■ CASE STUDIES

Mansonella perstans Isolated on Aspiration Puncture of a Salivary Gland

Lourdes Mateu,[a] Nieves Sopena,[a] Montserrat Giménez,[b] and Lluís Valerio[c]
[a]Unidad de Enfermedades Infecciosas, Hospital Germans Trias i Pujol, Universitat Autònoma de Barcelona, Badalona, Barcelona, Spain
[b]Servicio de Microbiología, Hospital Germans Trias i Pujol, Universitat Autònoma de Barcelona, Badalona, Barcelona, Spain
[c]Unitat de Salut Internacional del Barcelonès Nord i Maresme, SAP Santa Coloma de Gramenet, Institut Català de la Salut, Santa Coloma de Gramenet, Barcelona, Spain

Chronic parasitosis due to nematode worms (*filariae*) in tissue are very common in tropical ecosystems; their larvae (*microfilariae*) have been isolated in lymph vessels, skin, and blood. The case reported here is of a Nigerian patient suffering severe renal failure and admitted owing to the presence of a right sub-mandibular gland abscess. In the FNAP, the presence of a *Mansonella perstans* microfilaria was identified. Post-surgery examination of pathology samples from the gland reported an acute inflammatory infiltrate including abundant eosinophils and microfilariae. There are some reports in the literature of haematic or lymphatic microfilariae, especially in epithelial or glandular tissues, and, as in this case, in inflammatory or malignant conditions. Both ivermectin and mebendazol are used for treatment; the scant pathogenicity of the genus *Mansonella* should, however, lead to an individualized decision.

Key words: Filariosis. *Mansonella perstans*. Salivary gland.

***Mansonella perstans* aislada en una punción-aspiración de glándula salival**
Las parasitosis crónicas por nematelmintos de localización hística (filarias) son comunes en los ecosistemas tropicales; sus larvas (microfilarias) se aíslan en los vasos linfáticos, la piel o la sangre. Se presenta el caso de un paciente nigeriano afecto de insuficiencia renal grave que ingresó por un absceso en la glándula submandibular derecha. En la punción-aspiración con aguja fina se identificó una microfilaria de la especie *Mansonella perstans*. En el posterior estudio anatomopatológico posquirúrgico de la glándula, se determinó infiltrado inflamatorio agudo con abundantes eosinófilos y microfilarias. En la revisión bibliográfica efectuada se constatan algunos aislamientos de filarias hemáticas o linfáticas especialmente en tejidos epiteliales o glandulares; todos ellos, al igual que el que se presenta, en condiciones de inflamación o malignidad. La decisión de tratar o no con ivermectina o mebendazol una filariasis de escasa patogenicidad como las del género Mansonella debe individualizarse en cada caso.

Palabras clave: Filariasis. Mansonella perstans. Glándula salival.

INTRODUCTION

Mansonellosis, or chronic infection due to *Mansonella perstans*, is one of the most common parasitic infections in western and central Africa. Its prevalence varies by geographical areas, but infection rates of 15%-20% are common in the general population. The adult nemathelminth or macrofilaria lives in the vessels of the peritoneum, from which it expels the haematic forms or microfilariae without any know circadian rhythm. Insofar as it is an infection with a very low level of pathogenicity, it is possible to insolate microfilariae in the peripheral blood of asymptomatic patients, especially immigrants, for many years. It is not unusual to encounter diagnoses of mansonellosis due to serendipity or in the course of an eosinophilia study.[1,2]

Although it does not require specific treatment on occasions, because of its scant pathogenicity, its correct taxonomic diagnosis is very important, as it must be distinguished from other blood filariae causing serious morbidity: *Loa loa* and *Wuchereria bancrofti*. Screening for filariasis in healthy African immigrants is a controversial issue for due to its low cost-effectiveness.[3] There are ELISA and protein C-reactive assays that allow the detection of antibodies and genetic material, respectively, of *L loa* and *Onchocerca volvulus* (lymph node filaria), but there are no equally effective diagnostic resources available for the

The authors have not indicated any conflict of interest.

Correspondence: Dr. L. Valerio.
Unitat de Salut Internacional del Barcelonès Nord i Maresme.
Jacint Verdaguer, 118. 08923 Santa Coloma de Gramenet.
Barcelona. España.
E-mail: lvalerio.bnm.ics@gencat.net

Received May 10, 2007.
Accepted for publication September 3, 2007.

detection of filariae of Wuchereria or Mansonella.[4] We report on the case of Nigerian patient who was admitted due to a sub-mandibular tumour lasting for 7 days and severe kidney failure.

CASE STUDY

Twenty-seven-year-old male born in a rural area of south-eastern Nigeria and resident in Europe for 10 years and in Spain for the last 3; he has not visited his country of birth since arriving in Spain. His medical history reflects arterial hypertension and nephropathy of unknown origin diagnosed during a medical check-up at work 8 months ago, currently untreated.

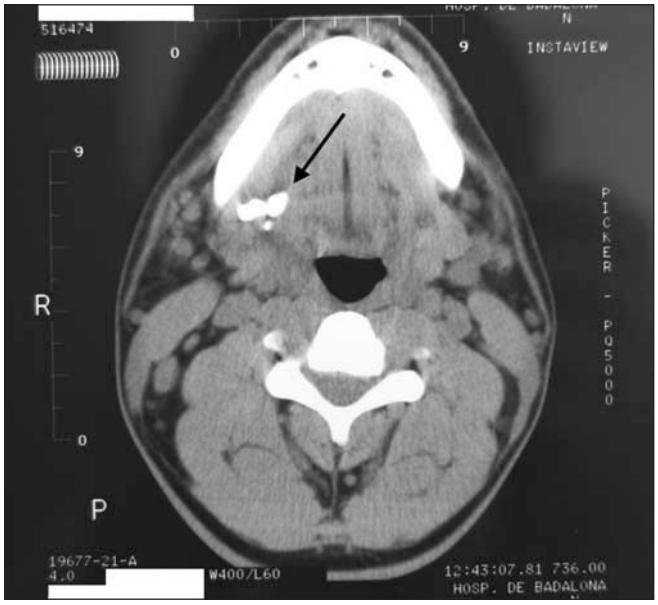

Figure 1. Computerized tomography showing the inflammatory infiltrate with the intraglandular calcifications (arrow).

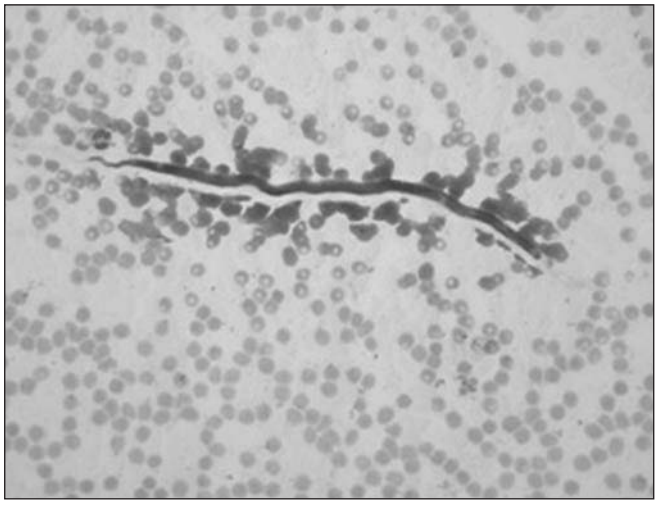

Figure 2. Smear cytology of the fine needle aspiration of the sub-mandibular gland. Presence of an unsheathed microfilaria identified as *Mansonella perstans* (May-Grünwald-Giemsa stain, 40).

At the time of his admission the patient had no fever and was oriented, with a blood pressure reading of 173/107 mm Hg. He complained of an intense spontaneous pain and pressure in the right sub-mandibular area, where an unmoving, hard but not rock-hard tumour of 3 2.5 cm was identified by palpation. The oropharyngeal cavity and the floor of the mouth were normal. No adenopathies were found on palpation of the neck. The rest of the physical examination using equipment showed no alterations, except for signs compatible with grade II hypertensive retinopathy. The blood test showed a non-septic haemogram with mild eosinofilia (9%), moderate normochromic normocytic anaemia, and a globular sedimentation velocity of 90 mm. His biochemistry results showed creatinine to be 5.3 mg/dL and potassium at 5.4 mmol/L. His urine presented proteinuria of 2.42 g/dL (creatinine clearance, 31.5 mL/min.).

The large drip and smear of peripheral blood on admission, as well as the detection of parasites in faeces, on skin, and in urine, were all negative. The computerized tomography of the neck revealed images compatible with an abscess in the right sub-mandibular gland that extended into the subcutaneous cell tissue. Inside this tissue there were 2 calcified nodular images measuring approximately 1 cm (Figure 1).

With the clinical suspicion of right sub-mandibular suppurative sialoadenitis, treatment was begun with antibiotics, amoxicillin and clavulanic acid (875/125), and anti-inflammatory drugs. A fine-needle aspiration puncture (FNAP) was performed, revealing a smear compatible with peripheral blood, scant glandular cells, eosinophils, and a single sheath-less vermiform extracellular parasite with nuclei to the caudal end, which the microbiology department identified as *M perstans* (Figure 2).

Due to the scant response to treatment, the gland was surgically removed. The pathology report revealed an acute and chronic inflammatory infiltrate with plasma cells, eosinophils, and abundant microfilariae. Culture in conventional media and media for mycobacteria was negative. The skin samples (double skin-snip) in search of *Onchocerca volvulus* and parasites in urine and faeces, in search of other pathogens causing eosinophilia and/or glomerulonephritis, were all negative. The patient presented good post-surgical progress and, after assessment by the nephrology department, he was diagnosed as having hypertensive nephropathy and was seen at the out-patient clinic, with gradual deterioration of his kidney function to the point where dialysis was required.

DISCUSSION

In the case reported here, it is worth highlighting the finding of microfilariae of *M perstans* in the FNAP of a salivary gland. There is only one other reference to isolation in that location (and in similar circumstances) in MEDLINE, EMBASE, and IME, so the present communication would represent the second record of a salivary gland and the third

Parasites Related to Otorhinolaryngological Conditions

Larval cysts of *Taenia soleum* (cysticercosis)
Larval cysts of *Echinococcus granulosus* (hidatidosis)
Tripanosoma cruzi (Chagas disease in salivary glands)
Toxoplasma gondii
Leishmania spp

referring to the organs of the ears, nose, and throat.[5] Table lists other parasites with pathological expression in otorhinolaryngology. Another species, *W bancrofti*, has many other records concerning culture isolations from the thyroid, conjunctive, dermal, breast, and ovarian tissue by FNAP, especially in cases malignancy or, as in the case presented here, inflammation.[6-8] Although it is evident that there is no bibliographical basis to state this definitively, it is possible that the lymph node or haematic microfilariae present a certain tropism for epithelial and glandular tissues in the context of an acute inflammatory reaction.

In any case, although treatment with mebendazol or ivermectin drastically reduces the concentration of microfilariae in blood, this massive microfilarial necrosis may lead to acute renal tubular necrosis in patients with underlying glomerulonephritis.[9,10] After assessing the risk/benefit ratio of treating a non-pathogenic filariosis in a patient with advanced kidney failure, it was decided not to treat the parasitosis. The patient required dialysis after a few months, at which time he received therapy with ivermectin not only to reduce the microfilarial burden but also to treat other parasitoses (especially *Strongyloides stercolaris*) that might reproduce on a massive scale in the event of immunodeficiency induced to avoid rejection in a future kidney transplant. Follow-up after treatment showed that the concentration of microfilariae had diminished by 80%. Subsequent cycles of mebendazol have led to the negativization of the microfilariae in blood. At present, he continues to attend out-patient check-ups and has been registered on the kidney transplant programme.

REFERENCES

1. Wanji S, Tedongfor N, Esum M, Ndideng M, Enyong P. Epidemiology of concomitant infections due to Loa loa, Mansonella perstans and Onchocerca volvulus in rain forest villages of Cameroon. Med Microbiol Immunol (Berl). 2003;192:15-21.
2. Roca C, Balanzó X, Sauca G, Fernández-Roure JL, Boixeda R, Ballester J. Imported hookworm infection in immigrants in Spain: study of 285 patients. Med Clin (Barc). 2003;121:139-41.
3. Carrillo E, Iglesias B, Gómez i Prat J, Guinovart C, Cabezos J. Cribaje de microfilariasis sanguínea (Loa loa) en la población de Barcelona inmigrante de zonas endémicas. Rev Esp Salud Publica. 2004;78:623-30.
4. Walther M, Muller R. Diagnosis of human filariases. Adv Parasitol. 2003; 53:149-53.
5. Sahu KK, Pai P, Raghuveer CV, Pai RR. Microfilaria in a fine needle aspirate from the salivary gland. Acta Cytol. 1997;41:954.
6. Bregani ER, Ceraldi T, Rovellini A, Ghiringhelli C. Case report: intraocular localization of Mansonella perstans patient from south Chad. Trans R Soc Med Hyg. 2002;96:654.
7. Goverde AJ, Schats R, van Berlo PJ, Claessen FA. An unexpected guest in follicular fluid. Hum Reprod. 1996;11:531-2.
8. Baird JK, Neafie RC, Connor DH. Nodules in the conjunctiva, bung-eye and bulge-eye caused by Mansonella perstans. Am J Trop Med Hyg. 1998;38:552-7.
9. Pakasa MM, Nseka NM, Nyimi LM. Secondary collapsing glomerulopathy associated with Loa loa filariasis. Am J Kidney Dis. 1997;30:836-9.
10. Mehta KS, Halankar AR, Makwana PD, Torane PP, Satija PS, Shah K. Severe acute renal failure in malaria. J Postgrad Med. 2001;47:24-6.