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ORIGINAL ARTICLE

Lymphadenitis due to non-tuberculous mycobacteria: Experience over 15 years☆

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KEYWORDS
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Lymph node excision;
Drainage

Abstract
Objective: To study the epidemiology, clinical features, diagnosis, therapeutic management, and outcome of non-tuberculous mycobacterial lymphadenitis in a paediatric population of Aragón (Spain).
Material and methods: A retrospective study was conducted on patients under 15 years-old diagnosed with non-tuberculous mycobacterial lymphadenitis between the years 2000 and 2015. Inclusion criteria: patients with lymphadenitis and positive culture. Quantitative values are shown as mean, rank, and standard deviation, and qualitative data as frequencies.
Results: Twenty-seven cases were registered, with a mean age of presentation of 39.9 months (range 10 months–8 years). The mean time between the symptoms onset and first consultation was 1.7 ± 1.1 months. The most frequent location was sub-maxilar in 17/27 cases (63%), on the right side in 59.3%, and size 2.96 ± 1.26 cm. Fistulae were observed in 16/27 cases. Tuberculin test was greater than 10 mm in 7/24 (29.1%). Microbiological cultures were positive for *Mycobacterium avium* in 14/27 (51.9%), *Mycobacterium intracellulare* 3/27 (11.1%), and *Mycobacterium lentiflavum* 3/27 (11.1%). Combined treatment of antibiotics and surgery was given in 16/27 cases (59.8%), medical treatment only in 7/27 (25.9%), and surgical exeresis alone in 4/27 (14.8%). Two patients required a new surgery, and one showed severe neutropenia secondary to rifabutin. Only one case (3.7%) suffered from temporary facial palsy as sequel.

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Conclusions: The most frequent treatment was the combination of antibiotics and surgery. Delay in diagnosis seemed to be responsible for the limited number of exeresis as first option, only one for every seven patients.

PALABRAS CLAVE
Linfadenitis;
Micobacteria;
Micobacteria no tuberculosa;
Macrólido;
Exéresis nódulo linfático;
Drenaje

Linfadenitis por micobacterias no tuberculosas: experiencia de 15 años

Resumen
Objetivo: Estudiar la epidemiología, las manifestaciones clínicas, el manejo diagnóstico-terapéutico y la evolución de las linfadenitis por micobacterias no tuberculosas en la población pediátrica de Aragón.
Material y métodos: Estudio retrospectivo de pacientes menores de 15 años diagnosticados de linfadenitis por micobacteria no tuberculosa entre 2000 y 2015. Criterios de inclusión: pacientes con linfadenitis y cultivo positivo. Los resultados se expresan como medias, rango y desviación típica para las variables cuantitativas, y porcentajes para las cualitativas.
Resultados: Se detectan 27 casos, edad media de presentación 39,9 meses (rango 10 meses-8 años). El tiempo desde inicio de los síntomas hasta la primera consulta especializada es $1,7 \pm 1,1$ meses. La localización más frecuente es submaxilar en 17/27 casos (63%), lado derecho en el 59,3%, con tamaño de $2,96 \pm 1,26$ cm. Solo 16/27 presentan fistulización. Prueba de tuberculina superior a 10 mm en 7/24 (29,1%). El cultivo es positivo para *Mycobacterium avium* en 14/27 (51,9%), *Mycobacterium intracellulare* 3/27 (11,1%), *Mycobacterium lentiflavum* 3/27 (11,1%). El 92,6% (23/27) es tratado inicialmente con amoxicilina-clavulánico. La combinación de antibióticos y cirugía se aplica en 16/27 casos (59,3%), solo antibioterapia 7/27 (25,9%) y únicamente exéresis 4/27 (14,8%). Dos pacientes precisan reintervención y un caso desarrolla neutropenia grave secundaria a rifabutina. Solo un caso (3,7%) presenta parálisis facial transitoria como secuela.
Conclusiones: La combinación de antibioterapia y cirugía es el tratamiento más frecuente. El retraso en el diagnóstico hace que la exéresis como primera opción terapéutica se realice únicamente en uno de cada 7 pacientes.

Introduction

Cervical lymphadenitis is the most frequent manifestation of infection by nontuberculous mycobacteria (NTM) in immunocompetent children. It amounts to 10%–20% of cervical, submandibular and preauricular lymphadenitis cases in early childhood. It affects children aged 1–5 years and is rare after age 10 years, unlike tuberculous lymphadenitis.[1]

Nontuberculous mycobacteria are found in water, soil, milk, animals (mainly birds) and health care equipment. Infection of the cervical lymph nodes follows inhalation, inoculation or ingestion of contaminated material in 90% of cases. The development of a unilateral, painless isolated mass in the submaxillary or cervical region is the most frequent clinical manifestation. The natural history of the disease involves a progression of the adenopathy, with the overlying skin taking a violaceous hue, fistula formation in three to four months, and a chronic indolent course that ends with skin scarring in 12–18 months.[2]

In recent years, its incidence has increased both in Spain and worldwide.[3–5] Until the mid-1990s, the incidence was of 1.2 cases/year based on data from a study conducted in a tertiary hospital of the Autonomous Community of Madrid. At present, in this same hospital, it is of 5.25 cases/year.[3] In countries such as the Netherlands or Australia, the incidence reaches up to one case per 100 000 inhabitants.[4,5]

However, most of the data published in the literature is from case series, and no conclusive controlled clinical trials have been conducted to assess the efficacy and safety of the different approaches to its management. Although complete excision of the affected lymph node is currently the gold standard, this is difficult to implement due to delays in diagnosis. However, other alternatives such as close observation and pharmacological treatment alone or combined with surgery are management options that seem to achieve similar outcomes.[6]

Materials and methods

We conducted a multicentre, retrospective observational study between 2001 and 2015 in patients aged less than 15 years with a diagnosis of nontuberculous mycobacterial lymphadenitis (NTMLA) recorded in the database of the