

case report

Abscess of C1/C2 cervical vertebrae – errors in diagnosis and therapy

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Background. Nonspecific upper cervical spine vertebra osteomyelitis is very rare. It is caused most often by contiguous spread from an adjacent focus of infection and rarely by haematogenous dissemination from an extraspinal one. We present a rare case of Coagulase-negative *Staphylococcus* spp cervical vertebra osteomyelitis, where the clinical presentation of the disease is often atypical.

Case report. We analysed the case of 57-year-old female, where we found the diagnostic error in identification of the atlas subluxation on the radiograph and neglected laboratory findings indicating the urinary infection. These led to the disease progression and occurrence of neurological symptomatology, presented with tetraparesis. A prompt surgery in two steps was planned: the urgent surgical anterior decompression and then the occipitocervical fixation, but the patient died after the first surgical session.

Conclusions. The early recognition of symptoms and a prompt diagnosis are always essential for the onset of the accurate therapy. An additional destabilization of the affected segment done by the surgical decompression in the first step without the adequate stabilization may lead, as shown here, to a sudden fatal outcome.

Key words: vertebral osteomyelitis; magnetic resonance imaging; surgery

Introduction

Nonspecific vertebral osteomyelitis accounts for 3-4% of all spinal infections. The cervical spine is affected in 3-10% cases, while the epidural abscess in this region

is estimated to occur in approximately 1/100.000 people.¹ The infection is developed by haematogenous dissemination from an extraspinal focus or direct inoculation of a causative agent into the venous plexus around odontoid process during the intervention in the pharynx.

In most cases the causative pathogen is *Staphylococcus aureus*. Despite the fact that the clinical presentation is often atypical, which leads to a delayed diagnosis, an early diagnosis of the disease is essential for the

Received 24 April 2009

Accepted 14 June 2009

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adequate treatment.² The erroneous interpretation of radiographs additionally delays a proper diagnosis and commencement of the treatment. A two-step surgical strategy can lead, as shown in here, to the risk of developing an additional destabilization of the vertebral segment. The risk is even greater if the decompression is not accompanied by the adequate fixation.

Pyelonephritis as a source of haematogenous dissemination of the pathogen, to our knowledge, has not been specifically reported. Long time wasted, till a proper diagnosis was made due to serious overlooks in reading radiographs and subsequent inadequate treatment, rare cause of the disease, and an unexpected outcome despite the favourable clinical course, make this case interesting.

Case report

A 57-year-old female admitted to the Neurology Clinic with progressive weakness and numbness in extremities, more on the right side. On the clinical examination, she complained of pain in the neck and head, behind the ears and in the region of temporomandibular joints, and hard swal-



Figure 1. Lateral radiograph of the cervical spine taken one month after the onset of symptoms. Anterior atlantodental interval of 9 mm indicates anterior atlas subluxation.

lowing. The discomfort lasted for about a year, while progressive weakness in her extremities appeared one week before her admittance. The symptoms began after a cold, left undiagnosed and untreated, which had stopped spontaneously. She did not complain of urinary system discomfort. With "the neck-pain syndrome" diagnosis, she was treated by a physiatrist for several months, without success, X-ray films of the cervical spine taken as outpatient clinic six months before her admittance revealed atlantoaxial subluxation (Figure 1). Duplex US – carotid, TCD-VB – normal, electromyography made three months before admittance indicated mild neurogenic lesion of the C7 and C8 myotome, bilateral Carpal Tunnel Syndrome, and thoracic outlet syndrome to the left.

The clinical examination revealed that the patient is subfebrile with anterior, posterior cervical, submandibular and suboccipital lymphadenopathy. A mild torticollis to the right with a spasm of the paravertebral musculature, more pronounced on the right was observed as well as a mild protrusion of the head in relation to the trunk. Neurological findings showed somewhat lower left angle of her lips, and tetraparesis of spastic type affecting extremities, also more pronounced on the right side. Three days before the admittance, the patient was not able to walk alone. Relevant laboratory findings were: RBC: $4.75 \times 10^{12}/L$, WBC: $9.0 \times 10^9/L$, Ht: 40.1, Hgb: 13.4 g/L, SR: 50/76, CRPN: 59.8 mg/L, Fibrinogen: 4.77 g/L, Glucose: 6.21 mmol/L. T3 and T4 – normal, Urine: slightly turbid, yellow in colour, pH = 6.0, relative density: 1025. Urine sediment: elongated erythrocyte 4-6, leukocytes 2-3, epithelium cells 2-3, uric acid crystals 3-4.

During the hospitalization: contrast enhanced CT of the brain and chest X-ray was normal. MRI (magnetic resonance imaging) of the cervical spine showed an extensive soft tissue infiltration at the C1-2 level with



Figure 2. MRI images of the cervical spine:

- A. Axial T1-weighted image of the cervical spine at the level of C1. Formed abscess infiltration, partial erosion of the right atlantoaxial joint, surrounded dens, and abscess penetration into the epidural space.
- B. Sagittal T1-weighted image demonstrates narrowing of the spinal canal at the craniocervical junction to 11 mm. Enlarged posterior longitudinal ligament. Myelopathy signal at the level of dens.

a massive prevertebral and minor epidural extension followed by the narrowed spinal canal with a sagittal diameter of 11 mm. Signs of myelopathy at the C1 level. The pathological change caused a shift in the anterior longitudinal ligament up to the C4 vertebra (Figures 2 a,b), PPD₃ showed normal reaction.

The inflammatory process was diagnosed and treated by cefotaxime 2.0 g/12 h, vancomycin 1.0 g/8 h and metronidazole 1.0 g/8h. The haemocultures were negative.

A surgical treatment was pursued urgently. The patient underwent the incision via an anterior, transverse approach to the upper cervical spine. After the incision of the abscess membrane, greyish pus was evacuated. The abscess was removed by curettage and aspiration. As the surgery was planned in two steps – decompression and then occipitocervical fixation – the instrumentation in the first step was not performed.

Postoperative complications were not registered and the clinical presentation improved. Pain in the neck decreased. The

cervical spine was immobilized by a hard cervical collar. The antibiogram revealed *coagulase-negative Staphylococcus spp* as the cause of infection. The further therapy was continued with the same antibiotics. During the next ten postoperative days, the signs of tetraparesis were in gradual regression, while the laboratory indicators confirmed a decrease in infection.

During the night of the tenth postoperative day, however, the patient died. The autopsy performed revealed spinal cord tissue oedema, at the level of change, with overfilled blood vessels. In the upper spinal cord region, signs of myelopathy were evident, but not those of bleeding. There were no signs of pus penetration into the dural space (Figure 3a). Pyelonephritis was found, probably the cause of haematogenous spread of infection (Figure 3b).

Discussion

Coagulase-negative Staphylococcus, present in our case, is very rare as an agent of the spi-

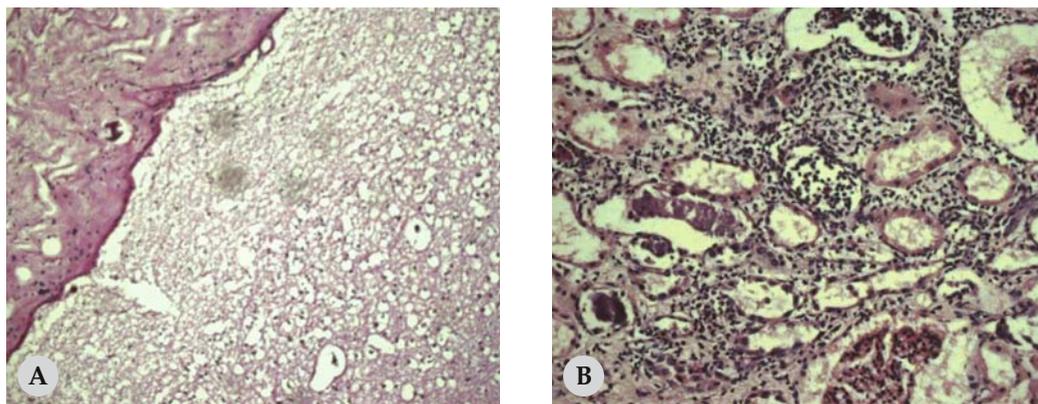


Figure 3. Microscopic images of the spinal cord and the kidney taken at autopsy:

- A. Cross-sectional image of the spine cord at the craniocervical junction. Spinal cord edema. Presence of microthrombi in some blood vessels obliterating the lumen. Pia mater is edematogenic with dilated blood vessels. Very close contact between the two maters around medulla, with partial obliteration of the subarachnoid space and pia and dura mater grown together.
- B. Interstitial tissue of the kidney is edematogenic with dilated blood vessels and irregularly distributed granulocytic inflammatory infiltrator. Tubuli are characterized by serious parenchymatous degeneration of cells, while extended lumens are filled with homogenous eosinophilic cylinders. Glomeruli are intact.

nal infection. Hadjipavlou *et al.*³ reviewed 34 cases of the spinal infection managed with the surgical percutaneous transpedicular drainage and found positive cultures in 73.5% cases out of which 16% case of infection with *coagulase-negative Stahylococcus* as the causative pathogen. A meta-analysis of 915 patients with the spinal epidural abscess, made by Reihnsaus *et al.*⁴ confirmed the pathogen in 753 cases, but only in 35 ones *coagulase-negative Stahylococcus spp* was found as an etiologic factor.

The disease is usually insidious and progresses slowly over a long period of time so that in a large number of cases it has been recognized with a delay. Zigler *et al.*⁵ described the difficulties a clinician is faced with in diagnosing this disease due to the atypical clinical presentation. Ross *et al.*⁶ reported on a rarely present symptomatic triad for the early diagnosis of the disease - elevated temperature, neck pain, and progressive neurological deficit. Also, cervical spine lymphadenopathy, spasm of the neck musculature and progressive torticollis followed by subfebrility -as early symptoms of

the upper cervical spine inflammation were described by Busche *et al.*⁷

Her diagnosis was "the neck pain syndrome", which was treated with the physical therapy for several months. The proper diagnosis of the disease was delayed for more than a year. Busche *et al.*⁷ described a young man with diabetes and similar symptoms in the upper cervical spine of six months' duration before a sudden change for the worse and a proper diagnosis was made. Noguchi *et al.*⁸ described the case of an elderly patient who complained of neck pain and stiffness of 7 days' duration and had symptoms of meningeal irritation. CT and MRI scans revealed odontoid osteomyelitis with the abscess. The authors recommend magnetic resonance imaging in the early stage of the disease emphasizing its important role in monitoring the disease evolution. Their diagnosis was based on CT and an early use of magnetic resonance imaging.^{9,10}

Additional radiological diagnostics in our case was not adequately carried out. Plain X-ray of the cervical spine was taken

relatively early, a month after the onset of the symptoms, but was interpreted erroneously. On a lateral X-ray image, taken at that time, an increase in atlantodental interval for more than 2.5 mm can be seen, as a sign of anterior atlantodental subluxation. Neither an open mouth radiograph, which is standard in diagnostics of upper cervical spine pathology¹⁰, nor dynamic lateral radiographs were taken. Abnormalities in the upper cervical spine region are frequently accompanied by abnormal parameters on plain radiographs even before clear clinical symptomatology. In our case, the protrusion of the patient's head might have arisen additionally suspicion of the changes in the cervical spine. As shown in some biomechanical papers, alterations in the static alignment of the cervical spine segments as a result of the pathological process cause alterations in radiographic parameters on dynamic radiographs.

The patient did not receive immobilization for the following six months. The neurological symptoms got worse with the progression of the infection followed by destruction of ligaments and odontoid, due to the generated vertebral instability.¹² At the moment of her admittance, tetraparesis of spastic type was evident. Magnetic resonance imaging of the cervical spine revealed odontoid abscess and, therefore, the surgical therapy followed.

The untreated infection of the urinary tract, confirmed in our case by autopsy, probably triggered the occurrence of odontoid osteomyelitis. In 854 patients with the epidural spinal abscess, out of 377 extraspinal foci of infection Reihnsaus *et al.*⁴ reported 23 urinary infections as the source of the abscess. The infection spread through the bloodstream contaminating the venous plexus of the occipitocervical region and further the epistropheus odontoid. Young and Weaver¹³ stressed the patient's age (over 60 years), diabetes, oral infection, and

medication misuse as crucial risk factors. Only the clinically undeveloped urinary infection as a cause of temporary bacteriuria was present. It was not, however, recognized as a possible focus of infection, and was left untreated, haemocultures were found to be positive in about two thirds of cases, but in our case they were negative.

The treatment of odontoid osteomyelitis with the abscess is controversial. Some authors emphasize successful clinical outcomes achieved in patients with vertebral osteomyelitis treated without surgery, especially in its early phase, when the segment is still stable.¹⁴ However, successful outcomes by the operative treatment performed in one or two stages⁵ have been reported in numerous recent papers. Neurological findings revealed a progressive cervical spine myelopathy, required an urgent surgical decompression in our patient. A two-step surgical treatment was planned, as suggested by Nakase *et al.*¹⁵ We had no experience with anterior instrumentation at the C1/2 level. It seemed at that moment that a simultaneous posterior occipitocervical fixation would be too invasive for our patient. Therefore, the patient underwent the anterior decompression. Postoperatively, some signs of resolving of neurological deficits were registered, laboratory findings improved and the patient felt better.

Sudden death of the patient in spite of promising clinical course has not been described until now. Mortality caused by cervical spine osteomyelitis complicated by abscess accounts for 15% of all cases.⁴ The autopsy revealed brain tissue swelling, ischemic necrosis at the site of spinal cord compression, and lung tissue swelling. No macroscopic changes in other organs were found, except for the right kidney pyelonephritis.

The abscess extended through the retropharyngeal space along the anterior longitudinal ligament up to the C4 level. The au-

topsy did not reveal the intradural abscess in the upper segment of the cervical spine cord. Probably sudden hypotension and respiratory arrest occurred during the spinal cord shock due to subarachnoid inflammation of small blood vessels and their thrombosis.

Such a small blood vessels thrombosis may, or may not be seen macroscopically at autopsy. Brain swelling could also be a result of the intracranial complication of bacterial meningitis.¹⁶ Neither clinical presentation of meningitis nor acute central cord syndrome was found in our patient. Probably the cause was the repeated compression of the spinal cord by unstable C1 segment. Such unstable segment, after decompression, caused repeated *microtraumas* - transient spinal cord ischemia, which led to blood vessel thrombosis in that part of the spine cord. Our case confirms that even in the situation of "partially repositionable" atlas subluxation there is a certain risk of the spinal cord compression, which can be fatal. This leads to a conclusion that the very evacuation of the granular tissue – as the first surgical act, can hide a potential risk of the additional destabilization of the segment and can impose a need for more secure postoperative stabilization of the cervical spine. The cervical spine orthosis, provided here, proved as unsatisfactory. Postoperatively, it was necessary to use, up to the following surgical step, halo-traction or halo traction-apparatus. Additionally, diagnostic and therapeutic overlooks, which to a great extent contributed to such an outcome, remind us of the fact that the patient should always be considered as a whole, and especially in cases of the treatment of so severe infections.

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