approach.
Fig. 2. The SPECT-CT images provide greater definition of the pathological uptake in the right mastoid and petrous bone with no extension beyond the midline.

References