Community-acquired granulomatous mastitis superinfected with Mycobacterium bolletii

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Mastite granulomateuse à *Mycobacterium bolletii* d'origine communautaire

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Introduction

Mycobacterium bolletii is an emerging pathogen [1], either responsible for soft tissue infections following mesotherapy injections, acupuncture, and piercing procedures, or respiratory tract infections in patients presenting with cystic fibrosis. Disseminated infections in immunocompromised patients have also been reported [2]. We reported a case of mastitis superinfected with *M. bolletii* in an immunocompetent patient.

Case presentation

A 51-year-old previously healthy nulliparous patient presented to our infectious diseases department for a five-month history of painful and inflammatory nodule in the superior external quadrant of the left breast. Several aspiration cytologies and biopsies and a surgical excision yielded the diagnosis of granulomatous mastitis, with no sign of malignancy. An abscess developed under the surgical scar after a short steroid treatment; several bacterial cultures of the purulent discharge remained negative. The patient received several courses of amoxicillin-clavulanic acid with no improvement. Surgical drainage was followed by new recurrence and the patient was referred to the infectious disease department.

The patient evasively reported a wound of the thoracic region with a tree branch, some time before the nodule developed. Two consecutive samples obtained by needle aspiration yielded mycobacteria after prolonged culture (10 days for the first, and 20 days for the second) on standard medium (Middlebrook 7H9 broth supplemented with 10% oleic acid-albumin-dextrose (OADC) at 37°C under microaerophilic condition). The isolate was identified as *Mycobacterium abscessus* complex by mass spectrometry (MALDI-TOF: Microflex, Bruker Daltonik GmbH, Germany) (score 1.92). Sequencing of *rpo*B gene confirmed *Mycobacterium bolletii* on the basis of 99.3% sequence similarity with the reference sequence (Genbank HQ404268) [3]. The patient received multiple antibiotic treatments because the susceptibility of the strain to antibiotics was difficult to ascertain (finally found to be *in vitro* susceptible only to amikacin and tigecycline), and because the patient experienced drug intolerance: linezolid/amikacin for one week, then linezolid/clarithromycin for four weeks, clarithromycin/rifabutin for three weeks, and finally

intravenous tigecycline /amikacin for five weeks. Pain and local sign of inflammation slowly disappeared, and treatment was stopped 12 weeks after the diagnosis. No relapse was observed after one year of follow-up.

To distinguish inoculation by initial vegetal wound from superinfection of the surgical site, biopsies of the nodule were newly examined with specific stains (Ziehl, PAS, Gomori), without evidence of acid-fast bacilli. Culture of a branch of the tree that might have caused the initial wound was not contributory, and culture of water from the tap at the patient's house yielded *Mycobacterium gordonae*, but not *M. bolletii*. The nosocomial origin of the infection was unlikely because of the absence of any other case of similar infection in the gynecologic ward during this period.

Discussion

Granulomatous mastitis is a rare disease (1.8% among 1,106 women with benign breast disease) [1], occurring in women aged around 30 years, with a past history of pregnancy and breastfeeding. Microbiological findings are usually negative, and an autoimmune mechanism has been recently suggested [4]. Steroid treatment associated with surgical removal usually leads to clinical cure [5].

Non-tuberculous mycobacteria such as *Mycobacterium chelonae* and *M. abscessus* complex can grow on blood agar within one week. They are present in the environment, essentially in water.

M. abscessus infections can be due to contaminated endoscopes during disinfection process [6] or cutaneous injection of unapproved alternative intravenous medication [7]. *M. abscessus* has also been described as responsible for granulomatous mastitis caused by breast "piercing" [8], and breast abscess after silicone injection , or surgical breast augmentation [9]. *M. bolletii* has first been described in 2006 as one of the species of the *M. abscessus* complex but its precise taxonomy remains controversial [1]. Sequencing the h*sp*65 and *rpo*B genes allows for differentiating between taxons in the *M. abscessus* complex [1, 3]. Localized infections due to *M. bolletii* have rarely been reported: a few cases of abscess after mesotherapy subcutaneous injections, three cases of furunculosis associated with pedicure footbaths [10]. Lesions were described as granulomatous and suppurating, similarly to our case report. Only four antibiotics are known to be active against *M. bolletii*: tigecycline,

amikacin, imipenem, and clarithromycin.

Conclusion

The link between granulomatous mastitis and *M. bolletii* infection deserves further investigations and prolonged culture of tissue on specific medium is warranted to establish the diagnosis. Cure of subcutaneous infection with *M. bolletii* can be achieved with tigecycline and amikacin.

Contribution of authors

MD and EP performed the investigations related to this patient.
CJ and CP wrote the article.
JLS and JPL managed the patient.

Disclosure of interest

The authors declare no conflict of interest.

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