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CASE REPORT

Lemierre's syndrome and right adnexal abscess; A case report and review of the literature

Malak Al-Hakeem[1 3], Thomas Schneider[2], Cronen Thomas[2], and Ilmi Behluli[1]

[1] *Dept. of Gynaecology and Breast Surgery, Charité, University Hospital of Berlin, Hindenburgdamm 30, Berlin, Germany*
[2] *Dept. of Infect. Diseases, Charite University Hospital of Berlin, Campus Benjamin Franklin, Hindenburgdamm 30. Germany*
[3] *Department of Gynecology, King Saud University Medical College, Riyadh, Saudi Arabia*

ABSTRACT

Lemierre's syndrome is an extremely rare but a completely curable condition. A high index of suspicion is needed for early diagnosis and proper treatment. We report a 20 year old virgin Caucasian lady presented with sore throat, fever, neck pain, nausea and vomiting followed by acute abdomen. Her laboratory investigations revealed white blood count 13,300/mm^3, C-reactive protein 332 mg/L. Abdominal computed tomography scan showed pelvic abscess 6.2 x 6.1 cm mainly involving the right ovary. Intra-abdominal swab showed growth of *Fusobactrerium necrophorum*. Right adnexectomy and proper antibiotic was carried out. High index of doubt is the most important key in diagnosing this fatal syndrome. In our case the diagnosis of the syndrome was made only after the isolation of *F. necrophorum* from the intra-abdominal swab culture. *J Microbiol Infect Dis 2014; 4(3): 114-117*

Key words: Lemierre's syndrome; adnexal abscess; virgin

Lemierre sendromu ve sağ adneks absesi: Bir olgu sunumu ve literatür incelemesi

ÖZET

Lemierre sendromu çok nadir görülen ve tamamen tedavi edilebilen bir durumdur. Erken tanı ve uygun tedavi için çok erkenden şüphelenmek gerekmektedir. Burada boğaz ağrısı, ateş, boyun ağrısı, karın ağrısını takip eden bulantı ve kusmayla başvuran 20 yaşında evlenmemiş beyaz bir kadını rapor ettik. Laboratuvar testlerinde beyaz kan hücre sayısı 13.300/mm^3, C-reaktif protein değeri 332 mg/L idi. Batın bilgisayarlı tomografi incelemesinde başlıca sağ overi tutan 6,2 x 6,1 cm boyutlarında pelvik abse görüldü. Karın içinden alınan sürüntü kültüründen *Fusobacterium necrophorum* üremesi oldu. Sağ adneksektomi ve uygun antibiyotikle iyileşti. Bu öldürücü sendromun tanısında erken dönemde şüphelenme en önemli anahtar rolünü oynamaktadır. Bizim olgumuzda tanı *F. necrophorum*'un batın sürüntü kültüründen izolasyonu ile konuldu.

Anahter kelimeler: Lemierre sendromu, adneks absesi, bakire

INTRODUCTION

Lemierre's syndrome is also known as postanginal sepsis and necrobacillosis. The disease was first described by Courmont and co-workers in the early 1900s.[1] In 1936, the diseases was characterized by Lemierre. He reported 20 cases of oropharyngeal infections which were followed by the anaerobic sepsis.[2] According to epidemiological data, the number of reported cases reported as one per million yearly.[3] The definition of Lemierre's syndrome is not consistent, and there is an elementary differences in the literature regarding the site of the initial infection or the presence of a thrombophlebitis of the internal jugular vein (IJV). In our case, we diagnosed Lemierre's syndrome according to Riordan, stating "an infection arising in the pharynx with subsequent metastatic lesions.[1]

In a case report, Chayachinda et al. reported a case postpartum, post-sterilization tubo-ovarian abscess caused by *F. necrophorum*, with a full recovery after surgical and medical interventions[4]. To our knowledge, there are no published case reports of a tubo-ovarian abscess caused by *F. necrophorum* in a virgin woman.

Correspondence: Univ. Ilmi Behluli, Department of Gynaecological Oncology and Breast Surgery Charité, Universitätsmedizin Berlin, Campus Benjamin Franklin Hindenburgdamm 30, 12203 Berlin, Germany Email: ilmi.behluli@charite.de
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CASE REPORT

A 20-year-old virgin Caucasian lady from the United States of America, arrived in Germany on the May 16, 2013. Five days later, she started to complain of sore throat, lasting for several days. Ever since, she developed severe headache, followed by fever, photophobia, nausea, vomiting associated with minimal dry cough. One day later, she developed cervical pain, urging her to seek medical advice in our emergency department.

During physical examination, she gave a past history of enlarged cervical lymph nodes. In addition, she gave a history of tonsillectomy and adenoidectomy with insertion of bilateral tympanic aeration tubes. Her past gynecological history was irrelevant. No history of contraception use or the administration of sanitary tampons.

After the initial investigations in main Emergency Department which included a lumbar puncture, she was admitted to the Department of Infectious Diseases. On examination she was febrile (38.7°C), blood pressure, heart rate and percutaneous oxygen saturation were within normal ranges. Apart from small left cervical group lymphadenopathy, her physical examinations results were normal. Immediate investigation of the cerebrospinal fluid (CSF) showed normal values. The laboratory analysis showed raised inflammatory markers with a C-reactive Protein (CRP) of 332 mg/l and a white blood count (WBC) of 13.300/mm^3 with 85% neutrophils. Also, hepatic transaminases were slightly elevated (Aspartate transaminase (AST): 45 IU/L and Alanine Transaminase (ALT): 63 IU/L), albumin and bilirubin levels were normal (4.2 g/dcl and 0.8 mg/dcl respectively). Further investigations (blood cultures, microbiological/virological CSF-analysis, virological analysis for hepatitis, urine analysis, chest X-ray, initial ultrasound of the abdomen and pelvis) revealed no abnormal findings. During the first two days after admission, the patient remained afebrile with recession of the initial symptoms. Additionally, inflammatory markers dropped slightly.

At the third day after admission, her general condition deteriorated suddenly, developing fever up to 39°C, acute abdomen with local peritonism in the lower abdominal quadrants accompanied by signs of sepsis. Abdominal Ultrasound was performed at once showing a suspicious mass in the right lower abdomen adjacent to the appendix and the right ovary. The patient was consulted by general surgeons and gynecologists, and underwent a surgical exploration. The explorative laparoscopy showed a normal appendix, the right fallopian tube was distended and adhered to the right inflamed ovary and pelvic side wall, where the intestine associated with adherent omentum. After adhesiolysis, pus was observed draining out from the right fimbrial end. Also, there were dense adhesions between the rectosigmoid colon and the uterus. Appendix appeared normal. Swabs were obtained for culture. Abscess drainage and pelvic lavage were then performed. Intravenous ceftriaxone 2 grams (50 mg/kg) once daily and 500 mg metronidazole intravenously every 8 hours were given intra-operatively and continued in post-operative period for 3 days. Regarding the etiology of the tubo-ovarian abscess, the patient denied repeatedly and convincingly previous sexual intercourse when questioned privately.

Post-operatively, her general condition was stable. At the third-post operative day, fever reached 39°C, accompanied by severe lower abdominal pain. Her clinical condition was deteriorating steadily associated with a very tense abdomen. Computed Tomography (CT) scan of the thorax, abdomen and pelvis was performed showing extensive multifocal pelvic abscess (6.2 x 6.1 cm), mainly involving the right ovary (Figure 1, see arrow). In the meantime, the microbiological analysis of the initially obtained blood culture as well as the intra-abdominal swab culture showed no growth.

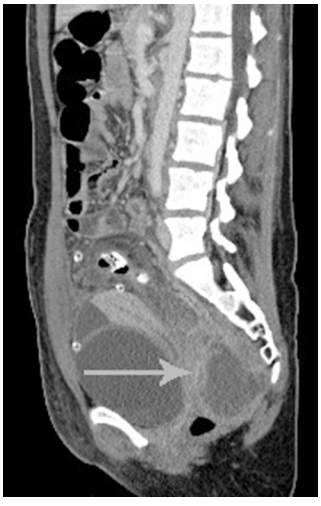

Figure 1. Computed tomography of the abdomen showing extensive multifocal pelvic abscess (6.2 x 6.1 cm) mainly involving the right ovary (Figure 1, see arrow).

A second emergency laparoscopy was decided due to recurrent pelvic abscess and peritonitis. Laparoscopic findings were; chronic right tubo-ovarian abscess with necrotic tissue, which was adherent to

the pelvic side wall with anatomical distortion associated with draining pus out from the right tube. The pelvic peritoneum was thick and inflamed. Left adnexa appeared normal. Pelvic swabs were obtained for aerobic and anaerobic culture. The decision of right adnexectomy was made to ensure complete eradication of the infection and necrotic tissue in order to prevent septicaemia and other complications despite she was nulliparous. Therefore, adhesiolysis, ureterolysis, and adnexectomy were carried out. Pelvic lavage, careful hemostasis was secured and a pelvic drain was left in. On the next day, the result of the culture of the first intra-abdominal swab showed growth of *F. necrophorum* with sensitivity to penicillin G and metronidazole. Therefore antibiotics were shifted to intravenous Penicillin G (1.5 million unit 4 times daily) and intravenous metronidazole (500 mg 3 times daily) for 14 days. We used a combination therapy because of the previously described clinical resistance to penicillin despite in vitro activity against *F. necrophorum* strains causing serious infections.[1] The second intra-abdominal swab showed no growth on culture.

After isolating *F. necrophorum*, the diagnosis of Lemierre's syndrome was made and further investigations were initiated in respects to potential complications and signs of the syndrome. A Doppler ultrasound of the neck was performed, with no signs of suspicious lesions, particularly signs of thrombophlebitis of the internal jugular veins. The patient was consulted by an otolaryngologist and an oral and maxillofacial surgery specialist, and no pathological findings were found.

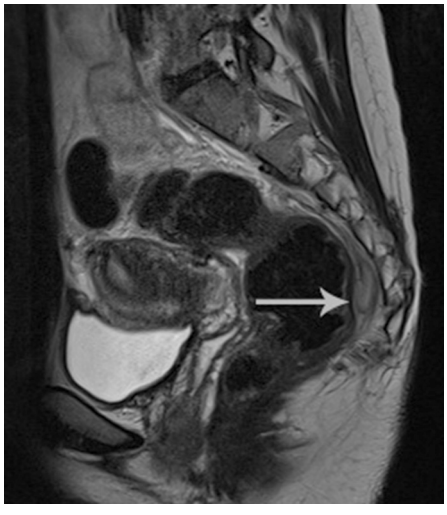

Figure 2. Magnetic resonance imaging of the abdomen showing normal findings after the surgical drainage of the abscess.

The patient showed progressive amelioration in her post-operative period and recovered steadily. Inflammatory markers dropped to normal levels, and a final abdominal ultrasound and magnetic resonance imaging (MRI) (Figure 2), showed no significant findings. We used MRI evaluation for the follow up of the treatment rather than CT as the latter is usually used for primary screening in abdominal pathologies, but in the second time, MRI was preferred for specific organ evaluation, . Nineteen days after admission, the patient was discharged fully recovered.

DISCUSSION

Usually previously healthy adolescents and young adults are reported with Lemierre's syndrome. The syndrome followed pharyngitis and tonsillitis as a complication. The pathogenesis might involve local suppurative thrombophlebitis, because of metastatic spreading of septic emboli to distant areas, commonly the lung. After the initial infection, thrombophlebitis of the IJV usually follows, presenting clinically as pain or swelling over anterior sternocleiodomastoid muscle, with probably absent local findings.

As mentioned earlier, there are different existing definitions of Lemierre's syndrome, which are summarized and discussed by Riordan.[1] However, a lot of reports and case presentations described the classical findings of Lemierre's syndrome with metastatic lesions and a preceding history of an infection of the pharynx in the absence of a thrombophlebitis of the IJV.[1,5,6] We therefore consider the definition of Lemierre's syndrome given by Riordan as compelling and reasonable, "an infection of the pharynx in the preceding four weeks, metastatic lesions in lungs and/or other organs, evidence of a thrombophlebitis of the IJV or isolation of *F. necrophorum* from blood cultures or a normally sterile site.[1] According to this definition, we were able to diagnose Lemierre's syndrome in our patient. The definition here states (OR), and we found the causative organism in the culture.

Alternative considerations in our case consisted of the inquiry, whether the infection with *F. necrophorum* arising outside the oropharynx can be called Lemierre's syndrome or not. There are in fact some descriptions in the literature about infections with *F. necrophorum* arising from the female genital tract, especially in significantly older patients or immunocompromised patients with other underlying diseases like malignancy.[1,7-9] Not least, Fuso-

bacterium is part of the normal human microflora of the genitourinary tract as well as of the oropharynx. However, in our case, a myriad of facts emphasize the high probability of Lemierre's syndrome with hematogenous spread of *F. necrophorum* resulting in a tubo-ovarian abscess. First, our patient gave a past medical history of sore throat and cervical lymphadenopathy prior to her admission implying pharyngitis. Despite the fact that throat and neck appeared normal at initial presentation, she nevertheless kept complaining of symptoms located to the upper respiratory tract, head and neck. As to the absence of pathological local findings in the Doppler ultrasonography of the neck, especially in regards to a thrombophlebitis of the IJVs, as mentioned above, these findings are not mandatory in Lemierre's syndrome. Besides, it is possible that in our case was subjected to subclinical phlebitis without thrombosis of the jugular vein or an affection of smaller pharyngeal veins undetectable by Doppler ultrasonography, which took place twelve days after admission and after already nine days of antibiotic therapy. This delay was due to the fact that the first intra-abdominal swab culture took quite long time to show the growth of *F. necrophorum*, which is a usual period, since cultures of Fusobacteria can take 5-8 days to grow.

Second, as mentioned earlier, our patient was a virgin who developed a tubo-ovarian abscess, which is a highly rare condition.[10] Above that, no case reports exist which show the involvement of *F. necrophorum* in the development of a tubo-ovarian abscess in a virgin female. Therefore in our case, a local infection with *F. necrophorum* is extremely improbable.

CONCLUSION

Before antibiotic age, Lemierre's syndrome was not uncommon and usually these cases had a fulminant course. *F. necrophorum* can be an etiologic organism of adnexal abscess in a rather healthy host. The syndrome is frequently unnoticed and actually "quite overlooked" when it appears today. Furthermore, when usual complications like a thrombophlebitis of the IJV or pulmonary manifestations are not apparent, the diagnosis of Lemierre's syndrome can be quite challenging. With proper therapy, in the majority of patients a cure is to be expected. The clinicians should be aware of Lemierre's syndrome for early diagnosis and treatment.

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