Subdural Empyema Due to Lactococcus lactis cremoris: Case Report

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Abstract

Lactococcus lactis cremoris (L. lactis cremoris) infections are very rare in humans. Only three case reports of brain abscess have been reported and the infectious routes and pathological features are still unknown. We experienced a subdural empyema due to L. lactis cremoris in an immunocompetent adult. A 33-year-old man was admitted with fever, right facial pain, left hemiparesis, and left hemianopsia. Computed tomography demonstrated low density fluid collection in the right falcotentorial subdural space. Magnetic resonance (MR) images revealed a high signal lesion on a diffusion-weighted image (DWI) and fluid attenuated inversion recovery (FLAIR) images in the right paratentorial and parafalcine subdural space, right maxillary sinus, and bilateral ethmoidal sinus. He underwent two sequential open surgeries for removal and drainage of empyema and was treated with antibiotics including meropenem and ampicillin. To our knowledge, this is the first report of subdural empyema caused by L. lactis cremoris infection. We report the case and discuss the pathological features with the previous literature.

Key words: subdural empyema, abscess, Lactococcus lactis cremoris

Introduction

Lactococcus lactis cremoris (L. lactis cremoris) is a facultative anaerobic Gram-positive coccus. This organism was previously classified as streptococcus but was transferred to the genus Lactococcus in 1985. It is a commensal organism of mucocutaneous surfaces of cattle and is occasionally isolated from human mucocutaneous surfaces. It is an important organism in the process of making dairy products including fermented milk, sour cream, and cheese. Although L. lactis cremoris is commonly considered to be non-pathogenic for immunocompetent adults, human infections have been noted recently.1–18) Infection in the central nervous system is very rare and only 3 cases of brain abscess have been reported.1,6,16) To our knowledge, this is the first report of L. lactis cremoris infection presenting with subdural empyema.

Case Report

A 33-year-old man presented with fever, right facial pain, gait disturbance, and left visual field defect. He was diagnosed with trigeminal neuralgia and given carbamazepine and non-steroid anti-inflammatory drugs; however, his headache gradually worsened and he was referred to our hospital 10 days after onset. He had multiple dental caries and sinusitis, which were both untreated. On neurological examination he was alert (Glasgow Coma Scale [GCS] = 15), but had nuchal rigidity, left hemiparesis, and left hemianopsia. Body temperature was 37.8°C. White blood cell count was 16,100/mm³ and serum C-reactive protein (CRP), was 26.3 mg/dl on admission. Computed tomography (CT) demonstrated iso-intensity fluid collection in the right falcotentorial subdural space. Magnetic resonance (MR) images revealed a high signal lesion on a diffusion-weighted image (DWI) and fluid attenuated inversion recovery (FLAIR) images in the right paratentorial and parafalcine subdural space, right maxillary sinus, and bilateral ethmoidal sinus. He underwent two sequential open surgeries for removal and drainage of empyema and was treated with antibiotics including meropenem and ampicillin. To our knowledge, this is the first report of subdural empyema caused by L. lactis cremoris infection. We report the case and discuss the pathological features with the previous literature.
no obvious erosion in the bone flap, but it was removed because the infection might have extended to the bone. The wound was closed with subdural drainage. After the procedure, empirical antibiotic therapy was started with meropenem 6 g/day. A gamma globulin agent, glyceol, and phenytoin were also administered. Left hemiparesis improved after surgery, but left hemianopsia, headache, and nuchal rigidity remained.

Culture of the drained pus and venous blood yielded \textit{L. lactis cremoris}, which was sensitive to ampicillin. For species identification, biological tests based on the Rapid ID 32 strep Api kit (SYSMEX bioMérieux Co., Ltd., Tokyo) were used. We de-escalated the antibiotics by changing meropenem to ampicillin 8 g/day.

Follow-up magnetic resonance imaging (MRI) 6 days after surgery demonstrated an increase of empyema in the fronto-parietal area (Fig. 3), and the patient complained of aggravated headache. We therefore decided to perform a second surgery for removal and drainage. Right fronto-parietal craniotomy was performed with the patient in the supine position (Fig. 4). Remaining subdural empyema along the falx was evacuated and irrigated as much as possible, and a bone flap was removed for the same reason as in the initial surgery. Most of the subdural empyema along the falx was removed (Fig. 5).

Dental examination showed multiple dental caries. Tooth 2 (Wisdom tooth) was considered to have caused maxillary sinusitis and was extracted. Otolaryngological

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Fig. 3  MRI after initial surgery. A, B: MRI the day after surgery (A: upper: FLAIR, lower: DWI, B: Gd-T,WI). Subdural empyema along the occipital falx and cerebellar tentorium was well removed, but empyema along the frontal falx remained. C, D: MRI on the 6th day postoperatively (C: upper: FLAIR, lower: DWI, D: Gd-T,WI). Empyema in the frontal to parietal parafalcine area had increased. DWI: diffusion-weighted image, FLAIR: fluid attenuated inversion recovery, Gd-T,WI: gadolinium-enhanced T₁-weighted image, MRI: magnetic resonance imaging.

Fig. 4  Operative findings of the second surgery. A: With right fronto-parietal craniotomy, pus (arrow) was evacuated from the subdural space along the falx. B: After placing subdural drainage, the wound was closed with the bone flap removed.

examination showed purulent nasal discharge filling the right nasal cavity and bilateral nasal polyps. The patient was diagnosed with deteriorated chronic sinusitis extending from dental caries and was treated conservatively with clarithromycin.

Chest X-ray showed normal findings. CT from the
neck to pelvis demonstrated no abdominal abscess or other abnormal findings except for asymptomatic liver hemangiomas. Echocardiography showed no vegetation and no gurgitation. Cerebral angiography showed no aneurysms. Serological screening for hepatitis B virus, hepatitis C virus, syphilis, and human immunodeficiency virus (HIV) was negative.

A week after the second surgery, CT demonstrated a low density area in the right temporal and lobe, which showed high intensity on MRI FLAIR images and iso intensity on DWI (Fig. 5). Secondary encephalitis due to subdural empyema was suspected, but the patient showed no worsening of symptoms or new symptoms. Antibiotic therapy was continued and the temporal lesion gradually

Fig. 5  Magnetic resonance imaging after second surgery (A: FLAIR, B: DWI). Subdural empyema along the falx was removed, but it remained in the right frontal and tentorial space. High intensity on FLAIR emerged in the right temporal lobe. DWI: diffusion-weighted image, FLAIR: fluid attenuated inversion recovery.

Fig. 6  Magnetic resonance imaging 2 months after discharge (A: FLAIR, B: DWI). High intensity areas on FLAIR and DWI had disappeared. DWI: diffusion-weighted image, FLAIR: fluid attenuated inversion recovery.
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7 cases of infective endocarditis and 3 cases of liver abscess (Table 1). As for intracranial infection, only 3 cases of brain abscess have been reported. The reason why L. lactis cremoris could be pathogenic to humans is still unknown. In some previous reports, its infection was associated with a history of exposure to unpasteurized dairy products or immunodeficiency. Among the 19 cases we reviewed, exposure to unpasteurized dairy products was seen in 8 cases, and immunodeficiency was seen in 4 cases.

Discussion

L. lactis cremoris is an important organism in the process of making dairy products such as cheese and other fermented milk products. It is a commensal organism of mucocutaneous surfaces of cattle, and is occasionally isolated from human mucocutaneous surfaces. It is considered nonpathogenic in humans; however, some human infections have been reported recently and its pathogenic potential is becoming apparent. To our knowledge, 19 cases have been reported in the literature, including Table 1 Summary of the previous reports of Lactococcus lactis cremoris infection

| Author, Year | Infection site | Age | Sex | Consumption of unpasteurized dairy products | Dental history | Immune status |
|--------------|----------------|-----|-----|---------------------------------------------|----------------|--------------|
| Inoue et al. (Presented case) 2012 | Subdural empyema | 33 | M | None | Untreated dental caries | Normal |
| Feierabend et al. (2012)6) | Brain abscess | 8 | M | None | None | Normal |
| Topcu et al. (2011)16) | Brain abscess | 1 | F | Unpasteurized milk | None | Normal |
| Kim et al. (2010)8) | Liver abscess | 42 | M | None | None | Normal |
| Lin et al. (2010)11) | Endocarditis | 41 | M | None | None | Normal |
| Davies et al. (2009)4) | Ascending cholangitis | 72 | F | None | None | Normal |
| Resch et al. (2008) | Endocarditis | 49 | M | Unpasteurized cheese | Possible dental caries | Normal |
| Leung et al. (2006)4) | Canaliculitis | 80 | F | None | Dental caries under treatment | DM |
| Zechini et al. (2006)19) | Endocarditis | 55 | M | None | Dental surgery | None |
| Koyuncu et al. (2005)17) | Deep neck infection | 68 | M | Unpasteurized milk | Surgery and radiotherapy of buccal mucosa tumor | Previous malignancy |
| Antolín et al. (2004)2) | Liver Abscess | 79 | F | None | None | Normal |
| Halldórsdóttir et al. (2002)1) | Endocarditis | 67 | M | Unpasteurized milk | None | Normal |
| Akhaddar et al. (2002)1) | Cerebellar abscess | 45 | F | None | Dental surgery | Normal |
| Nakari et al. (2000)11) | Liver abscess | 14 | F | None | None | Normal |
| Pellizzer et al. (1996)11) | Endocarditis | 56 | M | None | None | Normal |
| Durand et al. (1995)11) | Septicaemia | 69 | M | Yoghurt | None | CLL |
| Campbell et al. (1993)11) | Septic arthritis | 57 | F | Unpasteurized milk | None | Normal |
| Mannion and Rothburn (1990)11) | Endocarditis | 65 | F | None | None | Normal |
| Torre (1999)11) | Necrotizing pneumonitis | 24 | M | Unpasteurized milk and cheese | None | HIV |
| Wood et al. (1955)11) | Endocarditis | 21 | M | Sour cream | Irritated gum surrounding a non-vital tooth | Normal |

CLL: chronic lymphocytic leukemia, DM: diabetes mellitus, F: female, HIV: human immunodeficiency virus, M: male.

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To our knowledge, this is the first report of subdural empyema due to *L. lactis cremoris*. Our patient had no history of unpasteurized milk exposure or immunodeficiency, but he had multiple dental caries and sinusitis, which were both untreated. *L. lactis cremoris* was identified from subdural empyema and blood culture on admission. Cultures from the oral cavity and nasal mucosa were performed after starting antibiotic therapy and *L. lactis cremoris* was not identified from these cultures. Two infection routes were considered for our case: hematogenous infectious extension from the oral cavity and direct extension from the oral cavity to the skull base via the paranasal sinus. Although the former route was suspected from bacteremia, the latter cavity to the skull base via the paranasal sinus. Although the former route was suspected from bacteremia, the latter might have caused sinusitis due to localized adjacent to the infectious lesions. Dental caries was more likely, because the subdural empyema was the former route was suspected from bacteremia, the latter route was considered for our case: hematogenous infectious extension starting antibiotic therapy and the oral cavity and nasal mucosa were performed after untreated dental caries, and chronic sinusitis, *L. lactis cremoris* in the oral mucosa might have extended to the subdural space. Emergent initial surgery, frequent imaging follow-up, repeated surgery for enlarged empyema, and appropriate use of sensitive antibiotics could successfully treat this rare and severe subdural empyema.

**Conclusion**

We experienced a rare case of subdural empyema due to *L. lactis cremoris*. In this case, with untreated dental caries and chronic sinusitis, *L. lactis cremoris* in the oral mucosa might have extended to the subdural space. Emergent initial surgery, frequent imaging follow-up, repeated surgery for enlarged empyema, and appropriate use of sensitive antibiotics could lead to a good outcome.

**Conflicts of Interest Disclosure**

The authors report no conflict of interest concerning this article.

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