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Case report

Community-acquired Klebsiella pneumoniae central nervous system infection after acute suppurative otitis

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ABSTRACT

Community-acquired Klebsiella pneumoniae (K. pneumoniae) central nervous system (CNS) infection combined with bacteremia is rarely identified worldwide. We received a 55-year-old woman on long-term corticosteroid therapy for Sjogren's syndrome. Onset began with acute suppurative otitis, followed by a severe headache and loss of consciousness. Cerebrospinal fluid (CSF) testing and brain imaging examinations were compatible with K. pneumoniae meningitis and likely brain abscesses, respectively. K. pneumoniae bacteremia was also found on blood cultures. Despite aggressive antibiotic and supportive therapy, the patient died after 2 day's therapy. Corticosteroid therapy may be a risk factor for a community-acquired K. pneumoniae infection. Appropriate antibiotics and abscess drainage are still recommended, despite the poor prognosis.

Background

Klebsiella pneumoniae (K. pneumoniae), a gram-negative, aerobic, rod-shaped bacterium, is usually hospital-acquired and occurs primarily in patients with impaired immune defenses [1]. Community-acquired infections caused by K. pneumoniae have mainly been reported in cases of invasive liver abscess syndrome (ILAS), meningitis, or endophthalmitis in Taiwan [2]. Although sporadic cases can occur elsewhere, community-acquired K. pneumoniae central nervous system (CNS) infections without liver abscesses are rarely seen [3–11]. We report our recent experience with an adult case of community-acquired K. pneumoniae CNS infection associated with acute suppurative otitis.

Case presentation

A 55-year-old woman from Hebei province in Mainland China with a two-year history of Sjogren's syndrome was taking oral methylprednisolone 24 mg/day. Eight days prior to arriving at our hospital, the patient noticed a purulent discharge from her right ear but did not seek any diagnosis and treatment. Over the next six days, she developed a fever and headache, then was found unconscious by her family on her way to our emergency department (ED). On examination in our ED, the patient's vital signs were normal, but she had altered mental status with a Glasgow coma scale of 5 (E1+V1+M3). On physical exam, she evidenced nuchal rigidity and had a positive right-sided Babinski sign.

Initial laboratory findings were as follows: white blood cell (WBC) count of 10.92×10^9/L with an elevated neutrophil ratio of 89.0 %, hemoglobin of 11.1 g/dL and a platelet count of 302×10^9/L. The C-reactive protein was >160 mg/L, liver and renal function tests were normal. Procalcitonin was 4.5 ng/mL. Lumbar puncture yielded pale yellow, cloudy cerebral spinal fluid (CSF) with an opening pressure of >330mmH_2O. CSF results were as follows: WBC count, 1.15×10^9/L, with a predominance of polymorphonuclear leukocytes; total protein, 2.48 g/L; and glucose, <0.11 mmol/L. Head and temporal bone CT scans revealed right mastoiditis, while an abdominal ultrasound and CT scan of the chest, abdomen and pelvis were normal. The patient was diagnosed as having purulent meningitis and right mastoiditis complicated by underlying Sjogren's syndrome and was treated with 2 g of cefatriaxone and 1 g of vancomycin every 12 h, as well as mannitol to help reduce intracranial pressure. The patient's level of consciousness did not recover, and she was transferred to the ED intensive care unit (EICU) that same day.

On hospital day two, K. pneumoniae grew from both her CSF and blood cultures and was found to be sensitive to all tested antibiotics. Antibiotics were adjusted to meropenem 2 g every 8 h

Abbreviation: K.pneumoniae, Klebsiella pneumoniae; CNS, central nervous system; CSF, cerebrospinal fluid; ILAS, invasive liver abscess syndrome; IRB, Institutional Review Board.

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and amikacin 0.4 g every 12 h, while the dose of methylprednisolone was gradually reduced to 10 mg daily to help control infection. However, the patient's condition worsened, and she was endotracheally intubated for airway protection.

Because of her continuous fever and altered mental status, a contrast-enhanced head CT scan and lumbar puncture were again performed on hospital day seven. The contrast-enhanced head CT demonstrated acute, multi-layered, low-density masses in the patient's bilateral frontal, parietal, occipital and right temporal lobes without abnormal enhancement (see Fig. 1: contrast-enhanced head CT). CSF pressure decreased to 220 mmH_2O, and her CSF WBC count dropped to 0.16×10^9/L, with a CSF protein of 3.55 g/L. We next planned a contrast-enhanced head MRI and consult neurosurgery for potential operative options. However, the patient's relatives refused further treatment due to her already poor prognosis. The patient was extubated, and an ambulance was arranged to take her back home. She died on her way home secondary to a lack of spontaneous breathing.

Discussion and conclusion

Cases of community-acquired K. pneumoniae meningitis are exceedingly rare. In mainland China, the only such patient reported so far had ILAS [12]. Reviewing the literature to date, this patient is the first one we have found with community-acquired K. pneumoniae CNS infection and bacteremia with no neurosurgical or ENT (ear, nose and throat) surgical history or implants or procedures. We now review the current literature about community-acquired K. pneumoniae meningitis and try to analyze the reasons of our patient's demise.

A total of eight patients with community-acquired K. pneumoniae meningitis not associated with liver pathology have been reported in the Caribbean, as well as in Italy, Singapore, Taiwan, the United Kingdom, and the United States [3,4,9,13–15]. However, not all of them had brain abscesses or bacteremia. Two of them had uncontrolled diabetes and another two had chronic alcoholic diseases. In our case, long-term corticosteroid therapy may explain

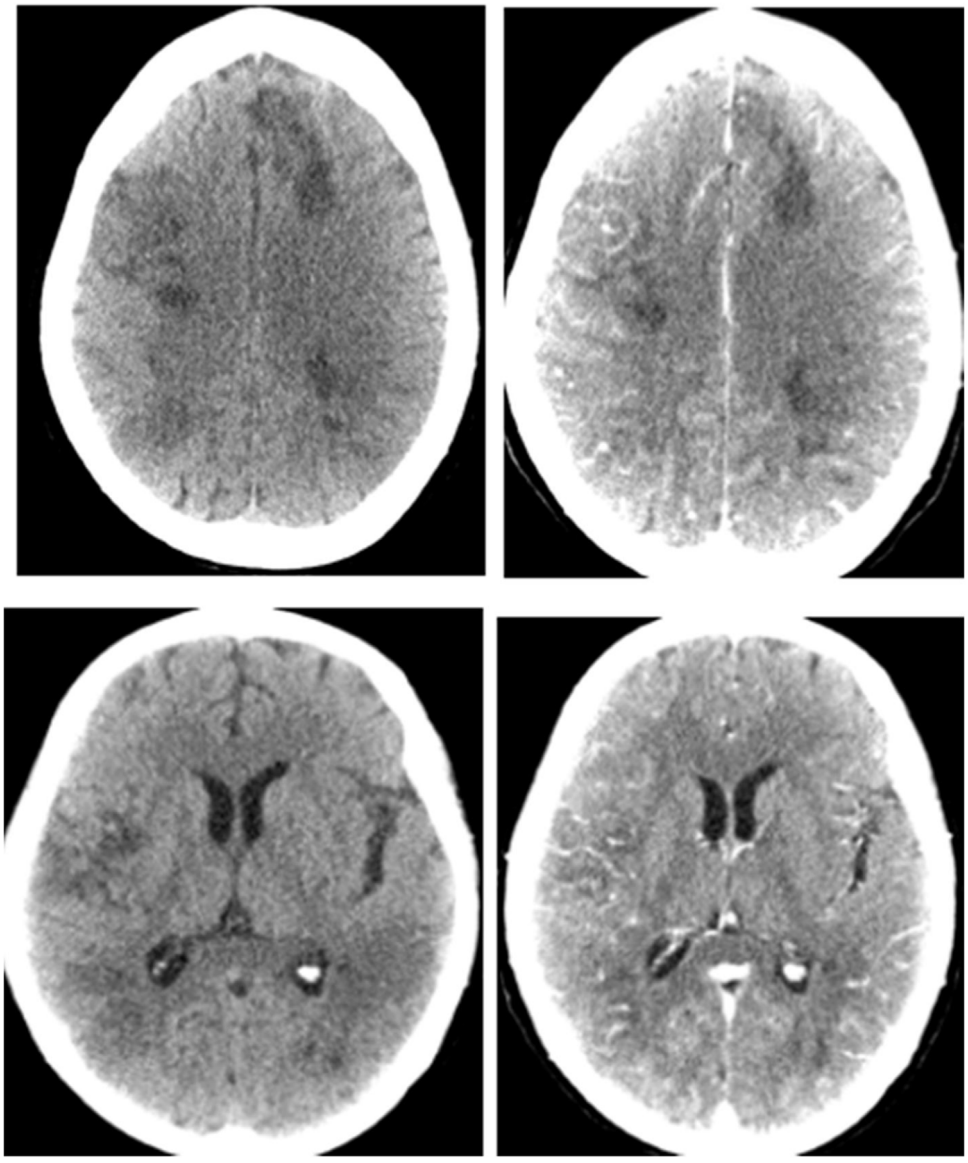

Fig. 1. Acute, multi-layered, low-density masses in the patient's bilateral frontal, parietal, occipital and right temporal lobes without abnormal enhancement.

the patient's increased risk for developing meningitis with K. pneumoniae.

The primary infection for our patient was likely suppurative otitis, which is similar to four other patients reported in the literature with preceding infections of endophthalmitis, otitis and sphenoid sinusitis [3,4,8,14]. However, K. pneumoniae was also cultured in our patient's blood, and the low-density CT scan showed likely multiple brain abscesses. This is the first patient shown to have K. pneumoniae bacteremia associated with meningitis, since the direct spread of organisms from a contiguous site (such as sinusitis) usually causes a solitary brain abscess [16]. The multiple brain abscesses in this case may be from the hematogenous spread of bacteria instead of direct spread from the suppurative otitis. Common conditions leading to hematogenous seeding of the brain usually involve chronic pulmonary infections, skin infections, pelvic and intraabdominal infections, bacterial endocarditis, esophageal dilation or the endoscopic sclerosis of esophageal varices [16,17]. Although the pathogenesis of this case is still unclear, the CNS infection combined with bacteremia could be a risk factor for the patient's poor prognosis.

Besides supportive treatment, the main therapy for patients with K. pneumoniae meningitis is timely antibiotics. The seven surviving patients with K. pneumoniae meningitis in the literature were treated with a third-generation cephalosporin combined with pefloxacin or gentamicin or amikacin, and only one case used meropenem [3,4,9,13–15] (sensitivity results were not provided in these cases, however). In our case, K. pneumoniae cultured from the CSF was susceptible to all of the remaining antibiotics tested, meropenem and amikacin included. Those two were chosen based on in vitro sensitivity, but, unfortunately, the ending was still tragic.

Drainage of the patient's brain abscesses was another point of consideration. Once an abscess has formed, surgical excision or drainage remains the treatment of choice [16]. Only one patient in the literature infected with K. pneumoniae has had a brain abscess, but this patient still died two weeks after lateral and ventricular drainage [12]. Other potential treatments of K. pneumoniae CNS infections, such as intrathecal antibiotics or brain abscess resection were not mentioned. Three patients with endophthalmitis, sphenoid sinusitis or liver abscesses, respectively, survived after eliminating the focus of infection [4,14], but these foci were outside the CNS. Because our patient had an already quite poor prognosis, her relatives chose to give up further efforts such as those mentioned above.

We also suspect that the strain of K. pneumoniae in this case may have had special virulence factors. K. pneumoniae strains related to ILAS harbors capsular serotypes K1 or K2, which are more virulent than those with non-K1/K2 serotypes. Besides capsular serotypes, the hypermucoviscosity phenotype, lipopolysaccharide, siderophores, and pili also contribute to the pathogenesis of K. pneumoniae [1,2]. Unfortunately, the molecule gene of K. pneumoniae in this case was not tested, but future studies may attempt to analyze the genetic code of K. pneumoniae for virulence factors leading to community-acquired meningitis without ILAS.

In conclusion, a community-acquired K. pneumoniae CNS infection without ILAS is a rare disease. Corticosteroid therapy was likely a risk factor for infection in our case. Appropriate antibiotic use and abscess drainage should be attempted for treating this condition. Our patient's death was likely a combination of K. pneumoniae bacteremia, presence of intracranial lesions and potential virulence factors for this strain of K. pneumoniae. This case is reported to help identify this rare disease and promote a further progress on understanding its pathogenesis and treatment.

Statement

The patient's next to kin has given their consent to publish the patients' cases for this study.

Ethics approval and consent to participate

The Institutional Review Board (IRB) of Peking Union Medical College Hospital has reviewed the study and has determined that this is a retrospective study and the design is scientifically and is up to the ethics standards. The IRB thus approve the study.

Consent for publication

Written consent for publication has been obtained from all the study participants and the patients reported in this article.

Availability of data and materials

The datasets used and analyzed during the current study are available from the corresponding author on reasonable request.

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Author contributions

All authors contributed to the study conception and design. Case collection and description were performed by Ruixue Sun and Jun Xu. The first draft of the manuscript was written by Ruixue Sun and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

Declaration of Competing Interest

The authors declare that they have no conflicts of interests.

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