



A Rare Combination of Citelli's Abscess and Lemierre's Syndrome due to *Proteus Mirabilis*

Wanda Siyabonga Tshabalala*, Bradley Naidoo and Shaun-Ray Garth Temmers

Department of Internal Medicine, University of KwaZulu-Natal, South Africa

Abstract

We present a case report of an 18-year-old male with chronic Otitis media that complicated with Lemierre's Syndrome (LS) and a Citelli's abscess due to infection by *Proteus mirabilis*. Citelli's abscess is infrequently encountered in patients with suppurative otitis media; the presence of LS in combination is a rare occurrence. Prior to the wide distribution of antibiotic use LS was an often-fatal condition. Though its prevalence has decreased in recent years, there has been a resurgence due to antibiotic resistance. *Fusobacterium* species have long been reported as the most prevalent organism in this disease. The presents of *Proteus* species as source pathogen has not been identified to date. Following early surgical and medical intervention the patient had a favourable outcome in this case. Difficulties in theatre time and access to imaging may have contributed to delay in diagnosis; however this did not lead to worsening morbidity.

Keywords: Lemierre's syndrome; *Proteus mirabilis*; Chronic otitis media; Citelli's abscess

Abbreviations

COM: Chronic Otitis Media; CT: Computer Tomography; ENT: Ear, Nose, and Throat; HIV: Human Immune Virus; Hb: Hemoglobin; LS: Lemierre's Syndrome; PCR: Polymerase Chain Reaction; RNA: Ribonucleic Acid

Introduction

Chronic middle ear infections are the most important preventable cause of hearing loss [1]. The global burden of chronic otitis media is between 65 to 330 million [1].

The intracranial complications of otitis media include meningitis, brain abscess, otitic hydrocephalus and lateral sinus thrombosis [2]. Extracranial complications include mastoiditis, with or without cholesteatoma through contiguous spread. Other complications are encountered less commonly and the list includes labyrinth fistulae, facial nerve palsy, and subperiosteal abscesses of the mastoid [1,2].

Subperiosteal abscess pathways can occur by direct inflammatory spread from the overflowing mastoiditis into nearby spaces and muscles attached to the temporal bone [3]. Citelli's abscess as an extratemporal complication of otitis media and occurs when pus from the mastoid trickles along the posterior belly of the digastric muscle to the cervical and occipital region [4]. This is a rare occurrence and is not well document in literature [3,4].

Lemierre's Syndrome (LS) is a rare complication resulting in septic thrombophlebitis of the internal jugular vein that is associated with ear, nose and throat infection [5]. This syndrome was labeled a "forgotten disease" in the 1980's because of the widespread introduction of antibiotics [6-8]. This is a lethal syndrome with a 90% mortality rate when left untreated [9]. Successful treatment of LS depends on early detection and treatment [5-10]. The number of cases has increased over the years demonstrated by a systematic review of LS in 2009 that reviewed 114 cases globally with ages of occurrence ranging between two months to 78 years [6]. In 2016, Johannesen and Bodtger did a five-year review of cases with LS and found 137 cases [7]. This increase in number of cases of LS is due to a change in pattern of antibiotic prescription, as well as an increase in the incidence of antibiotic resistance [6,7]. A definite diagnosis of LS is made on bases of a recent pharyngeal illness; complicated by septic emboli; thrombosis of internal jugular or blood cultures of *Fusobacterium necrophorum* [7]. The thrombophlebitis of the internal jugular is associated with septic embolization's commonly to the lungs [6,7]. The less common sites of septic embolization's are the liver, muscle, pericardium, brain and skin [6,7]. *Fusobacterium* species are reported as the commonest offending

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*Correspondence:

Wanda Siyabonga Tshabalala,
Department of Internal Medicine,
University of KwaZulu-Natal, Grey's
Hospital, Pietermaritzburg, South Africa,
E-mail: doctortshabalala884@gmail.com

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Table 1: Results of tests done during admission of an 18-year-old male with Citelli's abscess and Lemierre's syndrome at a hospital in KwaZulu-Natal.

Investigations		Admission	Day 13
Urea & electrolytes		Normal	Normal
Liver function tests	Reference ranges		
Total protein	60-80 g/L	61	74
Albumin	35-52 g/dL	17	20
Bilirubin	5-21 µmol/L	126	12
ALT	10-40 U/L	45	21
AST	15-40 U/L	56	23
ALP	53-128 U/L	286	127
GGT	<68 U/L	370	89
LDH	100-190 U/L	266	142
Troponin		negative	
Pro-B-natriuretic peptide		negative	
SARS-COV-2		negative	
C-reactive protein		259 g/mL	8 g/mL
Procalcitonin		69.98 ug/L	0.31 µg/L
Lactate		0.6 mmol/L	
Ferritin		1784 ug/L	405 µg/L
White cell count		24.70 × 10 ⁹ /L	9.10 × 10 ⁹ /L
Differential count dominantly neutrophils		22.70 × 10 ⁹ /L	
Haemoglobin		12.2 g/dL	8.8 g/dL
Platelets		648 × 10 ⁹ /L	256 × 10 ⁹ /L
Blood cultures		Two cultures of <i>Proteus mirabilis</i> pre antibiotics <ul style="list-style-type: none"> • sensitive to sulfamethoxazole/trimethoprim, penicillin, cephalosporins and gentamicin • Resistant to tigecycline 	
Intraoperative cultures taken in peri-auricular and citelli's abscess region		Organism cultured: <i>Proteus mirabilis</i> <ul style="list-style-type: none"> • sensitive to sulfamethoxazole/trimethoprim, penicillin, cephalosporins and gentamicin • Resistant to tigecycline 	
Sputum microscopy culture and sensitivities		Negative (six specimens)	
GeneXpert sputum and fluid from abscess		Negative	
Sputum cytology		no evidence of malignancy	
Histopathology report of tissue from radical mastoidectomy		Free-lying keratinous debris admixed neutrophil and bacterial colonisation. The differential diagnosis includes inadequate representation of a cholesteatoma.	

family of organisms in 90% of the cases of LS [6,7]. *Fusobacterium necrophorum* it is part of normal flora of the oropharyngeal cavity [5-7]. Two cases of LS that have been reported in case reports in South Africa in the past 20 years [10,11].

Case Presentation

An 18-year-old male patient presented to a hospital in KwaZulu-Natal, South Africa; with a two-week history of a productive cough with yellow sputum with blood streaks, right sided pleuritic chest pain, fever, and rigors. He also reported a purulent left ear discharge with left anterior neck and posterior auricular pain. He has a medical history of multiple episodes of AOM that were treated with antibiotics at district level clinics in childhood. In 2012 he had Computed Topography (CT) that revealed chronic mastoiditis which was treated with prolonged antibiotics. He had recently tested negative for *Human Immunodeficiency Virus* (HIV) and received all his vaccinations. This was his first hospitalization for this problem.

His vital signs on the day of admission revealed a temperature of 40.3°C; a respiratory rate of 22 breaths per minute with oxygen

saturation of 96% while breathing ambient air; a radial pulse rate of 124 beats per minute; and a non-invasive blood pressure of 106/74 mmHg.

His physical examination revealed left posterior auricular swelling with small posterior auricular lymph nodes as well as a tender, swollen left carotid-angle. He was pale however his cardiovascular examination was normal and there were no features of infective endocarditis. His respiratory examination revealed features of consolidation involving his right midzone and right lower zone, furthermore there was reduced breathe sound in both lung basis. His abdominal and central nervous system examinations did not reveal any abnormalities. He was diagnosed as septic chronic otitis media complicated by mastoiditis.

Case Management

The results of investigations done during this admission are summarized in Table 1. Day one of admission, he was taken to theatre for radical mastoidectomy with incision and drainage. He received amoxicillin/clavulanic acid 1.2 g three times daily from



Figure 1: Lung cavity with a fluid level in the right lower zone of a patient with Lemierre's syndrome.

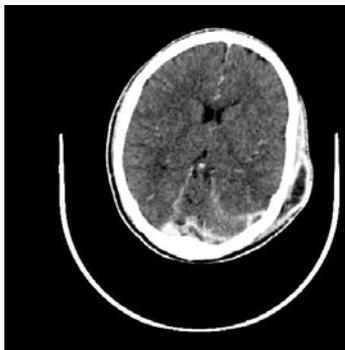


Figure 2: CT scan showing transverse sinus thrombosis and left-sided peri-auricular abscess collection.



Figure 3: Citelli's abscess in an 18-year-old with chronic mastoiditis: (green arrow heads).

day one to three. On day three, his blood cultures grew *P. mirabilis* (PM) with sensitivity to trimethoprim/sulfamethoxazole, penicillin, cephalosporin and gentamycin. The amoxicillin/clavulanic acid was changed to ceftriaxone 1 g three times daily as he had worsening hyperbilirubinemia and cholestasis. On the fifth of his admission, his clinical condition had not improved despite directed antibiotics and surgical drainage; he still had tenderness and swelling at the left carotid angle with fever, dyspnea, tachypnea and on-going production

of blood streaked sputum. Four additional blood cultures and six sputum cultures were negative at this point. He was now assessed as worsening septicemia and his antibiotics was changed to piperacillin/tazobactam and amikacin. Repeat imaging of his head, neck and chest was performed. The chest X-ray was repeated and it showed a cavity in right lower zone (Figure 1). The Computed Tomography (CT) scan found that there was reorganization of the abscess with transverse sinus thrombus (Figure 2). The CT scan of the neck and chest revealed rim enhancing collection in the soft tissues of the left peri-auricular region and deep neck tissues with an Internal Jugular (IJ) thrombus extending to the transverse sinus (Figure 3 and 4). The CT scan of the chest further revealed multiple lesions that were in different stages of breakdown with cavities (Figure 5). The transverse sinus thrombosis was an extension of internal jugular thrombus due to sepsis related thrombophlebitis and the distribution of his lung lesions suggest they were due to septic embolization from his primary site of infection. He was subsequently diagnosed with Lemierre's syndrome secondary to COM complicated with mastoiditis and Citelli's abscess.

On day six, he was referred to Ear, Nose and Throat (ENT) department for emergency incision and drainage. His surgery was delayed because of the South African lockdown due to coronavirus prevention strategy. Surgery was performed on day eight after his initial presentation; he underwent drainage of post-auricular and Citelli's abscess in theatre which evacuated fifteen milliliters (15 ml) and twenty milliliters (20 ml) of purulent debris. His clinical condition showed marked improvement post-surgery. The respiratory symptoms and productive cough immediately ceased day one post-surgery. On the eleventh day of his admission, the intraoperative



Figure 4: Left internal jugular vein thrombus: (green arrow heads).



Figure 5: Multiple bilateral lung lesions with cavitation in an 18-year-old immunocompetent male with Lemierre's syndrome.

specimen from two sites both cultured PM with similar sensitivity as his index blood culture from admission. The patient continued to improve and inflammatory markers normalized. The tazobactam/piperacillin 4.5 g three times daily was continued for 14 days and amikacin continued for 5 days after the drainage of abscess. During the course of admission, the patient had worsening of the mild anemia with Hemoglobin (Hb) of 12.2 g/dL that was seen at admission with a minimum drop to Hb of 8.1 g/dL. The anemia was a normocytic and normochromic anemia resulting from surgical blood loss, multiple blood sampling, and severe acute illness. The anemia gradually normalized after the septicemia had resolved. The lesions in his lungs improved with a six-week course of amoxicillin. He was counseled on anticoagulation and the decision was to continue anticoagulation for 6 months.

Discussion

Our patient had four positive cultures of *P. mirabilis* from different sites that were obtained at different times during his admission. *P. mirabilis* as an offending organism in LS is rare. Ninety percent of proteus infections are due to *P. mirabilis*, a gram-negative bacillus [12]. This organism is considered to be a community acquired organism that is able to survive on solid surfaces [12]. It is reported to be one of the common pathogens cultured in chronic middle ear infections and is also commonly cultured in patients with urinary tract infection in the setting of prolonged catheterization [1,6,13,12]. A systemic review from 2003 to 2019, found that *P. aeruginosa*, *S. aureus*, and *P. mirabilis* were there main reported pathogens associated with bacterial otitis media in the Sub-Saharan Africa [14]. *P. mirabilis* was found to be the culprit organism in 36% (18/50) adults with otitis media in Western Cape, South Africa [15]. Furthermore *P. mirabilis* was also the most frequently cultured organism in a system review of 1,307 cases with otogenic brain abscess in 2018 [13]. During the initial stages of management, this *Proteus* organism proved to be a challenge in the treatment of the patient due to the uncertainty of whether the organism had resulted in the chronic mastoiditis or if it was a contaminant. This proved to be a challenge during the initial stages of management of this case. Due to LS being relatively rare, it was difficult to ascribe the patient's index clinical state being due to *P. mirabilis* infection and thus the choice to direct antibiotics as per cultures became an area of contention. In spite of five days of directed antibiotic therapy, the patient showed no clinical or biochemical improvement.

Prolonged antibiotics are suggested to be the mainstay treatment of internal jugular vein thrombosis in patients with LS. There is no suggested regimen of antibiotics as there has never been randomized controlled trials to assist with clear guidelines to the management of LS. Surgical removal of the thrombus was previously explored in the pre-antibiotic era but not in the post antibiotic era [6,7].

The presence of chest lesions and infiltrative liver function test with hyperbilirubinemia should alert treating physicians to explore the upper respiratory tract for a possible source of infection as delay in diagnosis can have near fatal consequences.

Conclusion

Early diagnosis and initiation of antibiotics is key to the outcome of these patients. Knowledge about complications of middle ear

infections and Lemierre's syndrome is vital for practitioners managing patients with middle ear infections.

Permission

The patient gave consent for the case report to be published. The permission to publish was granted Grey's Hospital Chief executive officer and KwaZulu-Natal Provincial health research & knowledge management office.

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References

1. WHO. Chronic suppurative otitis Media Burden of Illness and Management Options. Geneva: World Health Organization, 2004.
2. Erasmus T. Chronic suppurative otitis media. Continuing Medical Education. 2012;30(9):335-6.
3. Brożek-Mądryz E, Waniewska-Lęczycza M, Robert B, Krzeski A. Head and neck abscesses in complicated acute otitis media-pathways and classification. Otolaryngol. 2018;8(2):345.
4. Sahoo AK, Preetam C, Samal DK, Sarkar S. Citelli's abscess following Otitis Media: A Case Report. Iran J Otorhinolaryngol. 2017;29(92):161-3.
5. Eilbert W, Singla N. Lemierre's syndrome. Int J Emerg Med. 2013;6(1):40.
6. Karkos PD, Asrani S, Karkos CD, Leong SC, Theochari EG, Alexopoulou TD, et al. Lemierre's syndrome: A systematic review. Laryngoscope. 2009;119(8):1552-9.
7. Johannesen KM, Bodtger U. Lemierre's syndrome: Current perspectives on diagnosis and management. Infect Drug Resist. 2016;9:221-7.
8. Coutlas JA, Bodasing N, Horrocks P, Cadwgan A. Lemierre's syndrome: Recognising a typical type of presentation of a rare condition. Case Rep Infect Dis. 2015.
9. Lemierre A. On certain septicaemias due to anaerobic organisms. Lancet. 1936;227(5874):701-3.
10. Such R, Joseph E. Lemierre's syndrome - the uncommon cold. SA J Radiol. 2005;9(2):22-4.
11. Roos M, Harris T, Seedat R. Fatal Lemierre's syndrome as a complication of chronic otitis media with cholesteatoma. South Afr J Child Health. 2016;10(4): 231-2.
12. Jamil RT, Foris LA, Snowden J. Proteus mirabilis infections. [Updated 2019 Sep 3]. In: Stat Pearls [Internet]. 2020.
13. Duarte MJ, Kozin ED, Barshak MB, Reinshagen K, Knoll RM, Abdullah KG, et al. Otogenic brain abscesses: A systematic review. Laryngoscope Investig Otolaryngol. 2018;3(3):198-208.
14. Tesfa T, Mitiku H, Sisay M, Weldegebreal F, Ataro Z, Motbaynor B. et al. Bacterial otitis media in sub-Saharan Africa: A systematic review and meta-analysis. BMC Infect Dis. 2020;20:225.
15. Meyer E, Whitelaw A, Edkins O, Fagan JJ. Chronic otorrhea: Spectrum of microorganisms and antibiotic sensitivity in a South African cohort. S Afr Med J. 2013;103(7):471-3.