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Total hip prosthesis complication, periprosthetic infection with external fistulizing due to *Enterobacter cloacae complex* multiple drugs resistance: A clinical case report

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ABSTRACT

INTRODUCTION: The *Enterobacter cloacae* is a microorganism found in the intestinal flora of the majority of animals, including humans. Primary infections caused by *E. cloacae* are rare in immunocompetent patients, but are very common in hospital settings in newborns and immunocompromised patients, and can be aggravated by the insurgence of antibiotic resistance. The incidence of periprosthetic hip infections is just below 2%.

CASE PRESENTATION: A 76 year old woman with multiple comorbidities underwent surgical implantation of intermediary total hip prosthesis of the left hip, in a different health facility, in February 2014, after the basicervical fracture of the upper femur extremity due to trauma. After an episode of dislocation of the prosthetic implant, in September 2014, she underwent a surgical operation to implant the acetabular component. A month later not in our facility, following a re-hospitalization for the dislocation of the arthroprosthesis, an infection from *E. cloacae complex* was discovered. After 2 years of chronic infection she came to our attention; the clinical picture featured coxalgia and secreting fistula in the surgical wound. Following a specific antibiotic therapy, carried out intravenously over the course of a month, we decided to intervene removing the left hip arthroprosthesis and placing an antibiotic spacer following the direction deduced from the antibiogram study of August 2016.

CONCLUSION: The patient was hospitalized in our facility and 2 months later she underwent another operation to remove the antibiotic spacer and to place a new total hip arthroprosthesis. Multiple swabs showed the complete healing from the infection, which was confirmed a couple of months later.

1. Introduction

The *Enterobacteriaceae* are ubiquitous microorganism found in soil, water, and plants, and are part of the intestinal flora of the majority of animals including humans. These bacteria cause a variety of human pathologies, a third of all bacteremia, more than 70% of the urinary tract infections and many intestinal infections. Some microorganisms (*Salmonella typhi, Shigella* spp., *Yersinia pestis*) are always associated with human pathologies, while others (*Escherichia coli, Klebsiella pneumonie, Proteus mirabilis*) are part of the normal commensal flora and can cause opportunistic infections [1].

Enterobacteriaceae infections can originate from an animal reservoir, a human carrier or from an endogenous diffusion of the microorganism.

The *E. cloacae* is a Gram-negative bacterium, it can grow rapidly in aerobiosis and anareobiosis, in a large variety of selective and non-selective media.

The primary infections caused by *E. cloacae* are rare in immunocompetent patients, but are very common in the hospital setting in newborns and in immunocompromised patients, and can be aggravated by the insurgence of the antibiotic resistance.

2. Case presentation

A 76 years old woman, suffering from senile dementia, anxiety-depressive syndrome, lower extremities venous insufficiency, biliary lithiasis, spondylosis, chronic obstructive pulmonary dis-

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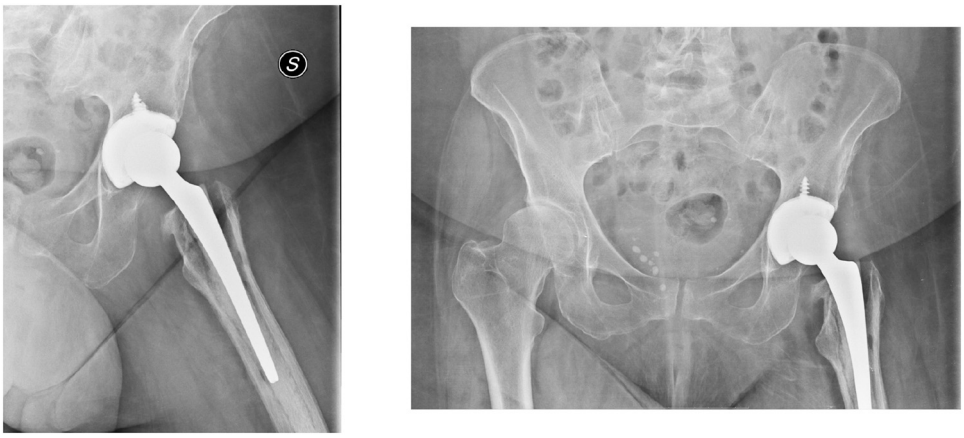

Fig. 1. The images highlighted an infectious/inflammatory process in the acetabular system.

ease, obesity and arterial hypertension, in February 2014, following a road accident, suffered a basicervical fracture of the left femur and underwent a surgical operation to place an intermediary total hip prosthesis in a different health facility, on the 6th of February 2014.

In September 2014, following a trivial trauma, the patient suffered from dislocation of the intermediary total hip prosthesis implant and, on the course of that operation, on the 22nd of September 2014, the acetabular component was implanted.

About a month after the acetabular component implant, the patient suffered a new dislocation of the arthroprosthesis, which was treated with a closed reduction, on the same premises of the first operation; simultaneously, some swabs of the surgical wound were made, which tested positive to *E. cloacae*.

In January 2016, the patient started lamenting an ingravescent coxalgia on the left side, with functional limitation and the appearance of secretion through sinus on the previous surgical scar; the following month, the patient developed a urinary tract infection, with a positive urine culture for *E. cloacae* and *E. coli*.

The monitoring of the inflammation indexes showed an ESR of 86 and a CRP of 2.71.

Later, a cutaneous swab of the wound was repeated, indicating again a positivity to *E. cloacae*, therefore, an antibiotic therapy of Ceftazidime 1fl/8 h IM for 14 days was carried out, followed by Imipenem 1fl/8 h for 2 more weeks.

In March 2016, the inflammation indexes were still high, with an ESR of 112 and a CRP of 1.71.

In May 2016, a new cutaneous swab of the wound was repeated, and again resulted positive for *E. cloacae*; the inflammation indexes were mostly unchanged.

An infectious diseases consultancy was asked for our patient, who was then prescribed a new antibiotic therapy of Ceftazidime 2 g/12 h IM and was suggested to remove the prosthetic implant.

In June 2016, the patient underwent new diagnostic investigation: new pelvis RX and leukocyte-marked scintigraphy; the images highlighted an infectious/inflammatory process in the acetabular system, in the greater-lesser trochanter zone and along the femoral stem. Elevated inflammation indexes: ESR 74 and CRP 2.28 (Fig. 1).

The patient came to our attention on the 27th of June 2016, with a clinical picture characterized by severe coxalgia on the left, tumefaction above the previous surgical wound and purulent fistula. At the moment of hospitalization, the previously prescribed antibiotic therapy continued.

On the 2nd of August 2016, she underwent in our facility the total removal of the left hip arthroprosthesis and of the antibiotic spacer, and the placement of cerclage bone wires: (Fig. 2) alongside that operation, intraoperative swabs were made to confirm the infectious process from *E- cloacae*. Consequently, we asked for a new infectious disease consultancy to update the antibiotic therapy with Meropenem 1 g bolus IV over 2 h, followed by 1 g every 6 h, in addition Amikacin 15 mg/kg/24 h.

The patient, hospitalized in our ward, continued the antibiotic therapy for about 2 months, with slight changes. The clinical course was complicated by the worsening of the anxious depres-

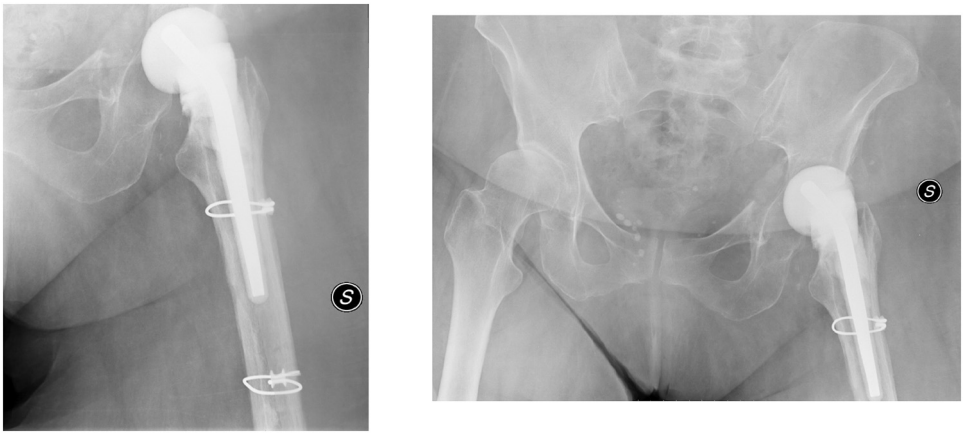

Fig. 2. The images show the antibiotic spacer, and the placement of cerclage bone wires.

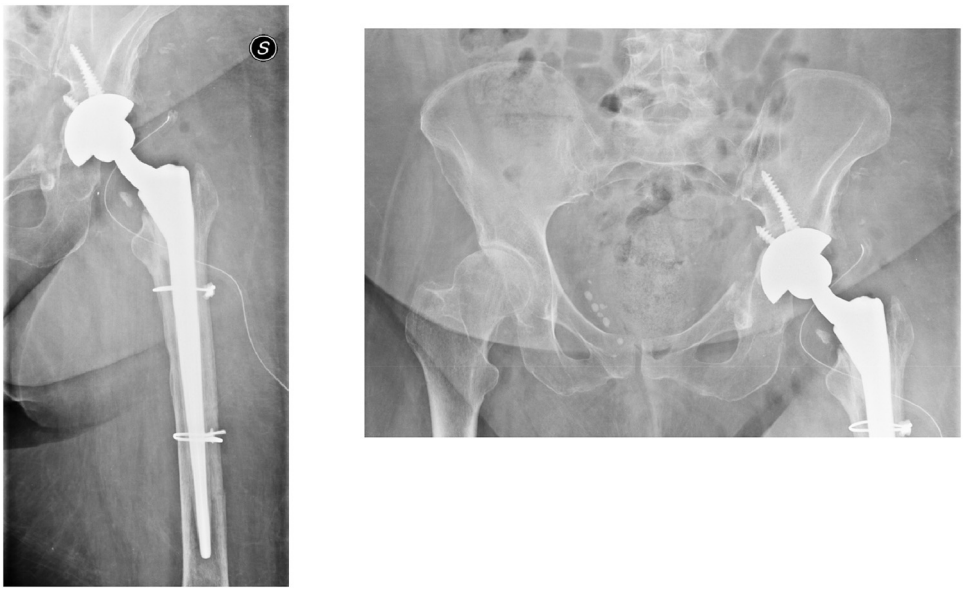

Fig. 3. New left hip revision prosthesis.

sive syndrome, of the chronic obstructive pulmonary disease and by a urinary trait infection from *Enterococcus faecium*.

On the 31st of October 2016 the patient underwent another surgical treatment to remove the antibiotic spacer covered with Meropenem and Amikacin and to place a new left hip revision prosthesis, stabilized by 3 screws over the acetabulum.

A fragment of the antibiotic spacer (Fig. 4) was examined on the scanning electron microscope (SEM) to verify the eradication of the infection. In the SEM picture (Fig. 5) the rod-shaped *E.cloacae* are evident, adhered to the biological matrix which presents blebs.

During the operation, new samples were taken for culture tests, and they all came back negative.

The inflammation indexes, during the following two weeks, went towards normalization, with the last results of ESR of 9, PCT of 0.03 and CRP of 1.9 and consequent interruption of the antibiotic therapy.

The correct positioning of the arthroprosthesis was confirmed by post-operation X-ray (Fig. 3).

The patient was discharged 30 days after the hip revision prosthesis operation, and transferred in the physical and rehabilitative medicine ward in another facility, for a specific rehabilitation program.

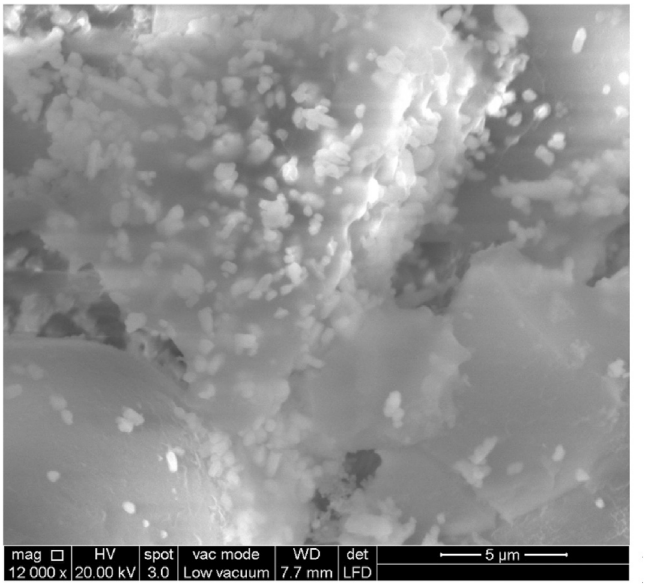

Fig. 4. A fragment of the antibiotic spacer was examined on the scanning electron microscope (SEM) to verify the eradication of the infection.

3. Discussion

The incidence of periprosthetic hip infections is just below 2%; usually the pathogens responsible for these infectious process are the more common gram-positive cocci: *S. aureus*, enterococci and streptococci; among the gram-negative it is worth mentioning the *P. aeruginosa* [2].

Periprosthetic hip infections are caused by gram-positive cocci: coagulase-negative staphylococci and *S. aureus* are responsible for 50–60% of the cases; moreover, lately, a rise of methicillin-resistant strains has been noticed [3].

Other gram-positive bacteria are the enterococci, representing 3–7% of the cases, while the streptococci are responsible for 9–10% of the cases. Gram-negative bacteria, specifically *P. aeruginosa* and some representative of the *Enterobacteriaceae* family are the cause of infection in 3–6% of the cases and are rather uncommon.

It is worth mentioning that the microbial contamination of the prosthesis can occur during the operation through airborne contamination or direct inoculation during the handling of the prosthesis [4].

Alternatively, the microorganism can reach the prosthesis by the hematogenous route for a bacteremia or from a septic source adjacent. Prosthetic infections are usually caused by the presence of microorganism growing inside a structure called biofilm [5].

The pathogenesis is determined by the interaction of the microorganism and the biofilm production, from the inflammatory response of the host.

The prosthesis has no blood circulation, therefore the local penetration of the antibiotic is impossible; consequently, on the surface of the prosthesis, an extracellular matrix with high concentration of water is formed, and the microorganism shifts from a planktonic phase to a sessile phase with low energy requirement [6].

Studies have shown that overweight and obesity can be influenced by hereditary factors (genes) and by the presence of some

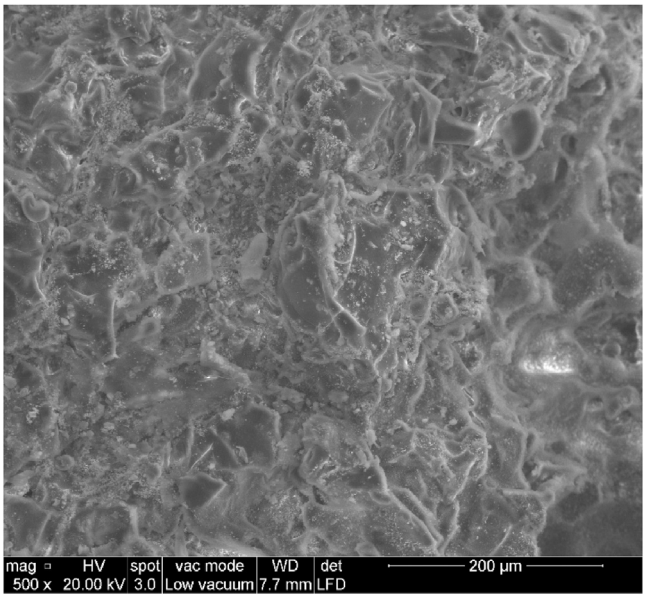

Fig. 5. In the SEM picture the rod-shaped *E.cloacae* are evident.

bacteria, like *E. cloacae* in the gastro-intestinal flora: those are the new research areas of the fight against obesity [7].

Our patient's infection from *E. cloacae* proved to be rare and particularly difficult to eradicate. The sole antibiotic therapy was of little benefit, therefore we had to resort to the revision of the prosthetic implant, in two times, a complex operation, with the consequent prolonged hospitalization of the patient in our facility and complications derived from that fact [8].

4. Conclusion

We are satisfied with the result obtained at the end of the clinical/surgical treatment we proposed, bearing in mind that the patient's comorbidity could have further complicated the clinical picture, leading to the non-resolution of the infectious process. The importance of the result is even more valuable since the pathogen causing the infection is very rare. A correct multidisciplinary approach has allowed a good resolution of the clinical case.

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Conflict of interest

The authors have no conflicts of interest to declare.

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Consent

Written informed consent was Obtained from the patient for publication of this case report and Accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

V. Amorese: study concept or design.
M. Corda: writing the paper.
M. Donadu: writing the paper.
D. Usai: writing the paper.
F. Pisanu: others.
F. Milia: others.
F. Marras: others.
A. Sanna: others.
D. Delogu: others.
V. Mazzarello: writing the paper.
G. Manzoni: others.
M. Conti: writing the paper.
G.B. Meloni: writing the paper.
S. Zanetti: writing the paper.
C. Doria: writing the paper.

Ethical approval

A written informed consent was obtained from the patient before the preparation of manuscript.

Guarantor

Veronica Amorese. Veronica.amorese@gmail.com.

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