

# Rare Postoperative Pelvic Lymphocyst Infection by *Streptococcus mitis/oralis*: A Case Study

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**Abstract:** Lymphatic cyst with infection is caused by interruption of lymphatic vessels during para-aortic lymphnode dissection in patients with gynecological malignancies. It is generally considered that the infection is caused by bacteria invading through the lower limb and perineal wound, but the pathogenic factors of most lymphatic cysts are not clear. Our literature search revealed no previously reported cases similar to ours, in which a lymphatic cyst infection resulted from a co-infection with *Streptococcus mitis/oralis* (*S. mitis/oralis*) and *Klebsiella pneumoniae* (*K. pneumoniae*). A woman of 61 years of age, was diagnosed in endometrial cancer, carcinosarcoma FIGO grade 3, stage IIIC of the FIGO system. During the third postoperative pelvic replacement and drainage procedure, the patient developed a fever with a peak temperature of 39.2°C on the second day after the removal of the drainage tube. Empiric broad-spectrum antibiotics were started upon hospital readmission, but the patient showed poor response to therapy, with persistent and recurrent lower abdominal tenderness, raising concern for ongoing intra-abdominal or pelvic infection. As a result, it is important to identify pathogenic bacteria associated with lymphatic cyst infections and determine antimicrobial regimens. We further investigated the pathogenicity and antibiotic resistance of *S. mitis/oralis*, formulated an appropriate anti-infective treatment strategies. Consequently, the patient recovered and was discharged.

**Keywords:** gynecological malignancies, lymphatic cyst, infection, *Streptococcus mitis/oralis*

## Background

Pelvic lymphocyst complicated by infection represents an inflammatory complication that arises secondary to pelvic lymphadenectomy. This pathological condition stems from the extensive intraoperative transection of regional lymphatic vessels, resulting in the abnormal accumulation of protein-rich lymph fluid within the retroperitoneal or pelvic spaces. The process ultimately forms an encapsulated cavity, known as a lymphocyst.<sup>1</sup> If the cystic cavity persists with poor drainage, it can be secondarily colonized and invaded by bacteria, particularly in the setting of impaired local immune defense or iatrogenic contamination, subsequently developing to an infected lymphocyst. Clinically, it often presents with persistent or remittent fever, elevated white blood cell count, and significantly increased inflammatory markers such as C-reactive protein (CRP) and procalcitonin (PCT). These findings are accompanied by dull pain and tenderness in the lower abdomen or deep pelvis. Some patients may present with urinary irritative symptoms, bowel dysfunction, or exacerbated unilateral or bilateral lower limb lymphedema. Severe cases can progress to pelvic abscess, sepsis, or even septic shock. Therapeutically, the combination of antimicrobial agents and percutaneous catheter drainage (PCD) is required, while surgical debridement is necessary if conservative measures fail. Above all, early identification and intervention of infected lymphocysts are crucial for preventing clinical deterioration, shortening the length of hospital stay and improving prognosis.

In our case, a patient with a postoperative diagnosis of Grade 3 uterine carcinosarcoma (FIGO Stage IIIC2) underwent laparoscopic surgery, including para-aortic lymphadenectomy. The patient received six cycles of adjuvant chemotherapy with the paclitaxel and carboplatin (TC) regimen and external beam radiotherapy following surgery. Following the third percutaneous drainage procedure, the patient developed a fever with a peak temperature of 39.2°C on the second day after drain removal. Percutaneous drainage and antibiotics initially controlled the purulent leakage and infection, however, the patient



subsequently experienced recurrent fever and lower abdominal pain. On the sixth day of admission, pus culture results revealed abundant growth of *S. mitis/oralis* complex and scant growth of *K. pneumoniae*, which was ESBL-negative. Following the adjustment of antibiotic therapy, the patient recovered and was discharged. This study aims to provide a valuable reference for the antimicrobial management of lymphatic cyst infections following gynecological oncology surgery.

## Case Presentation

A 61-year-old female who presented with fever for 2 days. Eight months ago, initial examinations suggested endometrioid endometrial carcinoma (FIGO Grade 3, Stage IA). Consequently, she underwent radical hysterectomy, bilateral salpingo-oophorectomy, pelvic and para-aortic lymphadenectomy, and partial omentectomy were performed. The final postoperative pathology confirmed uterine carcinosarcoma with upstaging to FIGO Stage IIIC. Following the third percutaneous drainage procedure, the patient developed a fever, with a peak temperature of 39.2°C. She went to an external medical facility and received a single intravenous dose of cefoperazone/sulbactam. On admission, the patient presented with a fever of 38°C and significant inflammatory markers: leukocytosis of  $11.5 \times 10^9/L$ , neutrophils of 90.4%, CRP of 138.8 mg/L, and PCT of 0.11 ng/mL. Upon diagnosis of infection, empiric antibiotics therapy was initiated with intravenous cefoperazone/sulbactam (Pfizer Inc. 1.5g) 3g every 12h, along with other supportive therapy. However, the patient highest temperature rose to 39.5°C. Laboratory results showed leukocytosis of  $9.0 \times 10^9/L$ , neutrophils of 89.6%, CRP of 137.7 mg/L, PCT of 0.13 ng/mL, and total bile acids of 6  $\mu\text{mol/L}$  on the second day of admission. Given the sustained high fever, the antibiotic regimen was immediately switched to intravenous meropenem ((Haibin Pharmaceutical Co., Ltd. 0.5g) 1g every 8 h. Simultaneously, 10g of human albumin Injection (Shandong Taibang Biological Products Co., Ltd. 20% 10g) was administered to provide nutritional support. Concurrently, due to persistent abdominal distension and right lower limb soreness, ultrasound-guided pelvic abscess catheter drainage was performed. Ultrasonography revealed a cystic mass measuring 12.1×5.0×2.9 cm in the right abdomen, with clear boundaries and an irregular shape. On the third day of admission, the patient remained febrile (peak 39.0°C). In the past 24 hours, 90 mL of turbid, light-brown fluid was drained. Considering the escalation of fever, the antibiotic regimen was adjusted to intravenous moxifloxacin (0.4g, Bayer Vital GmbH) combined with meropenem. Additionally, somatostatin (3mg, ALFASIGMA S.p.A) 3mg IV micro-pump was administered via continuous intravenous infusion to inhibit intestinal fluid secretion. Despite treatment, the patient remained febrile with a peak temperature of 38.7°C. On examination, there was localized tenderness in the right lower abdomen, but no rebound tenderness was elicited. The patient's temperature gradually decreased on day 5. On day 6, pus culture results revealed abundant growth of *S. mitis/oralis* and scant growth of *K. pneumoniae*. Drug susceptibility testing revealed sensitivity to penicillin, cefotaxime, and linezolid. Subsequent laboratory investigations showed a leukocyte count of  $3.9 \times 10^9/L$ , neutrophils of 84%, CRP of 64.6 mg/L, and PCT < 0.04 ng/mL. Guided by the pus culture results, the antibiotic regimen was adjusted to intravenous piperacillin/tazobactam (8:1) (4.5g, Zhuhai Federal Pharmaceutical Co., Ltd.) 4.5 g every 8 h and intravenous moxifloxacin 0.4 g every 24 h. On the ninth day of admission, the patient remained afebrile. With significant clinical improvement, the patient was successfully discharged.

## Discussion

### Main Pathogenic Bacteria and Related Treatment Options for Pelvic Lymphocysts

Lymphocyst is a common complication in gynecological cancer patients after lymph node dissection. Most lymphocysts are asymptomatic and gradually resolve spontaneously without specific treatment. However, a small number of lymphocysts can cause symptoms of varying degrees, including local pain, lower limb edema, secondary infection, compression of the ureter or blood vessels leading to hydronephrosis, ureteral dilation, or even lower limb venous thrombosis, which can severely affect postoperative quality of life and subsequent treatment.

Postoperative infected lymphocysts are generally thought to be caused by bacterial invasion through wounds in the lower limbs and perineum, with a pathogenesis similar to cellulitis. However, the predisposing factors for most such infections are not clearly identified. A retrospective analysis assessing risk factors and microbiological management of lymphocyst infection in patients undergoing systemic lymphadenectomy for gynecological cancer included 619 gynecological cancer surgery patients, of whom 115 developed postoperative lymphocysts, with an infection rate of 4.36%. This study identified that combined pelvic and para-aortic lymphadenectomy, a higher yield of removed pelvic lymph

nodes, lower postoperative serum hemoglobin levels, and an elevated neutrophil percentage were associated with a higher incidence of infected lymphocysts. Furthermore, a cyst diameter exceeding 60 mm was found to be an independent risk factor for infection.<sup>2</sup> These findings suggest that the etiological characteristics of infected lymphocysts differ significantly from those of gastrointestinal-derived pelvic abscesses, implying a unique pathogenic flora. Bacterial community profiles were analyzed using next-generation sequencing (NGS) in a recent study. Of the nine patients who developed infected lymphocysts following gynecological cancer surgery, three responded to conservative antibiotic therapy alone, while six required ultrasound-guided puncture and drainage. The majority of cases were characterized by monomicrobial infections, involving pathogens such as *Staphylococcus lugdunensis*, *Streptococcus agalactiae*, *Streptococcus equi*, *Enterococcus saccharolyticus*, and *Escherichia coli*. Appropriate antibiotics should be selected to effectively eradicate these specific strains.<sup>3</sup> A retrospective study conducted at the University of Tokyo Hospital identified Gram-positive cocci were the predominant pathogens following gynecological or urological surgeries.<sup>4</sup> According to Kawamura et al, lymphatic cyst infections are characterized by a monomicrobial derived mainly from skin or urogenital flora, with Gram-positive cocci and anaerobes being predominant. Based on this etiology, they advocate for empiric monotherapy using ampicillin-sulbactam or amoxicillin-clavulanate.<sup>5</sup> The management of infected lymphocysts typically involves antibiotic therapy and percutaneous catheter drainage. Recent studies indicated that mild infections responded to antibiotics alone, whereas severe infections necessitated adjunctive drainage. Notably, earlier drainage correlated with a reduced total treatment duration.<sup>6</sup>

Sclerotherapy is established as a safe and effective treatment for non-infected lymphocysts. The mechanism of sclerotherapy is to block lymph leakage by causing local inflammation and subsequent fibrosis of the lymphocyst. However, there is a paucity of literature specifically addressing the efficacy and safety of sclerotherapy in the context of infected lymphocysts.<sup>7</sup> Sclerotherapy for infected lymphocysts is indicated only after the cessation of suppuration, clinical stabilization, and normalization of inflammatory markers. Previous studies have demonstrated that sclerotherapy can significantly reduce drainage volume and help shorten the catheter indwelling time for infected lymphocysts in gynecologic oncology.<sup>8</sup>

In this case, the patient presented with high fever and was subsequently diagnosed with an infected lymphocyst. Initial empiric therapy with cefoperazone/sulbactam failed to resolve the symptoms. Several studies have demonstrated the potency of cefoperazone/sulbactam against commonly encountered pathogens causing intra-abdominal infections.<sup>9</sup> In vitro studies demonstrated that cefoperazone/sulbactam exhibited potency in vitro against *E. coli* and *K. pneumoniae*, irrespective of the presence of ESBL genes.<sup>10</sup> On the second day of admission, the antimicrobial regimen was switched to meropenem. Meropenem, a carbapenem antibiotic, exhibits potent activity against Gram-positive, Gram-negative bacteria, as well as anaerobes. However, it lacks activity against methicillin-resistant *Staphylococcus aureus* (MRSA). Furthermore, to ensure coverage of atypical pathogens, moxifloxacin was added to the regimen. The widespread utilization of antibiotics has led to a rising incidence of fluoroquinolone resistance in *Enterobacteriaceae*. As a result, clinical guidelines discourage the use of this drug class for such infections in areas where resistance rates are high. As the patient showed no significant improvement in antimicrobial therapy, presumably due to the large size of the lymphocyst, we recommend early source control via PCD or sclerotherapy. Following the drainage of purulent material, the patient experienced gradual defervescence and was eventually discharged.

## Drug Resistance and Antimicrobial Regimens for *Streptococcus mitis/oralis*

The *Streptococcus mitis* group includes *Streptococcus mitis* (*S. mitis*), *Streptococcus oralis* (*S. oralis*), *Streptococcus infantis*, *Streptococcus peroris*, and *Streptococcus cristatus*.<sup>11,12</sup> *Streptococcus mitis* is generally considered a low-virulence commensal of the oral microbiota. However, it can cause various invasive diseases, especially in neutropenic and immunocompromised patients. Notably, it is increasingly becoming a cause of bloodstream infections in patients receiving cytotoxic anticancer chemotherapy.<sup>13</sup> Data for England was extracted from the UK Health Security Agency's Second Generation Surveillance System (SGSS), based on 2023 data, the mitis group accounted for the largest percentage of non-pyogenic streptococcal bacteraemia. The pathogenic mechanism of *S. mitis* is not fully clear. It is known to successfully colonize the human oropharynx through various strategies, including secreting adhesins and immunoglobulin A proteases, forming

biofilm, and modulating the host immune system.<sup>14</sup> These colonization factors enable *S. mitis* to compete for ecological niches and nutrients with other oropharyngeal microorganisms.

In vitro antimicrobial susceptibility analysis of *S. mitis/oralis* often reveals significant therapeutic challenges, as a large proportion of isolates exhibit resistance to penicillin and cephalosporins, including third-generation cephalosporins like ceftriaxone.<sup>15</sup> Furthermore, vancomycin-resistant strains of *S. mitis* have also been identified.<sup>16</sup> Samuel et al performed multilocus sequence analysis and whole-genome analysis sequencing on 118 viridans group streptococci (VGS) strains isolated from the blood of patients at MD Anderson Cancer Center. Compared to patients infected with other viridans streptococci, those infected with *S. mitis/oralis* were more likely to develop moderate or severe clinical symptoms.<sup>17</sup> The management of infective endocarditis caused by *S. mitis/oralis* remains particularly difficult; despite in vitro susceptibility to vancomycin, the clinical efficacy is often unsatisfactory. Consequently, there is growing interest in the potential of lipopeptide antibiotics, such as daptomycin. Although clinical data are limited, daptomycin has been utilized to manage infections caused by  $\beta$ -lactam-resistant *S. mitis/oralis*.<sup>18,19</sup>

While *S. mitis/oralis* isolates were susceptible to  $\beta$ -lactams, the concurrent isolation of a hypervirulent *K. pneumoniae* strain complicated the clinical management. Given the increased complexity of the infection, piperacillin-tazobactam was administered. This regimen ensured comprehensive coverage of both isolates and represented an optimization of the therapeutic strategy.

## Conclusion

Pelvic lymphocyst infection is a recognized complication following radical surgery for gynecological malignancies, typically attributed to skin flora or enteric bacteria ascending from the perineum. However, this case presents a rare and complex microbiological profile: a co-infection with *S. mitis/oralis* and *K. pneumoniae*. *S. mitis/oralis* is generally considered a low-virulence commensal of the oral cavity, it has emerged as an opportunistic pathogen capable of causing severe invasive diseases, particularly in immunocompromised hosts such as cancer patients undergoing chemotherapy. The isolation of this organism from a pelvic abscess, rather than a bloodstream infection or endocarditis, is exceptionally uncommon and suggests a unique route of translocation, possibly hematogenous spread during periods of neutropenia or direct inoculation, although the exact mechanism remains unclear. The concurrent presence of *K. pneumoniae* further complicated management, requiring broad-spectrum antibiotics to cover potentially  $\beta$ -lactam-resistant Gram-positive cocci and Gram-negative bacilli simultaneously.

The clinical course of this patient underscores the limitations of antibiotic monotherapy in managing large, infected lymphocysts. Our experience reinforces the principle that “source control” is important; without adequate drainage of the purulent collection, even highly active antimicrobial regimens may fail. The subsequent adjustment to piperacillin/tazobactam combined with continuous drainage led to rapid defervescence and clinical recovery.

However, this study had some limitations. First, given the extensive 16S rRNA sequence homology (>99%) between *S. mitis* and *S. oralis*, conventional clinical diagnosis techniques including matrix-assisted laser desorption ionization-time of flight mass spectrometry fail to accurately and reliably differentiate between these two species.<sup>20,21</sup> As a result, isolates are frequently categorized broadly within the *S. mitis/oralis* group. Due to technical constraints in our laboratory, we were unable to employ advanced molecular techniques (such as whole-genome sequencing) to achieve definitive speciation, potentially obscuring species-specific epidemiological or virulence differences. Second, this is a single-case report, which limits the generalizability of our findings. The successful outcome may be influenced by patient-specific factors, such as her immune status and the specific anatomical characteristics of the lymphocyst, which may not apply to all patients with similar infections. Finally, the retrospective nature of the analysis means that certain data points, such as the exact bacterial load prior to drainage or detailed pharmacokinetic parameters of the antibiotics in the cystic fluid were not available.

In conclusion, implementing a protocol that mandates early imaging assessment and low-threshold drainage for large cysts, combined with empiric antibiotics covering both oral flora and enteric pathogens, may significantly reduce treatment failure rates and hospital stays.

## Data Sharing Statement

The data that support the findings of this study are available from Xiaoyu Cheng, but restrictions apply to the availability of these data, which were used under license for the current study, and therefore are not publicly available. Data are, however, available from the authors upon reasonable request and with permission of Xiaoyu Cheng.

## Ethics Approval and Consent to Participate

This study was approved by the ethics committee of the First Affiliated Hospital of USTC (approval no.2025-RE-450). We certify that the study was performed in accordance with the 1964 declaration of HELSINKI and later amendments. Institutional approval was not required for the publication of this case report.

## Consent for Publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. All the details of any images, videos, recordings can be published.

## Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

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## Disclosure

The authors declare that they have no competing interests in this work.

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