



Case report

Streptococcal sex syndrome: a curious association between sex and cellulitis

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ABSTRACT

Women undergoing pelvic lymphadenectomy and radiation therapy for gynaecological cancer are prone to acute and often recurrent cellulitis as a consequence of compromised lymphatic circulation. Vaginal intercourse can trigger the infection, a condition named *streptococcal sex syndrome* (SSS). We report a 63-year old female patient with a history of pelvic lymphadenectomy and radiation therapy for gynaecological cancer. She presented to the obstetrics/gynaecology department for vaginal haemorrhage during sexual intercourse the day before. Gynaecological examination was unremarkable but she was febrile and presented inflammatory signs on the abdomen and lower left limb. The diagnostic workup suggested cellulitis and intravenous antibiotics were initiated, with complete recovery. Blood cultures were positive for *Streptococcus mitis/oralis*. One month later, the infection recurred at the same location, promptly after sexual intercourse.

1. Introduction

Women undergoing pelvic lymphadenectomy for gynaecological cancer, with or without radiation therapy, are prone to develop acute and often recurrent cellulitis as a consequence of local anatomic abnormalities, compromising lymphatic circulation (Dankert and Bouma, 1987; Bouma and Dankert, 1988). The infection, which usually manifests in the abdominal wall and lower limb, is thought to occur by shunting of bacteria away from the affected intrapelvic and inguinal lymphatics, into the soft tissues of the lower extremities (Ellison III and McGregor, 1987). A minor skin trauma in the area of impaired drainage is usually the precipitating event but vaginal intercourse may be the trigger in a subset of patients, an association that was described in 1987 (Ellison III and McGregor, 1987).

2. Case Presentation

A 63-year-old woman was admitted to our hospital's emergency department (ED) on October 2016 with fever and inflammatory signs on the skin of the abdomen and lower left limb. She initially presented to the obstetrics/gynaecology department complaining of vaginal haemorrhage during sexual intercourse the day before but, after an unremarkable gynaecological examination, was referred to the general ED. She reported pain in her left thigh since that morning, associated with malaise and shivering. Past medical history was significant only for cervical epidermoid carcinoma, stage IB1, diagnosed in June 2013,

subjected to radical hysterectomy with bilateral adnexectomy and pelvic lymphadenectomy in September 2013, followed by external pelvic radiation and vaginal brachytherapy, completed in January 2014. No other relevant health problems and no allergies were reported. On examination, the patient was alert and oriented, febrile (38.5C degrees), tachycardic and hypotensive. The left thigh had exuberant inflammatory signs, with pain, erythema and oedema extending from the lower abdominal wall to the left knee (Fig. 1). Intravenous fluids were started which corrected the patient's hypotension. Cellulitis was suspected and the patient initiated intravenous amoxicillin/clavulanate and clindamycin, after collecting urine and blood cultures. Bloodwork showed leucocytosis with increased reactive C protein, with normal kidney and liver function. A pelvic computed tomography (CT) was performed to exclude abscesses or evidence of gas formation, and found signs of increased dermis thickness, related to cellulitis. The patient was transferred to the ward where she presented a favourable clinical course and was discharged on the fourth day on oral amoxicillin/clavulanate and clindamycin. The blood cultures were positive for *Streptococcus mitis/oralis*, susceptible to the antibiotics prescribed. One week later, she was seen on the outpatient clinic and was well, with no signs of infection.

One month after discharge, the patient returned to the ED, again complaining of fever and inflammatory signs on the left thigh. She reported sexual activity on the previous day. Once again she was admitted for cellulitis and treated with intravenous ceftriaxone and clindamycin with a favourable course. Microbiological cultures yielded no

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Fig. 1. Left leg erythema secondary to Streptococcal cellulitis.

results. She was discharged two days later with advice to moderate her sexual activity and a follow-up gynaecologic evaluation was requested so that an effective prophylactic strategy, to avoid further episodes, could be established.

3. Discussion

In the 1980s there were a series of reports of acute and often recurrent cellulitis in women who had undergone local lymphadenectomy for gynaecologic cancer (Dankert and Bouma, 1987; Bouma and Dankert, 1988; Ellison III and McGregor, 1987; Binnick et al., 1980; Baddour and Bisno, 1985). The events were exclusive or more frequent in patients subjected to postoperative radiation of the pelvic area, possibly due to additional impairment of lymphatic drainage. Cellulitis presented clinically with erythema over the thigh, the inguinal region or the lower abdominal wall associated with signs of systemic infection. When pathogens were identified they were mostly non-group A, beta-haemolytic streptococci. Antibiotic prophylaxis (e.g. monthly intramuscular penicillin) was effective in preventing recurrences in almost all of these patients, in contrast to patients who did not receive prophylaxis.

In 1987, Ellison III et al. described a curious temporal relationship between vaginal intercourse and the onset of cellulitis in two patients (Ellison III and McGregor, 1987). One had a history of recurrent nodular sclerosing Hodgkin's disease submitted to a staging laparotomy, followed by radiotherapy and multiple chemotherapeutic regimens. Disease developed in right inguinal nodes. After treatment, she began experiencing various episodes of fever and erythematous macular rash on her right leg, where she had a residual chronic lymphedema. All episodes had developed approximately 24 h after vigorous and prolonged sexual intercourse. *Streptococcus agalactiae* was repeatedly isolated from blood and vagina. She was advised to moderate her sexual stimulation and began using a water-soluble lubricant jelly during subsequent coitus. No further episodes have occurred in the follow-up period. The other patient was a pregnant woman who had undergone a modified radical vulvectomy with inguinal lymphadenectomy for squamous cell carcinoma of the labia majora, two months earlier. Cellulitis developed approximately one hour after the patient's first postoperative vaginal intercourse. One month later, the infection recurred at the same location promptly after sexual intercourse. It was postulated that in both of these cases the vaginal region was the site of inoculation after microtrauma from coitus. The authors named this new entity *streptococcal sex syndrome* (SSS).

Our patient provides a very similar description, referring to her

sexual intercourse as vigorous and confirming its timing to the onset of symptoms. The loss of blood in the first episode is also suggestive, because it may have resulted from a disruption of the genital tract mucosa barrier. As the situation was building up a great deal of anxiety in our patient she was referred to a follow-up gynaecologic evaluation, where she was given additional advice on her sexual activity. She also began using a lubricant jelly during coitus. This proved to be an effective prophylactic strategy with no recurrences after 1 year of follow-up.

The only microbiological isolate was of *Streptococcus mitis/oralis* from two blood cultures. *Streptococcus mitis/oralis* belongs to the viridans group of Streptococci. It is an alpha-haemolytic gram-positive coccus and a normal commensal of the oropharynx, skin, intestinal and female genital tracts (Ruoff, 2002; Kutlu et al., 2008). Despite its low virulence and pathogenicity, it can cause, under certain circumstances, serious infections like bacteraemia, endocarditis, meningitis, orbital cellulitis and streptococcal toxic shock syndrome (Kutlu et al., 2008; Lyytikainen et al., 2004; Ng et al., 2005; Madhusudhan et al., 2007; Seltz et al., 2011). It may also be a contaminant in blood cultures, although, in our case, the fact that it was isolated from both blood sets (collected from separate venipuncture sites), makes that possibility unlikely. In some cases, vaginal swab cultures can provide additional supporting evidence, if the same microorganism is isolated (Ellison III and McGregor, 1987).

We recommend that the initial empiric antibiotic therapy of SSS should provide activity against beta-haemolytic streptococci and *Staphylococcus aureus*, as other points of inoculation may be implicated, namely the skin.

After treatment of the acute episode, prevention is of paramount importance in SSS. If the behavioural measures, successfully implemented in our patient, prove to be ineffective, antibiotic suppressive therapy may be necessary. The available data seems to support this approach, as was previously mentioned. Both daily penicillin V and monthly intramuscular penicillin were used with good results, but the trials did not included patients with recurrent infection related to sexual intercourse (Bouma and Dankert, 1988; Binnick et al., 1980).

In conclusion, this is a case report from a somewhat forgotten and therefore most likely underdiagnosed syndrome. Only a careful medical anamnesis, with attention to a detailed sexual history, can provide the clues to its timely diagnosis and prevention.

Conflicts of interest

The authors declare that there are no conflicts of interest regarding the publication of this article.

Author contribution

Mario Bruno Santos: writing of manuscript.
 Rita Félix Soares: writing of manuscript.
 Filipe Basto: critical review of manuscript.

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