Rare disease

Osteomyelitis of zygoma in a schizophrenia patient

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Summary

A 43-year-old man with schizophrenia presented to our hospital with appetite loss and general fatigue lasting 1–2 months. His face was flared and swollen, and he shed tears of pus. He could answer any questions, but never complained of pain. We found dacryocystitis with subcutaneous abscess with contiguous osteomyelitis with culture-proven *Staphylococcus aureus*, and diabetes mellitus (DM). Although DM neuropathy was mild, he did not complain of pain. We searched thoroughly for other abscesses since *S aureus* grew in all four of his blood cultures. We re-examined his whole body by CT, which revealed multiple muscle abscesses in both legs. It is reviewed that schizophrenia patients are relatively insensitive to physical pain. Thus, we should keep in mind that they may have multiple, unpredictable and rare underlying diseases, such as our case. Careful and thorough examinations are essential for treatment in schizophrenia patients.

BACKGROUND

Schizophrenia is a mental disorder with blunted affect, emotional withdrawal and social withdrawal, as well as auditory hallucinations, paranoia, delusions and conceptual disorganisation. It has been well known that the mortality in patients with schizophrenia is significantly higher than in the general population. Death by natural causes accounts for about two-thirds of the excess mortality, and suicide accounts for only a small proportion of the excess mortality.¹ The mechanisms of higher mortality with schizophrenia patients are unclear. However, their relative insensitivity to physical pain, as reviewed in some papers,^{2–3} could be an explanation for some of this causes. A case report of a schizophrenia patient with perforated small bowel had a lack of pain, which led to a delay in diagnosis and surgical intervention.⁴ Such cases suggest that schizophrenia patient without any symptoms may have unpredictable and rare underlying diseases.

In this paper, we will present a case of schizophrenia lacking symptoms, but had complicated *Staphylococcus aureus* osteomyelitis of the zygoma, associated with bacteraemia and multiple asymptomatic secondary muscle abscesses of both legs.

CASE PRESENTATION

A 43-year-old man was admitted to our emergency department due to appetite loss and general fatigue lasting 1–2 months. He suffered from schizophrenia, making him socially withdrawn. He stayed indoors for the last 10 years and did not receive any medication. His mother called the ambulance due to his severe weight loss, and visited our hospital. On admission, he was conscious, could answer any questions, but never complained of any symptom. We did not obtain any information about previous infections and abscesses in his history. His blood pressure was 116/74 mm Hg, respiratory rate was 20 breaths per minute and body temperature was 38.3 °C.

INVESTIGATIONS

On examination, his face was flared and swollen with an ulcer on his right nasolacrimal duct which was discharging pus (figure 1), but he did not complain of any pain. There were no lesions in the teeth, nose, sinus, throat or neck.

Laboratory data showed elevated WBC (white cell count) of 16 600/μl with 80.5% neutrophils, and increased serum C-reactive protein (CRP) level of 35.1 mg/dl U/l. Fasting blood sugar on admission was 306 mg/dl, and glycosylated haemoglobin was 14.6%. Other laboratory data suggested the absence of diabetic ketoacidosis.

He never complained of any pain, although his Achilles tendon reflex, vibration sensation, pin prick sensation and temperature sensation were almost normal.

CT scan with contrast medium showed destruction of the right zygoma with lesion expanding the subcutaneous abscess (figure 2).

Figure 1 Pretreatment picture shows ulcer above right nasolacrimal duct.

Figure 2 Axial image shows destruction of the right zygoma (a) with subcutaneous abscess (b).

We diagnosed the dacryocystitis with subcutaneous abscess with contiguous osteomyelitis. Gram stain of pus tear with tissue swab and subcutaneous abscesses with

drainage showed Gram-positive clusters. We searched thoroughly for more abscesses since all four of his blood cultures were positive.

We ruled out endocarditis by transthoracic echocardiography, and re-examined his whole body by CT to find other abscesses. We found multiple muscle abscesses in both legs (figure 3). Gram stain showed Gram-positive coccus (GPC) from all of the muscle abscesses drainage. *Methicillin-sensitive S aureus* (MSSA) was cultured from all specimens: tears of pus, subcutaneous abscess, blood culture and muscle abscesses.

We finally diagnosed the dacryocystitis with subcutaneous abscess with contiguous osteomyelitis, infection of bloodstream and muscle abscesses of both legs with culture-proven *S aureus*, which possibly exacerbated by diabetes mellitus (DM).

TREATMENT

We empirically started intravenous drip of vancomycin 1.0 g twice daily as antibiotic treatment in the case of GPC clusters from Gram stain were *Methicillin-resistant S aureus*, and placed drain tubes in the abscess of the face and both legs. MSSA only was cultured from subcutaneous abscess, muscle abscess of bilateral legs, tear of pus and blood. Both nafcillin and oxacillin are not sold in Japan, making cefazoline the first choice of *S aureus* treatment in Japan. Furthermore, since we feared his poor adherence, we continued cefazoline intravenously 1.0 g every 8 h for 42 days.⁵ We also administered insulin for DM treatment.

OUTCOME AND FOLLOW-UP

Fever, ulcer and inflammatory data such as CRP and erythrocyte sedimentation rate (ESR) recovered rapidly after 2 weeks of antibiotic treatment. CT of the face on

Figure 3 Axial image shows deep-seated abscesses of the legs.

Figure 4 CT scan on day 42 shows improving of osteomyelitis of zygoma.

day 42 demonstrated significant improvement (figure 4). He was discharged from our hospital with continuation of oral clindamycin treatment, which has excellent tissue penetration, particularly into bones and abscesses. He was healthy and well at 6-month follow-up, with antibiotics treatment continuation due to slight ESR elevation and CT abnormality.

DISCUSSION

Schizophrenia represents a mental disorder that has the prevalence of 0.5–1%, and is a mental disorder with blunted affect, emotional withdrawal and social withdrawal, and as well as auditory hallucinations, paranoia, delusions and conceptual disorganisation. It is frequently accompanied by significant social or occupational dysfunction. It has been established that the mortality in patients with schizophrenia is significantly higher than in the general population. Pascal *et al* reported that death rates of schizophrenia patients are two to three times higher than those observed in the general population, but the mechanisms of the higher mortality with schizophrenia patients is unclear. However, their relative insensitivity to physical pain, as reviewed in some papers,^{2 3} could be an explanation for some of this cause. It is reported that schizophrenia patients are insensitive to physical pain associated with illness and injury compared with non-schizophrenia patients. A case study reported that a schizophrenia patient with lack of pain had a perforated small bowel, which led to a delay in its diagnosis and surgical intervention.⁴ Other literature suggested that the widely accepted notion of reduced pain sensitivity in schizophrenia is related more to a different mode of pain expression than to a real endogenous analgesia. In this case as well, our patient could answer any questions and did not have severe DM neuropathy, but never complained of pain, which led to a late diagnosis of the muscle abscesses of both legs. We searched thoroughly for more abscesses since *S aureus* grew in his blood culture. We carefully examined his whole body by CT scan, and found deep-seated abscesses.

Osteomyelitis of the zygoma is extremely rare, since the zygoma receives a relatively sufficient supply of blood, and

patients with facial disease visit hospitals early due to facial pain or abnormality of appearance. Only few literatures reported it in PubMed database, and these causes are usually due to odontogenic sources, infected fracture sites, tuberculosis or candidiasis.^{6–8} In this case, *S aureus* osteomyelitis of the zygoma was more likely to have spread a contiguously due to cellulitis or subcutaneous abscess, possibly exacerbated by DM, a different mode of pain expression, our patient's indifference to his own appearance, arising from blunted affect, emotional withdrawal and social withdrawal. If our patient was a non-schizophrenia patient, he would have visited a hospital earlier, due to facial pain or abnormality of appearance, and would not have had osteomyelitis of zygoma.

We should keep in mind that schizophrenia patients are insensitive to pain, and that they may have multiple, unpredictable and rare diseases like our case. Careful and thorough examinations are essential for treatment in schizophrenia patients.

Learning points

- ▶ It is reported that schizophrenia patients are insensitive to physical pain.
- ▶ Careful and thorough examinations are essential for treatment in schizophrenia patients.
- ▶ Schizophrenia patients may have multiples, unpredictable and rare diseases, such as complicated staphylococcal osteomyelitis with bacteraemia and multiple secondary abscesses.

Competing interests None.

Patient consent Obtained.

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