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ABOUT COVER

Editorial Board Member of *World Journal of Clinical Cases*, Dr. Iva Brčić finished medical studies at the Medical University of Graz and received her MD degree in 2003. She received her doctoral degree in 2006 at the same institution. In 2007, she enrolled in the pathology residency program at the University Hospital Center Zagreb. In 2012, she passed her board exam and, until 2015, worked as a staff pathologist at the University Hospital Center Zagreb. From 2015, she is working as the University Assistant at the Medical University of Graz. At the end of 2017, she joined the bone and soft tissue team and spent 4-mo observership at the University of Miami, FL, USA. Her ongoing research interests include bone and soft tissue neoplasms.

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The primary aim of *World Journal of Clinical Cases* (*WJCC*, *World J Clin Cases*) is to provide scholars and readers from various fields of clinical medicine with a platform to publish high-quality clinical research articles and communicate their research findings online.

WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

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Dental focal infection-induced ventricular and spinal canal empyema: A case report

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Abstract

BACKGROUND

Dental focal infection-induced ventricular and spinal canal empyema is an extremely rare, severe, acute disease that is clinically associated with extremely high morbidity and mortality. Traditional cerebrospinal fluid (CSF) bacterial culture is time-consuming, with a low positive rate, which frequently results in severe irreversible consequences. The next-generation sequencing technique is an emerging pathogenic microorganism detection method that can obtain results in a short time with high accuracy, thus providing great assistance in the clinical diagnosis and treatment of this disease.

CASE SUMMARY

This paper reports a rare case of dental focal infection-induced ventricular and spinal canal empyema. During the course of treatment at a local hospital, the patient had negative results from repeated CSF bacterial cultures and was empirically given vancomycin treatment. After transfer to our hospital, the next-generation sequencing technique was adopted to determine that the pathogenic microorganisms were multiple anaerobic infectious bacteria derived from the oral cavity. The antibiotic therapeutic scheme was adjusted in a timely manner, and the ventricular and spinal canal inflammation was also controlled. However, the antibiotics that had been applied at the local hospital were not able to cover all pathogenic microorganisms, which resulted in irreversible injury to the brain stem, finally leading to patient death.

CONCLUSION

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Dental focal infection-induced ventricular and spinal canal empyema is an extremely rare, severe, acute disease with high morbidity and mortality. Any delay in diagnosis and treatment will result in irreversible consequences. The early application of the next-generation sequencing technique can obtain results in a short time and clarify a diagnosis. Appropriate antibiotic treatment combined with suitable surgical intervention is the key to managing this disease.

Key words: Ventricular empyema; Next-generation sequencing; Dental focal infection; Antibiotics; Surgery; Case report

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Core tip: Dental focal infection-induced ventricular and spinal canal empyema is an extremely rare severe acute disease with high morbidity and mortality. Any delay in diagnosis and treatment will result in irreversible consequences. The early application of the next-generation sequencing technique can obtain results in a short time and clarify a diagnosis. Appropriate antibiotic treatment combined with suitable surgical intervention is the key to managing this disease.

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INTRODUCTION

Dental focal infection-induced ventricular and spinal canal empyema is an extremely rare, severe, acute disease with high morbidity and mortality. Fewer than 5% of brain abscesses that develop from a dental focal infection manifest as pyocephalus, which is extremely rare^[1,2]. Analysis by computed tomography (CT) is the preferred imaging examination, while magnetic resonance imaging (MRI) is an examination modality with higher identification ability than CT, and it is the essential imaging analysis, if feasible based on the patient condition^[2]. Traditional laboratory bacterial culture is time-consuming, with a low positive rate^[2,3]. The next-generation sequencing technique is an emerging pathogenic microorganism detection method that can obtain results in a short time with high accuracy and provide great assistance in the clinical diagnosis and treatment of this disease^[4].

CASE PRESENTATION

Chief complaints

A 53-year-old female patient was admitted to the Neurotrauma Surgery Department of Jilin University First Hospital on September 13, 2019, due to intermittent fever for 6 d and unconsciousness for 4 d.

History of present illness

Initially, the patient was admitted to a local hospital as a result of hypertensive disease; 1 day after admission, she had fever, with the highest body temperature of 38.8 °C; 2 d later, the patient had sudden unconsciousness. Emergency head MRI revealed the fester-traditional cerebrospinal fluid (CSF) level inside the posterior horns of bilateral lateral ventricles, the fester diffuse weighted imaging (DWI) showed a high signal, and the apparent diffusion coefficient map showed a low signal (Figure 1). Thereafter, the patient was transferred to the Neurosurgery Department to receive emergency extraventricular drainage. Multiple CSF bacterial cultures after surgery revealed negative results, and the local physician empirically administered vancomycin treatment to the patient. After surgery, the patient was in continuous narcosis, and her family visited our hospital to seek further diagnosis and treatment.

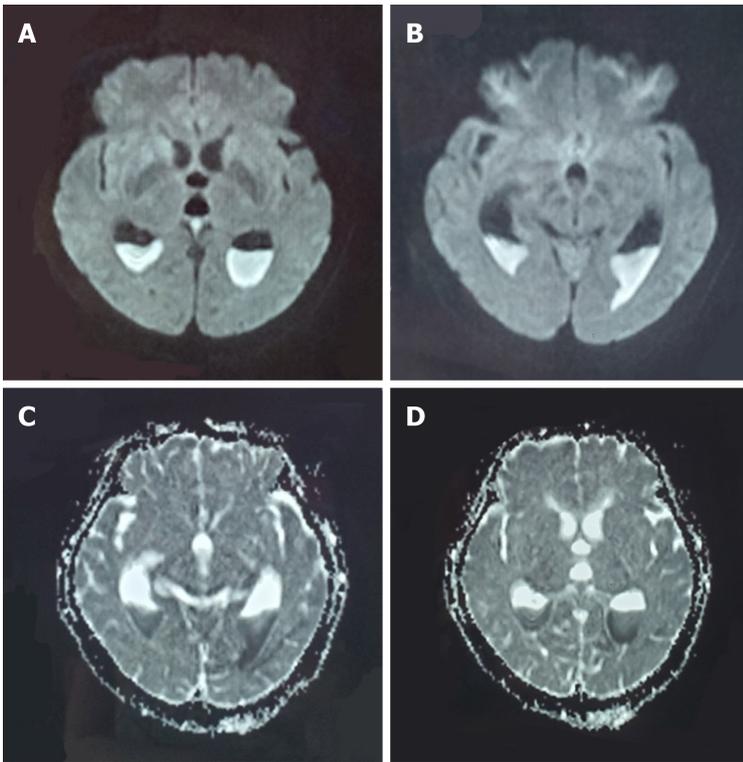


Figure 1 Head magnetic resonance imaging before admission. A and B: The fester-traditional cerebrospinal fluid level was seen inside the posterior horns of the bilateral lateral ventricles, and the fester diffuse weighted imaging showed a high signal; C and D: The fester showed a low signal on the apparent diffusion coefficient map.

History of past illness

The patient had a past history of hypertension for 10 years and received oral antihypertensive drug therapy, but no ideal control effect was achieved. In addition, the patient had had dental caries for 3 years, and she did not receive systemic treatment after the medulla oblongata treatment 1 month prior.

Personal and family history

The patient had a free personal and family history.

Physical examination upon admission

Physical examination upon admission showed that the patient was in narcosis with no autonomous respiration. The Glasgow Coma Scale score was E3V1M4. Extensive lesions and loosening of the second molars in the bilateral mandibles were found. CSF culture and next-generation sequencing of pathogenic microorganisms were carried out after admission. The intraventricular CSF culture results were negative, while pathogenic microorganism next-generation sequencing suggested *Fusobacterium*, *Porphyromonas*, *Solobacterium*, *Prevotella*, *Dialister*, *Fusobacterium nucleatum*, *Fusobacterium hwasookii*, *Porphyromonas endodontalis*, *Solobacterium moorei*, *Prevotella intermedia*, and *Dialister pneumosintes* (Figure 2A and B).

Imaging examinations

Head MRI revealed a quasicircular liquid signal opacity in the left cerebellum. The adjacent brain stem was compressed, the fourth ventricle became narrower, and ventricular hydrops accompanied by paraventricular edema was observed (Figure 3). Spinal MRI revealed stripe-like long T1 and long T2 signals in C7-T2 spinal canals (Figure 4).

FINAL DIAGNOSIS

The final diagnosis of the presented case was ventricular and spinal canal empyema.

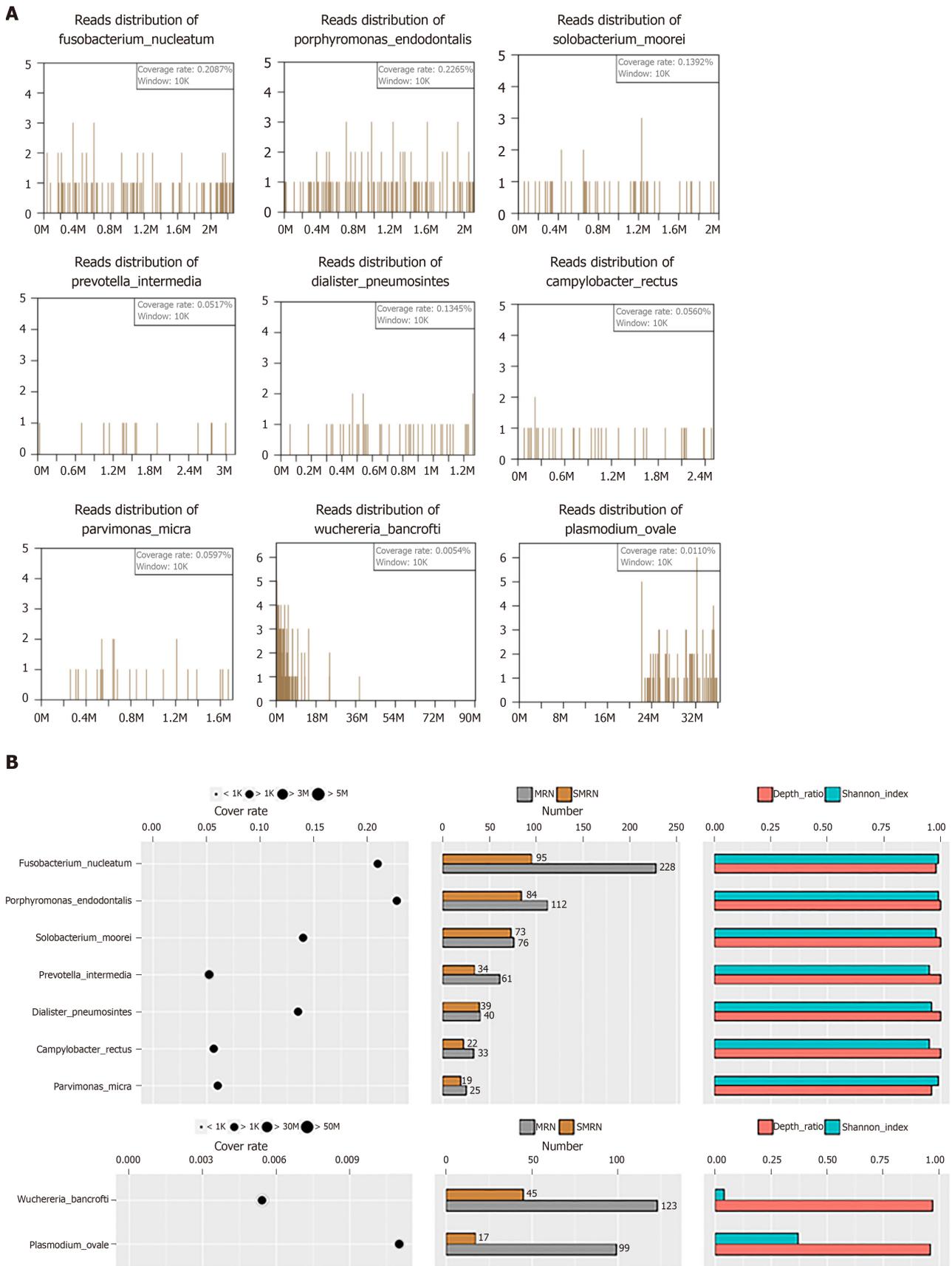


Figure 2 The next-generation sequencing results revealing the presence of the following pathogenic microorganisms in the traditional cerebrospinal fluid of the patient. A and B: *Fusobacterium*, *Porphyromonas*, *Solobacterium*, *Prevotella*, *Dialister*, *Fusobacterium nucleatum*, *Fusobacterium hwasookii*, *Porphyromonas endodontalis*, *Solobacterium moorei*, *Prevotella intermedia*, and *Dialister pneumosintes*.

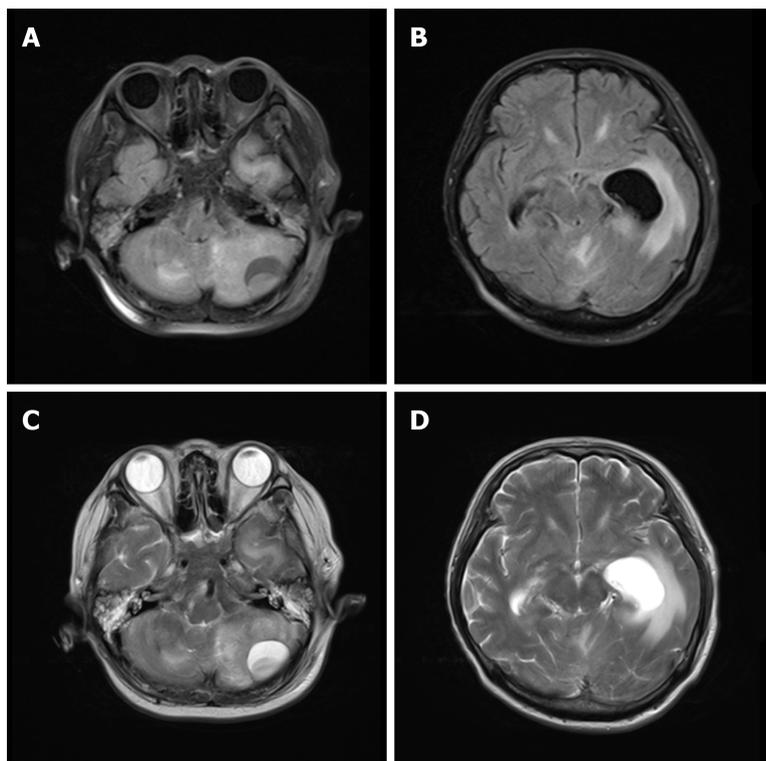


Figure 3 Head magnetic resonance imaging upon admission. A-D: A quasicircular liquid signal opacity was seen in the left cerebellum, with a low T1WI signal (A) and a high T2WI signal (C). The adjacent brain stem was compressed, the fourth ventricle became narrower (A and B), and ventricular hydrops accompanied by paraventricular edema were observed.

TREATMENT

After admission, the patient underwent several extracranial drainages and lumbar punctures. The antibiotic therapeutic scheme was adjusted immediately to vancomycin, meropenem, gentamicin, and S-ornidazole combined with anti-infective treatment.

OUTCOME AND FOLLOW-UP

The patient died of brain stem injury-induced respiratory circulatory failure at 2 mo after admission. The timeline of the case is shown in Figure 5.

DISCUSSION

Brain abscess is an extremely rare, life-threatening disease in neurosurgery, and its morbidity is reported to be 1/100000 in the United States^[1]. In the early 1970s, the mortality of brain abscess was 30%-60% but was reduced to 0-24% in recent years due to the rapid development of neuroimaging techniques (such as CT and MRI), the development of more effective antibiotics, and the improvement of surgical techniques^[5]. Among all causes of brain abscess, otogenic causes account for 65.75%, hematogenous causes account for 13%, and odontogenic causes account for a low fraction. In addition, only < 5% of dental focal infection-induced brain abscesses manifest as pyocephalus, which is extremely rare^[1,2].

As part of the Human Microbiome Project, the molecular identification method has been used to identify over 1200 kinds of microorganisms in the human oral cavity^[6]. Over 350 distinct microorganisms were isolated from marginal periodontitis^[6], and 150 different bacterial strains were isolated from endodontic infections^[7]. The commonly seen pathogenic microorganisms include *Streptococcus viridans* (especially the *anginosus* group), *Actinomyces*, *Peptostreptococcus*, *Prevotella*, *Fusobacterium*, *Aggregatibacter actinomycetemcomitans*, and *Eikenella corrodens*^[2]. The next-generation sequencing results suggested that the 11 infectious bacteria in our patient were mostly

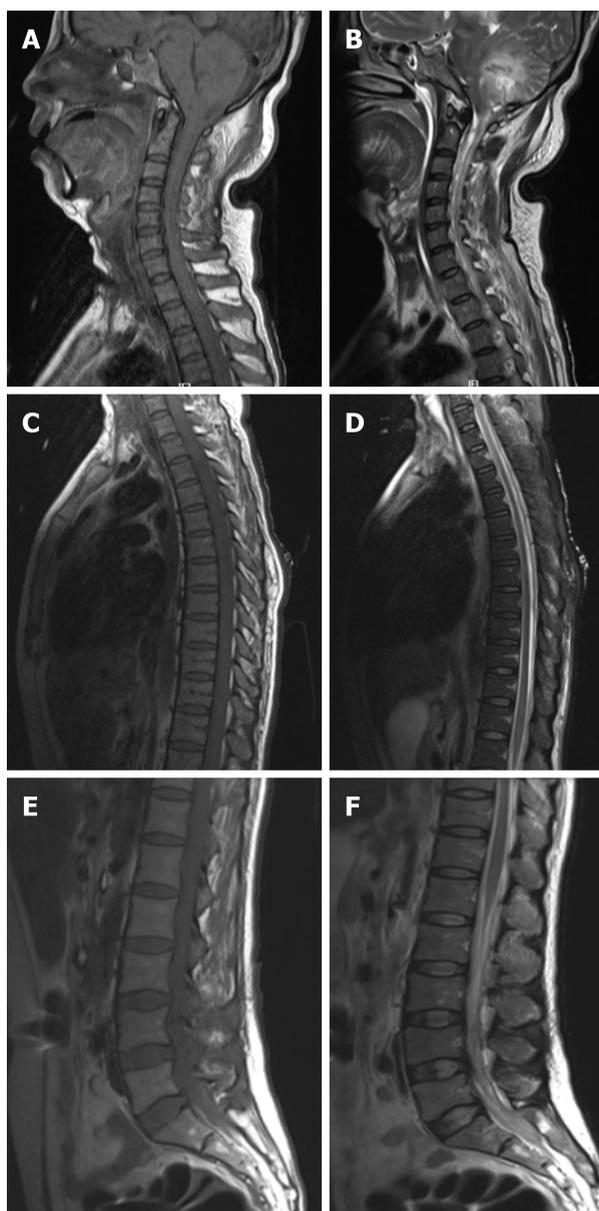


Figure 4 Spinal magnetic resonance imaging on admission. A and B: Flake-like and stripe-like long T1 and long T2 signals were seen in the cerebellar hemisphere and cervical spinal cords on spinal magnetic resonance imaging (MRI); C and D: Long stripe T1 and T2 abnormal signals were seen in the T1-2 spinal cord on chest MRI; E and F: A flake-like low signal was seen in the spinal cord on lumbar MRI, and multiple nodular low signals were observed in the cauda equine innervation area at the L3-4 levels.

colonizers of the oral cavity.

Research shows that the pathogenic microorganisms result in dental focal infection-induced brain abscess through the following four pathways: (1) Systemic bacteremia; (2) Direct drainage to the cavernous sinus through facial and pterygoid vein systems; (3) Infection through continuous diffusion; and (4) Lymphatic drainage^[8]. The most likely infection pathway of our patient was systemic hematogenous infection. The patient had a past dental operation history, and she did not receive systemic treatment. Dental operations cause periodontal vascular endothelial damage, and as a result, a large number of oral bacteria enter the blood. According to the research by Roberts *et al*^[9], periodontal small blood vessel injury results in bacteremia, and 38.5% of people have transient bacteremia even in daily tooth brushing. Our patient had a history of hypertension, and her blood brain barrier (BBB) integrity was destroyed. The BBB is an important defense barrier in the human body that provides physical protection for the central nervous system (CNS). However, in hypertensive patients, the BBB is broken, which may result in leakage of small arterioles, thus promoting the occurrence of bacteremia^[10].

CT examination is the preferred imaging modality, and a plain MRI scan combined with DWI should be performed subsequently^[2]. MRI-DWI can distinguish benign

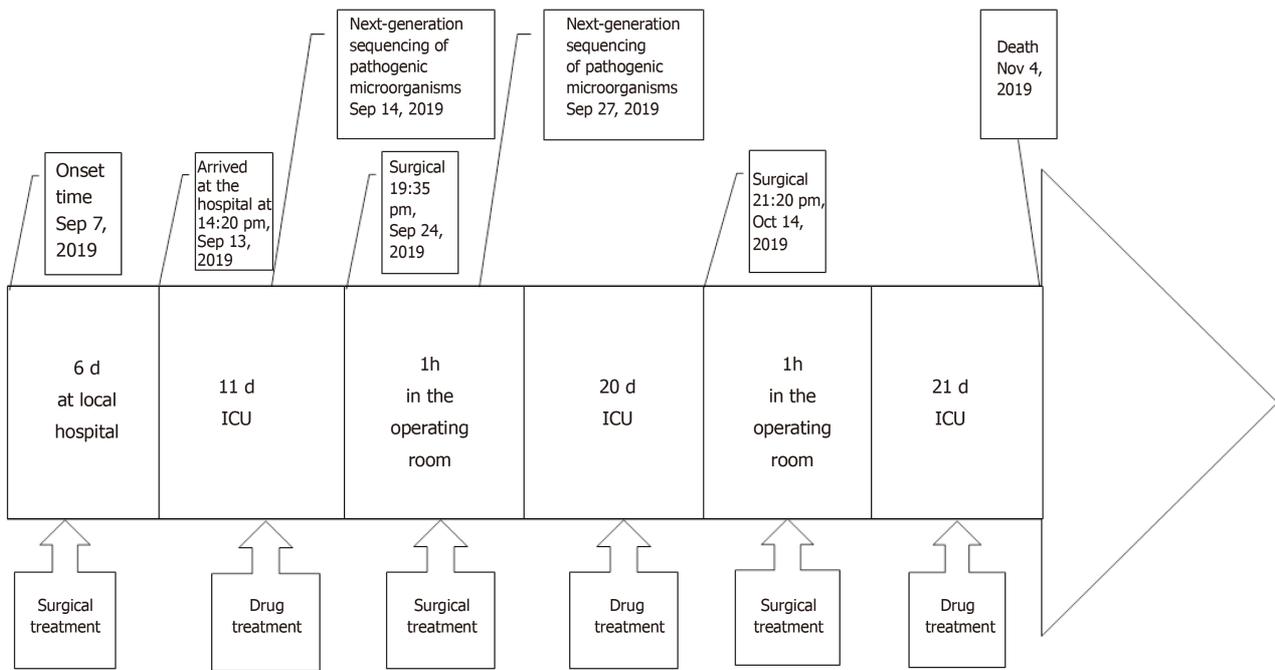


Figure 5 Timeline of the case. ICU: Intensive care unit.

abscesses from primary or metastatic tumors^[2]. Its typical MRI manifestation includes the liquid level formed by fester, necrotic debris, and CSF in the cerebral ventricle. Fester and debris are located at the weight-bearing sites in the occipital horn and trigonum of the cerebral ventricle; compared with the CSF signal, fester tissue debris shows a slightly low T2WI signal and a slightly high T1WI signal^[11-13].

Additionally, CSF bacterial culture is an essential laboratory examination. However, under certain circumstances, standard bacterial culture may not serve as an effective pathogen identification method, particularly when the source of infection is suspected to be derived from the oral cavity. This is because numerous oral bacteria are fastidious pathogenic bacteria, which may not necessarily grow in culture sufficiently to allow easy identification. Some bacteria induce infection only in the presence of other pathogenic bacteria, and they are called concomitant pathogens^[2]. In addition, traditional bacterial culture is time-consuming. Therefore, we adopted a new pathogen detection method, namely, next-generation sequencing. Its principle is to employ the second-generation high-throughput sequencing technique to sequence the nucleic acid in the sample, carry out a comparative analysis through the microorganism database according to the metagenomics principle, and obtain the species information of the suspected pathogenic microorganism by means of an intelligent algorithm^[4]. Using this microorganism detection method, results are obtained in 1 d, and it can achieve high accuracy^[4]. After the patient was admitted, we conducted traditional bacterial culture and pathogenic microorganism next-generation sequencing at the same time. The results of multiple bacterial cultures were negative, while pathogenic microorganism next-generation sequencing identified multiple oral microorganism infections. The antibiotic usage scheme was adjusted immediately based on the pathogenic microorganism next-generation sequencing results, and the CSF white blood cell count in the patient decreased from 551723×10^6 mmol/L to 33×10^6 mmol/L. The appearance of the CSF also changed from the original resemblance to rice water to that of a faint-yellow, clear liquid (Figure 6).

In terms of treatment, enough importance should first be attached to the progression of CNS disease, which should be regarded as a life-threatening, severe, acute disease^[14]. Once a diagnosis is confirmed, treatments from three aspects should be applied immediately: (1) Aggressive surgery for abscess debridement or drainage; (2) Suitable combined antibiotic treatment selected according to the pathogenic microorganisms; and (3) Treatment specific to the primary infection lesion^[14]. In our patient, the antibiotics combined with anti-infective treatment were adjusted in a timely manner, and local oral treatment was also applied, but the antibiotics that had been applied at the local hospital did not cover all pathogenic bacteria, and the brain stem function was subjected to irreversible damage, finally leading to patient death.



Figure 6 Traditional cerebrospinal fluid. A: Traditional cerebrospinal fluid (CSF) on admission. The CSF was a rice water-like purulent liquid; B: CSF at 2 wk after admission. The CSF was a faint-yellow, clear liquid.

CONCLUSION

Dental focal infection-induced ventricular and spinal canal empyema is an extremely rare, severe, acute disease with high morbidity and mortality. Any delay in diagnosis and treatment will result in irreversible consequences. The early application of the next-generation sequencing technique can obtain results in a short time and clarify a diagnosis. Appropriate antibiotic treatment combined with suitable surgical intervention is the key to managing this disease.

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