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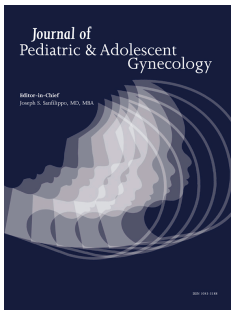

Lethal necrotizing cellulitis caused by ESBL-producing E. coli after laparoscopic intestinal vaginoplasty

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Lethal necrotizing cellulitis caused by ESBL-producing E. coli after laparoscopic intestinal vaginoplasty

Running head: Fatal ESBL-EC necrotizing cellulitis

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Key words. Vaginoplasty; Reconstructive Surgical Procedures; Necrotizing Fasciitis; Postoperative Complications.

ABSTRACT

Background. The absence of a functional vagina has a negative effect on the (sexual) quality of life of (transgender) women. Multiple surgical procedures have been described for vaginal reconstruction in these patients.

Case. We present a case of an 18-year-old transgender woman, who underwent laparoscopic intestinal vaginoplasty as vaginal reconstruction and subsequently developed septic shock and multiple organ failure based on an extended-spectrum beta-lactamase-producing *E. coli*. A severe progression of the necrotizing fasciitis was lethal, despite repeated surgical debridement, intravenous antibiotics and supportive care at the intensive care unit.

Summary and conclusion. Though vaginal reconstruction has a positive influence on the quality of life in (transgender) women, physicians and patients need to be aware of serious complications that may arise. Luckily, these complications arise rarely.

BACKGROUND

The absence of a functional vagina has a negative effect on the (sexual) quality of life of biological women and transgender women. Multiple surgical procedures have been described for vaginal reconstruction in these patients. In transgender women, penile inversion vaginoplasty is performed most frequently. Intestinal vaginoplasty is mentioned as a surgical alternative. [1]

Extended-spectrum beta-lactamases-producing organisms (ESBL) can cause life-threatening infections. The prevalence increases worldwide; percentages up to 43.5% of fecal carriage have been reported.[2] As it is multi-drug resistant, adequate treatment poses a challenge. We report on an 18-year-old transgender woman who developed necrotizing cellulitis caused by ESBL-producing *E. coli* as lethal complication after laparoscopic intestinal vaginoplasty.

CASE

An 18-year-old, healthy, transgender woman underwent total laparoscopic sigmoid vulvo-vaginoplasty as gender confirming surgery. Standard penile inversion vaginoplasty was not feasible, due to penoscrotal hypoplasia, which resulted from previous treatment with puberty suppressing hormones.[3] Transgender women with early-onset gender dysphoria, treated with puberty suppressing hormones, report fewer behavioral and emotional problems and an improvement of general functioning.[4] During surgery, a pedicled sigmoid segment was laparoscopically isolated and brought down to the dissected vesicorectal space. Intestinal continuity was restored by intra-abdominal side-to-side anastomosis. The procedure lasted 210 minutes without intra-operative complications. Cefuroxim and Metronidazole were administered intravenously as antibiotic prophylaxis.

After 24 hours, the patient developed abdominal pain, started vomiting bile and increased C-reactive protein (330 mg/L, normal < 5 mg/L) was noted. Due to a stable condition of the

patient, normal vital parameters and absence of visible signs of disease, no operative measures were undertaken. A gastric tube was placed and extra fluid was administered intravenously. After three days, an abdominal CAT-scan revealed a dilated small bowel and colon, multiple air-fluid levels, and free gas in the abdominal cavity, indicating anastomotic leakage. During emergency laparotomies, three and five days after initial surgery, anastomotic or air leakage was not observed. Intraperitoneal fluid was cultured and showed *Escherichia coli, Streptococcus viridans* and *Enterococcus faecium*. Vancomycine (1 gram) was administered. The patient was transferred to the intensive care unit (ICU) because of respiratory instability. A livid, non-blanchable skin lesion developed at the right flank (Figure 1A). The skin lesions rapidly spread in a few hours, expanding to the proximal upper legs (Figure 1B&C). She became inotrope-dependent and necrotizing fasciitis was suspected (Figure 2). During a third re-laparotomy, a large volume of brown, non-fecal fluid was aspirated and Extended-Spectrum Beta-Lactamase-producing *E. coli* (ESBL-EC) was cultured. Necrosis of subcutaneous fat and Scarpa's fascia was observed (Figure 3), but the underlying deep fascia and musculature were not affected. No air bubbles were present. The strain of ESBL-EC could not be matched to cultures of other admitted ICU-patients. The pathology report revealed necrotizing subcutaneous inflammation and presence of gram-negative, rod-shaped bacteria. Imipenem and clindamycin were administered intravenously.

Hemodialysis was started because of multiple organ failure (MOF) and further expansion of skin lesions was observed. About 60% of the body surface of subcutaneous tissue and skin was removed, at which the cutting edges appeared vital and the deep fascia was intact. However, the patient developed hypothermia (30.5°C) and hypoglycemia and deceased due to an irreversible septic shock with MOF based on necrotizing cellulitis caused by mixed (facultative) anaerobes, including an ESBL-EC.

SUMMARY AND CONCLUSION

We presented a case of lethal necrotizing cellulitis caused by ESBL-producing *E. coli* after laparoscopic intestinal vaginoplasty in a young transgender woman. Though vaginal reconstruction has a positive influence on the quality of life in non-transgender and transgender women, physicians and patients need to be aware of serious complications that may arise. Bouman et al., conducted a review of literature and reported all described surgical complications after sigmoid vaginoplasty. Severe complications arise rarely (0.6%). Reported severe complications, comprised necrosis of the sigmoid conduit, bilateral lower extremity compartment syndrome, intraluminal abscess in the top of the neovagina and necrotizing fasciitis. Mild complications occur in 4-5% in cases and comprise intra-operative damage to the rectum or bladder, intraoperative bleeding that requires blood transfusion, wound and urinary tract infections, hematomas, (mechanical) ileus, peritonitis and introital stenosis.[5] Generally, the procedure results in satisfactory functional and esthetic outcomes and a good patient-reported quality of life in transgender women. [6]

ESBL-producing bacteria can cause life-threatening infections. Risk factors for acquiring an infection are prolonged hospital stay, invasive medical devices, recent surgery, hemodialysis, decubitus ulcers, poor nutritional status, administration of antibiotics and being immunocompromised.[7] In our case, recent surgery was the only risk factor present. One of the main ESBL-producing organisms is *E. coli*.[8] The mortality rate of a bacteremia caused by ESBL-EC is high, up to 61% versus 24% for non-ESBL-EC.[9] The presence of several virulence factors and β-lactamase production contribute to the pathogenesis of ESBL-EC.[10] Therapeutic options are limited. Carbapenems are the treatment of choice, combined with wide surgical debridement, and supportive care.[8, 11] In hospitals, ICUs are often the origine of ESBL-producing organisms.[2] Genotypically similar strains have been identified in multiple patients, implying patient-to-patient transmission. In this case, no other patients at the ICU were identified with the causative pathogen, suggesting our patient was colonized before admission.

Necrotizing soft tissue infections (NSTIs) can be divided based on their anatomical location (i.e., Fournier gangrene), infection depth, and associated microbiological organisms. Typically, multiple deep layers of the body are involved, but in our patient only skin and subcutaneous tissue were affected, indicating leakage of fluids and gas of the perineal wound through the perineal tunnel. This leakage was not seen during re-laparotomy, suggesting a non-detected wound infect of the deeper layers. According to microbiological findings, three types of necrotizing cellulitis can be distinguished. Type I is a polymicrobial, often bowel flora-derived infection, type II a monomicrobial infection, usually caused by group A β-haemolytic streptococcus and type III is caused by gram-negative, often marine-related organisms.[7] Our patient suffered type I necrotizing cellulitis caused by mixed (facultative) anaerobes, including ESBL-EC. Rapid surgical intervention remains essential for survival, but unfortunately could not prevent rapid deterioration of our patient's condition with fatal consequences.

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FIGURE LEGENDS

Figure 1. Livid discoloration of the skin at post-operative day 5 at the ICU, right flank (A)**, ,** Expansion of livid skin discoloration post-operative day 6 at the ICU. (B) Right flank. (C) Inguinal and left upper leg.

Figure 2. The skin lesions expanded in merely a few hours. (A) Left flank. (B) Inguinal and upper legs. (C) Right flank.

Figure 3. Inspection during emergency surgery showed subcutaneous fluids and necrosis of subcutaneous fat and the fascia of Scarpa.