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## Case report

# Cervical spondylodiscitis following cricopharyngeal botulinum toxin injection

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## ABSTRACT

**Background:** Iatrogenic cervical spondylodiscitis is rare, but may occur after various medical interventions.

**Methods:** We report a case of a diabetic 70-years-old female with C5–C6 spondylodiscitis and symptomatic epidural abscess with neck pain and upper limb paresis after endoscopic botulinum toxin injection for the treatment of dysphagia. Treatment included antibiotic therapy with amoxicillin and later on benzylpenicillin for the next ten weeks and corporectomy with spondylodesis.

**Result:** The patient made an excellent recovery, with complete resolution of paresis and only minor residual hypoesthesia at one year after operation.

**Conclusion:** Cervical spondylodiscitis should be considered early, in patients with neck pain after endoscopic cricopharyngeal injection, as timely diagnosis and treatment can prevent serious and irreversible neurological deficit.

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## 1. Introduction

Pyogenic spondylodiscitis is a purulent bacterial infection of the vertebral body, intervertebral disc, or both. Due to the relatively small diameter of the vertebral canal in relation to the diameter of the cervical spinal cord, spondylodiscitis in the cervical spine often results in severe neurological deficits, such as paraparesis or hypesthesia [1]. Presenting symptoms can be relatively insidious and nonspecific, often resulting in a delayed clinical diagnosis [2]. However, timely diagnosis and adequate treatment are critical to prevent severe neurological deficit [3]. Endoscopic cricopharyngeal botulin toxin injection has never been associated with post-procedural spondylodiscitis. We present the first report of a patient that suffered progressive serious neurological deficit due to bacterial spondylodiscitis following botulin injection for the treatment of dysphagia.

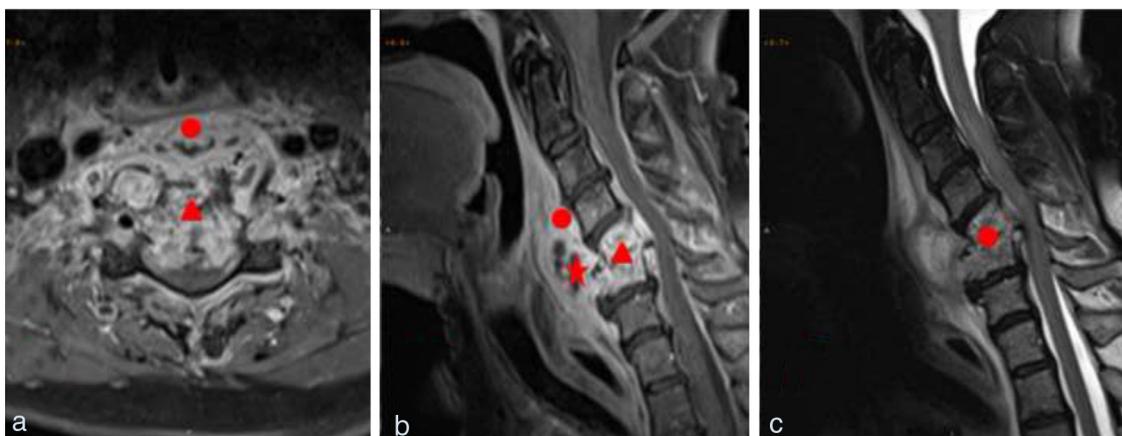
## 2. Case-report

A 70-year-old woman with type II diabetes treated with novorapid/levemir insulin injections, had received endoscopic injection of botulinum toxin in the cricopharyngeal muscles for the

treatment of dysphagia, caused by hypertonicity of the cricopharyngeal (CP) muscles. The injection was directed to the lower part of the CP muscle compartment via a direct laryngoscopic approach, near the C5 and C6 vertebra, and consisted of a bolus of 50 units of botulinum toxin. The endoscopy was straightforward, without any noticeable mucosal injury. After three weeks this patient started to complain about progressive neck pain and she was admitted to the neurological department. She was neurologically intact. Body temperature was 38 degree Celsius, laboratory examination revealed an elevated white blood cell count of  $18.7 \times 10^9 L^{-1}$ , Magnetic Resonance Imaging (MRI) of the neck demonstrated an anterolaterally located paravertebral and epidural mass with slight rim enhancement compatible with the diagnosis of abscess. Also there was a small amount of gas in the paraspinal tissue and inside the spinal canal. Intravenous administration of Amoxicillin 2000 mg every 4 hours was initiated empirically, after taking blood samples for microbiological culture. Over the next day, the patient noticed weakness of the right arm and numbness of the right shoulder, after which she was transferred to our neurosurgical unit. Neurologic examination revealed a paresis of the right deltoid and biceps muscles, as well as hypesthesia and hypalgesia in the C4, C5 and C6 dermatomes on the right side. An additional MRI scan demonstrated an increase of the epidural mass, now with spinal cord compression (Fig. 1A and B). An anterior C5 and C6 corporectomy with abscess-evacuation and spondylodesis (one stage procedure) was performed. Intraoperative acquired

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**Fig. 1.** Axial (A) and sagittal (B) T1 weighted magnetic resonance images with fat saturation after intravenous Gadolinium show enhancement of vertebral endplates C5 and C6 (▲) and enhancement of prevertebral and epidural soft tissues (●) around a low-density centre (\*) suggestive of an abscess. Sagittal (C) T2 weighted STIR magnetic resonance image shows destructive changes at the C5/6 disc (●) with endplate sclerosis.

collections of pus were sent for microbiological analysis. Gram staining and cultures were inconclusive, but the blood cultures revealed *Streptococcus Intermedius* and *Streptococcus Constellatus*. Antibiotic treatment was switched to Benzyl penicillin 12 million units intravenously. Postoperatively, the patient's condition improved, with progressive recovery of motor function. She was discharged to the referring hospital for further recovery. Antibiotic treatment was continued for the next 10 weeks; the first six weeks after surgery intravenously, the final four weeks with oral administration of clindamycin 600 mg every 8 hours. One year after surgery motor deficit has resolved completely, with a discrete C6 dysaesthesia as the residual symptom.

### 3. Discussion

To the best of our knowledge, this is the first report of spondylodiscitis and retropharyngeal epidural neck abscesses following endoscopic cricopharyngeal botulinum injection. Cervical spinal infection has been reported after several diagnostic or therapeutic procedures including corticosteroid or analgesic injection [4], esophagoscopy [5] and esophageal stenting [6]. Spondylodiscitis can result from direct inoculation or from hematological or lymphatic dissemination of a distant infection. The presence of *Streptococcus* species in the blood cultures in the present case, typically originating from the oral/oesophageal mucosa, seems indicative for a causal relationship between the botulinum injection and the spondylodiscitis, as these microorganisms reside in the gastrointestinal tract.

The most likely explanation for the development of spondylodiscitis and epidural abscess is hematogenous spread of local micro-organisms from the retropharyngeal soft tissues via the

anterior extravertebral venous plexus into the basivertebral veins and the anterior internal vertebral venous plexus. All these venous structures are part of the valveless vertebral venous system, which is a voluminous and relevant, but nevertheless ignored collateral venous network [7,8] (Fig. 2A and B).

The diabetic status of this patient may have served as a predisposing factor, contributing to the development of this infection. Omitting antibiotic prophylaxis in this particular case may have promoted the development of bacteraemia.

Early-stage vertebral osteomyelitis can be treated conservative, starting with intravenous antibiotics for two weeks, followed by oral antibiotics. The optimal duration of antibiotic treatment has been debated, although several studies suggested non-inferiority of 6 weeks versus 12 weeks in patients with pyogenic vertebral osteomyelitis [9]. Failure of antibiotic therapy, the development/occurrence of spinal instability, and/or neurological deterioration necessitate surgical intervention. In the present case, surgery was indicated because of the neurological deterioration. Anterior corpectomy with spondylodesis was opted because of the complete destruction of the vertebral bodies involved. Various studies have demonstrated that spinal instrumentation in patients with osteomyelitis is safe, showing similar recurrence rates of infection between instrumented and non-instrumented surgery [10]. Possibly more important than the specific treatment chosen, is the timely diagnosis of this complication, as the delay to diagnosis is an independent risk factor of poor outcome [3]. Therefore, patients with neck pain and/or neurological symptoms after cricopharyngeal botulin injection should raise suspicion of an infectious complication by the consulting physician.

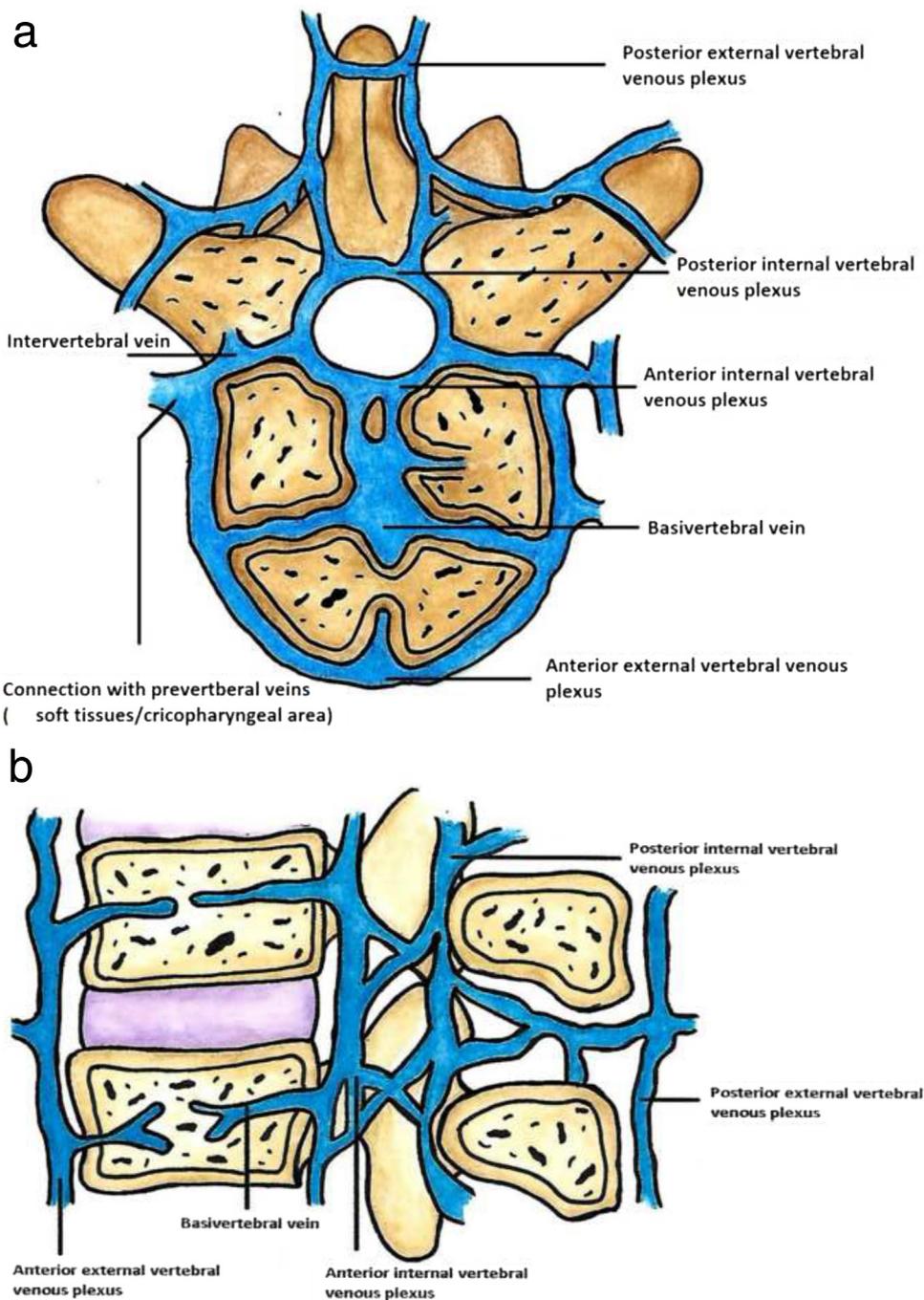


Fig. 2. Schematic representation of the vertebral venous system in a transverse (A) and a sagittal (B) section.

#### 4. Conclusion

Cervical spondylodiscitis with epidural abscess and spinal cord compression may occur as a complication of cricopharyngeal botulin toxin injection. Early recognition and prompt antibiotic treatment is the key to prevent serious neurological deficit. Knowledge of the related anatomical structures, especially awareness of the existence and function of the vertebral venous system, will help to understand the pathophysiology and to anticipate proper treatment.

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#### Disclosure of interest

The authors declare that they have no competing interest.

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