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Postpartum septic arthritis of pubic symphysis due to *Sneathia sanguinegens*, *Sneathia vaginalis*, and *Mageeibacillus indolicus*: Contribution of clinical metagenomics

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Abstract

Sneathia sanguinegens, *Sneathia vaginalis*, and *Mageeibacillus indolicus* have been recently described in the female genital tract. We present the first case of a postpartum septic arthritis of the pubic symphysis due to these organisms, identified by next generation sequencing.

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1. Introduction

Septic arthritis of the pubic symphysis is a rare infection, representing about 0.8–1.36% of all septic arthritis cases in adults [1]. Risk factors are previous gynecological or urological surgeries, delivery, drug abuse, diabetes, cancer, local macro- and micro-traumatism, and radiotherapy [2].

We present the case of a patient with septic arthritis of the pubic symphysis during the postpartum period [3,4], where metagenomic next generation sequencing (mNGS) allowed us to detect and identify unusual pathogens.

2. Case report

A 32-year-old woman presented at 40 weeks of gestation in the latent phase of labor with maternal fever and fetal tachycardia. Complaining of upper respiratory tract symptoms, a rapid influenza test (Cobas®Liat®, Roche) turned out positive for influenza A, motivating a treatment with oseltamivir for 5 days. She had an uncomplicated spontaneous vaginal delivery with only a superficial tear on the labia minora.

On Day 1 postpartum, she complained of pain in the groin and pubic symphysis, rapidly worsening and resulting in an inability to walk. In parallel, she had persistent fever. A pelvic magnetic resonance imaging (MRI) revealed on postpartum Day 9 a high suspicion of septic arthritis of the pubic symphysis (Fig. 1). An ultrasound-guided aspiration of the joint returned 3 ml of purulent fluid. Surgical drainage of the collection was then performed. During surgery, a pelvic exam was done by a senior gynecologist and no fistula nor previously undiagnosed lesions were found. Immediately following surgery, initial empiric therapy with cefuroxime was switched to broad-spectrum antibiotic (piperacillin-tazobactam). The culture of the ultrasound-guided joint aspiration (without antibiotics) and of

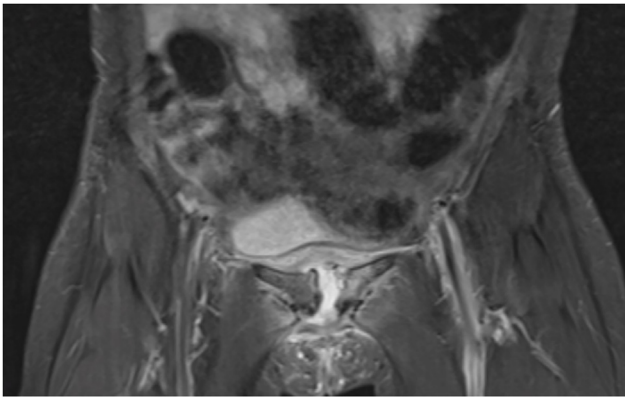


FIG. 1. Magnetic resonance image of the pelvis (T2 WI tse cor, STIR cor) shows an intra-articular effusion at the pubic symphysis with diastasis and contrast enhancement of the peripheral soft tissues.

multiple perioperative samples did not yield growth under standard conditions. These included blood agar, chocolate agar, MacConkey agar and colistin-nalidixic acid agar plates incubated under aerobic conditions (72 h incubation at 35 °C in 5% CO₂ atmosphere), CDC agar plates incubated under anaerobic conditions (48 h incubation at 35 °C) and brain-heart infusion broth (5 days incubation at 35 °C in 5% CO₂ atmosphere). Direct staining of the aspirate revealed gram-positive cocci and gram-negative rods. Owing to rapid clinical recovery, the patient was discharged on Day 4 postoperatively with a 3-week course of antibiotics (amoxicillin-clavulanate).

2.1. mNGS analysis

Suspecting a polymicrobial infection with fastidious organisms, we performed mNGS of joint fluid after bacterial DNA enrichment using the Ultra-Deep Microbiome Prep (Molzym) with modified sample pretreatment [5,6] protocol. Our mNGS pipeline (HUGe-MAP) consisted in Illumina 2 × 151 iSeq 100 sequencing, read quality filtering, removal of replicate and human reads, and classification of non-human (paired) reads using CLARK [7] as described previously [6] except for the minimum read length that was set to 90 nt. Of 3,062,842 generated reads (pairs), 2,510,457 passed the quality check and most of them corresponded to human DNA (2,057,545). Among non-human reads (31,992; deposited in the European Nucleotide Archive under study number PRJEB44108), 28,628 were assigned to bacteria, including *Sneathia sanguinegens* (27,489), *Sneathia vaginalis* (332), *Mageeibacillus indolicus* (45) and a common reagent contaminant *Cutibacterium acnes* (31). Other bacterial species (including *Gardnerella vaginalis*) were represented by < 24 reads. In the negative control (NEC), *C. acnes* was the dominant organism (8997 reads). Only one read was assigned to *S. sanguinegens*, and none to *S. vaginalis* or *M. indolicus*.

The reads assigned by the HUGe-MAP pipeline to *M. indolicus*, *S. sanguinegens* and *S. vaginalis* were queried against

corresponding reference genome sequences using *blastn* search application (-word_size 12 -evalue 1e-010 -perc_identity 80 -qcov_hsp_perc 80 -culling_limit 1) from BLAST+ v2.13.0 [8]. This analysis largely confirmed taxonomic assignments to *M. indolicus* [(100% of reads with *blastn* hits; median (Q1–Q3) hit sequence identity percentage 99.3 (98–100)] and to *S. sanguinegens* [(98.7% of reads with *blastn* hits; median hit sequence identity percentage 99.3 (98.7–100)]. The reads classified as *S. vaginalis* by HUGe-MAP had a lower similarity to the reference genome sequence of this species [(64.4% of reads with *blastn* hits; median hit sequence identity percentage 94.9 (91.3–97.3)] and to that of *S. sanguinegens* [(7.4% of reads with *blastn* hits; median hit sequence identity percentage 94.1 (89.5–96.7)]. We cannot exclude that (at least some of) these reads originated from a putative yet-to-be-described *Sneathia* species [9].

3. Discussion

The presence of *Sneathia sanguinegens* [10] [synonym *Leptotrichia sanguinegens* [11]], *Sneathia vaginalis* [12] [synonyms *Sneathia amnii* [9] and *Leptotrichia amnionii* [13]] and *Mageeibacillus indolicus* confirms the diagnosis of septic arthritis of pubic symphysis, the likely source being the female genital tract. In 2015, Austin et al. [14] isolated *M. indolicus* (family Oscillospiraceae, phylum Firmicutes), a strictly anaerobic gram-positive rod-shaped bacterium from the female genital tract associated with bacterial vaginosis. *S. sanguinegens* and *S. vaginalis* are gram-negative rod-shaped and pleomorphic coccobacilli, respectively, which belong to the family Leptotrichiaceae (phylum Fusobacteria) and are part of the vaginal microbiome [9,15]. During vaginal delivery, microtrauma to the pubic symphysis can predispose to local bacterial inoculation. In seven of the nine published cases of septic arthritis of the pubic symphysis during the postpartum period [2–4], *Streptococcus*,

Staphylococcus or *Pseudomonas* have been identified while *Sneathia* and *Mageeibacillus* have not been reported.

Sneathia species have been sporadically reported as causes of infection, including peripartum bacteremia [16], and have recently been considered potential emerging pathogens [15]. The underestimation of their potential pathogenicity may be linked to their complex growth requirements [15]. Therefore, such bacteria could be, for example, etiological agents of culture-negative postpartum septic arthritis reported by Ducrotoy et al. [17]. Our case highlights the advantage of mNGS in the detection of emerging pathogens in particular situations.

CRedit author statement

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Nadia Gaia: Methodology, Investigation, Validation.

Begoña Martinez de Tejada: Writing - Review & Editing.

Jacques Schrenzel: Investigation, Methodology, Validation, Resources, Supervision, Writing - Review & Editing.

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Transparency declaration

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