**Case Reports**

# *Haemophilus parainfluenzae* infective endocarditis associated with pelvic abscess: an uncommon complication of endometriosis

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**Summary**

The case of a woman with native mitral valve endocarditis due to *Haemophilus parainfluenzae* (HPI) associated with a pelvic abscess and endometriosis is reported. Although HPI is an infrequent pathogen involved in endocarditis, association to a gynaecological infection has never been reported. Endometriosis could increase this risk.

*Key words*: *Haemophilus parainfluenzae;* Infective endocarditis; Endometriosis.

## Introduction

Common predisposing factors for salpingitis and gynaecologic abscess include sexual behaviour [1], increased age, diabetes and immunocompromised status due to HIV infection or renal transplantation. Endometriosis, which is found in 25-40% of women with infertility and in 2-5% of the general population [2], has been suspected to increase occurrence of gynaecologic abscess [3]. Although *Haemophilus influenzae* (HI) is a common respiratory tract pathogen in humans but, rarely isolated from fallopian tubes, few reports have described HI salpingitis and tuboovarian abscess [4-7] and only two *Haemophilus parainfluenzae* (HPI) genital infections have been reported [5, 6]. HI and HPI are also recognised as rare causative agents ofpathogens implicated in bacterial infective endocarditis (IE) [8].

We report an unusual case of *Haemophilus parainfluenzae* endocarditis IE associated with a gynaecologic abscess in a patient with endometriosis.

## Case Report

A 33-year-old woman, gravida 0, para 0, suffering from severe endometriosis (Stage IV) and ovarian cysts was known to have a heart murmur, never explored. In March 2008, she presented suddenly with a flu-like syndrome and was prescribed ibuprofen and paracetamol. Laboratory investigations were normal. Four days later, the patient was still febrile (38.6°C) and complained of pelvic pain. Blood samples revealed a thrombopenia (28 g/l). White blood cell count was 23.0 g/l with a predominance of polymorphonuclear leukocytes and the C-reactive protein level was 437 mg/dl. At hospital admission, her temperature was 39.1°C, while her blood pressure and pulse rate were normal. A mitral insufficiency murmur was audible. No sign of heart failure, palpable spleen or peripheral cutaneous lesions were observed. Dental examination and radiography were normal. There was localised mild tenderness in the lower abdominal quadrant. Vaginal examination resulted in severe pain. Ultrasound exam revealed a right tuboovarian abscess (62 x 43 mm). Abdomino-pelvic computed tomography (CT) showed that the lesion was non homogeneous, with enhancement after contrast administration (56 x 45 mm) and was associated with a peripheric abscess in the Douglas pouch (63 x 54 mm). Sternal punction confirmed the peripheric thrombopenia. Amoxicillin-clavulanate (1 g x 3 per day), ofloxacine (200 mg x 2 per day) and gentamycin (150 mg per day) were started after bacteriological samples (blood, urine and vaginal cultures). Twenty-four hours later, 3/3 blood cultures became positive with small gram-negative rods. Urinalysis was negative. Microbiological identification confirmed a multi sensible *HPI*. Transthoracic and transoesophageal echocardiography were performed and confirmed the diagnosis of endocarditis, with vegetation on the mitral valve and a mitral prolapse. Left ventricular function was normal, but mitral regurgitation was evaluated Stage III. Antibiotherapy was switched to amoxicillin (12 g per day) plus gentamycin. Antinuclear factors, rheumatoid factor, complement and antiphospholipid antibodies were negative. No immunosuppression or sexually transmitted diseases were diagnosed (B and C hepatitis, TPHA VDRL and HIV serologies were negative). Because of persistent abdominal pain, laparoscopy was done ten days after initiation of antibiotherapyin. Examination of the pelvic cavity revealed the presence of moderate adhesions of the ileum, and a right abscess in the Douglas pouch. The ovaries were normal and an abscess had developed on an underlying pseudo-cyst of endometriosis. Drainage of the cavity was performed. Culture and 16S ribosomal RNA gene sequencing of perioperative sampling were negative, probably because of previous antibiotherapy. Biopsy was performed in the Douglas pouch and histological analysis showed typical lesions of endometriosis, with inflammatory reaction and altered polymorphonuclear leukocytes. After 24 hours, fever disappeared, abdominal pains and inflammatory syndrome decreased and further outcome was favourable. Control blood cultures were negative. After three weeks of amoxicillin, results of MIC allowed switching to ceftriaxone 2 g a day IM, for three weeks to simplify the treatment. Control of

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echocardiography showed stability of valvular damage, with persistence of mitral regurgitation Stage III. There have been no manifestations of valvular infection during the five following months and mitral valvuloplasty was performed to correct the valvular defect in September. LHRH analogues were dispensed to treat the endometriosis after this acute infection.

## Discussion

This patient had an IE due to HPI (positive blood cultures and echocardiographic findings). *Haemophilus spp* are a rare responsible pathogen of IE (3%) [8]. Darras-Joly *et al*. reported 42 cases of *Haemophilus spp* endocarditis [9]: HPI was the main pathogen (26/42), like in Vasquez *et al*'s study. In this study, the mean age was 27 years, 60% of patients had no identifiable predisposing illness [10]. Although surgery is often necessary, HPI endocarditis has a favourable outcomeprognosis [9].

In previous reports, origin of the micro-organism was unknown in 70-80% of cases and no case of gynaecologic infection was noted. In our case, a pelvic abscess was present. The lack of isolation of HPI from this site may be explained by the fact that samples were performed ten days after the onset of antibiotherapy. The role of HPI in genital infections has been described. HI is a small gram-negative rod that commonly inhabits the upper respiratory tract. HI biotypes II and III are found to predominate among strains from the respiratory tract and biotypes II and IV among strains from the genital tract. HPI, biotype II, is most frequent in both sites [11]. It has rarely been isolated in tuboovarian abscesses, salpingitis, endometritis and other obstetrical infections. In Vasquez *et al's*. study [6], *Haemophilus spp* was isolated from 2.8% of 5,572 genital specimens; of whom HPI was detected in 64.5% and HI in 29% of cases. Their pathogenic role is evocated when it is isolated as the single pathogen in infections as urethritis in men or Bartholin's abscess in women [6]. To explain urogenital infection with HPI, few hypotheses have been suggested: role of orogenital sexual contact or colonization by a reservoir in the colon. Genital infections with bacteremia caused by *Haemophilus spp* have been described [7], suggesting a possible risk of endocarditis in case of underlying valve disease. In our case, the origin of infection was probably gynaecologic.

Since it is of outstanding to describe HPI endocarditis from genital infection, it is a hypothesis that endometriosis may have favoured this rare complication. Occurrence of tuboovarian abscess seems to increase in women with endometriosis [12], particularly in women with Stage III and IV, at the age between 20 and 29 and those older than 40 [3]. It has been suggested that endometriosis could be responsible for impairment of local immunity (particularly cystic wall and ovarian epithelium) and that the presence of old blood in an endometrioma may provide a culture medium for bacteria to grow slowly after transvaginal inoculation and facilitate the spread of infection. Thus, infected endometriosis could be a risk factor for endocarditis, like any other infected site (dental, digestive...), particularly in case of underlying heart disease. The most probable mechanism is a bacteremia but we can wonder if migration of cells (like catamenial pneumothorax) could be incriminated.

In conclusion, we report an unusual case of HPI infective endocarditis probably related to endometriosis with pelvic abscess. Evolution was favourable with adapted antibiotherapy and surgery treatment. The patient remained asymptomatic for 15 months. The present case report suggests that in women with *Haemophilus spp* endocarditis, it may be of interest to detect pelvic inflammatory disease or endometriosis if no origin of infection (particularly dental) is identified. Gynaecologic tract infection is probably an underestimated cause of endocarditis, particularly in young women. It highlights the importance of considering the potential role of endometriosis in these settings. Prospective larger cohorts of patients with endometriosis would be necessary to define incidence and types of infective complications. Practioners should consider *Haemophilus* species as a potential pathogen in extra respiratory or oto-rhino-laryngologic infections.

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