Unexpected outcome (positive or negative) including adverse drug reactions

An infected branchial cyst complicated by retropharyngeal abscess, cervical osteomyelitis and atlanto-axial subluxation

Richard Bullock,¹ Deanne P Soares,^{1,2} Marsha James³¹ Section of Radiology, Department of Surgery, Radiology, Anaesthetics and Intensive Care, University of the West Indies, Kingston, Jamaica² Department of Radiology, University Hospital of the West Indies, Kingston, Jamaica³ Department of Otorhinolaryngology, Bustamante Hospital for Children, Kingston, Jamaica

Correspondence to Richard Bullock, richardbullock801@msn.com

Summary

We present a case of a 44-year-old man who visited his general practitioner for recurrent neck swelling, which was found to be a neck abscess. It was aspirated, later recurred, and then surgically excised. Histology was consistent with an infected branchial cyst. Eight months after discharge, the patient presented with a history of progressive neck pain and stiffness and eventually bilateral upper limb weakness. MRI demonstrated a prevertebral abscess complicated by cervical osteomyelitis and atlanto-axial instability. The abscess was drained and appropriate antibiotic treatment was administered. The patient responded well with full recovery of his upper limb strength and resolution of the abscess. However, he had mild persistent neck stiffness.

BACKGROUND

This case serves to highlight the importance of imaging in patients with persistent neck pain particularly when associated with a history of recurrent infection. Imaging, particularly MRI, facilitates diagnosis of underlying lesions such as branchial cysts. Imaging would also be valuable in excluding or demonstrating the extent of associated unusual but potentially serious complications.

CASE PRESENTATION

A 44-year-old man presented to his general practitioner with a 1-year history of recurrent left-sided neck swelling. On examination, he was found to have a tender left-sided neck mass with associated bilateral cervical lymphadenopathy. The presence of an abscess was confirmed by ultrasound, which demonstrated a thick walled collection in the anterior triangle of the neck lateral to the common carotid artery and internal jugular vein. The abscess was aspirated and culture positive for *Staphylococcus aureus*. Despite antibiotic treatment the abscess re-collected and 2 months after presentation the cavity was surgically excised. Histology revealed features of an infected branchial cyst.

Four months later the patient re-presented with fever and neck stiffness. Meningitis was considered. Lumbar puncture was negative for infection. A CT scan of the brain was performed, which showed no evidence of intracranial sepsis but images through the skull base suggested atlanto-axial instability. Two aetiologies were considered: rheumatoid arthritis and cervical osteomyelitis. The patient had no known chronic illnesses and his immune status was normal. Due to the lack of an available neurosurgical service at his local hospital he was transferred to another institution where a more detailed history revealed progressive

weakness and numbness of both upper limbs. On examination, he was found to have reduced power in both upper limbs. CT and MRI of the spine were requested and demonstrated a retropharyngeal abscess, cervical osteomyelitis and atlanto-axial subluxation. There was associated spinal canal stenosis and cord compression.

INVESTIGATIONS

The CT scan (figure 1) was performed with 1.5 mm slices from the skull base to C3 level. The predental space was widened measuring 1.2 cm with posterior displacement of the odontoid peg and narrowing the foramen magnum. The odontoid peg, lateral masses of C1, and the bodies of C2 and C3 all demonstrated erosion.

On axial and sagittal T2W MRI (figures 2 and 3), the predental space was again noted to be widened measuring 1.3 cm (normal <5 mm). The transverse atlantal ligament was lax with posterior displacement of the dens with resultant narrowing the foramen magnum and compression of the cervical cord and medulla oblongata. The compressed cord demonstrated increased signal intensity suggestive of myelomalacia. There were extensive prevertebral inflammatory changes from the skull base to the C6 vertebral level. A pocket of fluid was seen in the prevertebral soft tissues suggestive of a prevertebral abscess. Both anterior and posterior elements of C1–C5 show marrow signal changes likely representing osteomyelitis. The tip of the odontoid peg was eroded. There was also an epidural collection from C2–C5 narrowing the spinal canal and causing additional cord compression.

DIFFERENTIAL DIAGNOSIS

Rheumatoid arthritis and Grisel's syndrome.

Figure 1 Axial CT of atlanto-axial articulation.

Figure 2 Axial T2* MRI of atlanto-axial articulation.

Figure 3 Sagittal T2W MRI of upper cervical spine.

Figure 4 Sagittal T2W MRI of upper cervical spine (postsurgical).

TREATMENT

The prevertebral abscess was drained by the ear, nose and throat (ENT) specialists via an anterior approach. Pus was sent for culture and sensitivity, which revealed a significant growth of *S aureus*—the same strain as that previously derived from the infected branchial cyst. The patient was placed on culture-directed antibiotics.

OUTCOME AND FOLLOW-UP

The patient remains well 2 years post follow-up with only residual neck stiffness. The weakness in his upper limbs has resolved completely. He no longer has any clinical signs of a neck abscess or inflammatory changes in the neck. This was confirmed on follow-up MRI (figure 4). However, the latter demonstrated fusion of his upper cervical vertebra (C2–C4) and bony stenosis of the cranio-cervical junction with cord compression. This is the likely cause for his persistent neck

stiffness. Though neurologically he has improved, the bony stenosis of the cranio-cervical junction with cord compression remains cause for concern. Further ENT and neurosurgical surveillance has been arranged.

DISCUSSION

Atlanto-axial subluxation is a serious condition that can lead to paraplegia and death due to cervical cord compression. The upper cervical spine (C1–C2) is a complex articulation whose integrity is made up of bony and ligamentous structures. The bony structures include the combined anterior and posterior arches of C1 and the odontoid peg (dens of C2). The supporting ligaments are the transverse atlantal ligament, the paired alar ligaments and the apical ligament of the dens. Instability results from disruption or laxity of these ligaments and/or fractures of the dens or C1 arches.

Imaging of the C1–C2 articulation can be done with plain radiographs, CT or MRI. Plain radiographs are useful for delineation of bony alignment and for fractures, CT for patterns of bony destruction or fractures¹ and MRI for spinal cord and nerve root evaluation. MRI can also help in delineating the ligaments.²

Atlanto-axial dislocation has several possible aetiologies. The more common causes include trauma,³ congenital defects^{4–5} and connective tissue disorders such as rheumatoid arthritis.⁶ Adjacent neck infections can be associated with laxity of the transverse atlantal ligament, particularly in children. In such cases this is referred to as Grisel's syndrome.^{7–8} Osteomyelitis is an uncommon entity in the cervical spine. It is more commonly noted in the thoracic spine in association with direct spread from chest infections or the lumbar spine from genitourinary spread.

Branchial cysts are congenital lesions that can become infected. Infected branchial cysts are a recognised cause of persistent or recurrent neck infections/abscesses.⁹ A literature search revealed only two case reports of infected branchial cleft cysts becoming complicated by retropharyngeal abscess.^{10–11} No cases of cervical osteomyelitis or atlanto-axial subluxation resulting from spread of infection from a confirmed branchial cleft cyst were noted, although cervical osteomyelitis is a recognised complication of retropharyngeal infection from other causes, including upper airway infection, dental sepsis and foreign body perforation.^{12–14} Osteomyelitic destruction of the odontoid peg has been recorded in association with retropharyngeal abscesses^{15–16} and is associated with neck stiffness and meningism.

Recurrent neck infections should raise the possibility of an underlying congenital lesion such as a branchial cyst. In this case, despite aspiration and surgical excision of the infected branchial cyst, the infection recurred and spread through the retropharyngeal space to involve the upper cervical vertebrae from C1–C5. In association with infection at the C1–C2 level, there was relaxation of the transverse atlantal ligament leading to atlanto-axial instability with neck stiffness and meningism as seen in our case.

Learning points

- ▶ Persistent neck infections may suggest an underlying congenital lesion such as a branchial cyst.
- ▶ Retropharyngeal abscesses may become complicated by osteomyelitis.
- ▶ Persistent neck pain should always signal the possibility of occult spinal pathology.
- ▶ MRI can provide early diagnosis of osteomyelitis.
- ▶ Cervical osteomyelitis can become complicated by atlanto-axial instability.

Competing interests None.

Patient consent Obtained.

REFERENCES

1. Hua Q, Ma WH, Zhao LJ, *et al.* [Clinical application of multi-spiral CT thinner scanning and reconstruction in the diagnosis of atlantoaxial fracture and dislocation]. *Zhongguo Gu Shang* 2009;**22**:349–52.
2. Pfirrmann CW, Binkert CA, Zanetti M, *et al.* MR morphology of alar ligaments and occipitoatlantoaxial joints: study in 50 asymptomatic subjects. *Radiology* 2001;**218**:133–7.
3. Levine AM, Edwards CC. Traumatic lesions of the occipitoatlantoaxial complex. *Clin Orthop Relat Res* 1989;**53**:68.
4. Vickers ED. Atlantoaxial anomalies with particular emphasis on os odontoidum. *J Manipulative Physiol Ther* 1990;**13**:471–6.
5. Ali FE, Al-Bustan MA, Al-Busairi WA, *et al.* Cervical spine abnormalities associated with Down syndrome. *Int Orthop* 2006;**30**:284–9.
6. Timponi V, Lomasney L, Demos TC, *et al.* Radiologic case study. Diagnosis: atlantoaxial instability secondary to rheumatoid arthritis. *Orthopedics* 2006;**29**:963, 1037–41.
7. Youssef K, Daniel S. Grisel syndrome in adult patients. Report of two cases and review of the literature. *Can J Neurol Sci* 2009;**36**:109–13.
8. Ugur HC, Caglar S, Unlu A, *et al.* Infection-related atlantoaxial subluxation in two adults: Grisel syndrome or not? *Acta Neurochir (Wien)* 2003;**145**:69–72.
9. Nusbaum AO, Som PM, Rothschild MA, *et al.* Recurrence of a deep neck infection: a clinical indication of an underlying congenital lesion. *Arch Otolaryngol Head Neck Surg* 1999;**125**:1379–82.
10. Huang RY, Damrose EJ, Alavi S, *et al.* Third branchial cleft anomaly presenting as a retropharyngeal abscess. *Int J Pediatr Otorhinolaryngol* 2000;**54**:167–72.
11. Hewel K, Kioumehri F, So G, *et al.* Infected third branchial cleft cyst: retropharyngeal extension to the superior mediastinum. *Can Assoc Radiol J* 1996;**47**:111–13.
12. Karlidag T, Alpay HC, Kaygusuz I, *et al.* [Retropharyngeal abscesses: a retrospective analysis of 10 patients]. *Kulak Burun Bogaz Ihtis Derg* 2008;**18**:300–5.
13. Jang YJ, Rhee CK. Retropharyngeal abscess associated with vertebral osteomyelitis and spinal epidural abscess. *Otolaryngol Head Neck Surg* 1998;**119**:705–8.
14. Paul CA, Kumar A, Raut VV, *et al.* Pseudomonas cervical osteomyelitis with retropharyngeal abscess: an unusual complication of otitis media. *J Laryngol Otol* 2005;**119**:816–18.
15. Willenborg KM, St ver T, Becker H, *et al.* [Parapharyngeal abscess and osteomyelitic destruction of the odontoid process]. *Laryngorhinootologie* 2007;**86**:128–30.
16. Kubo S, Takimoto H, Hosoi K, *et al.* Osteomyelitis of the odontoid process associated with meningitis and retropharyngeal abscess—case report. *Neurol Med Chir (Tokyo)* 2002;**42**:447–51.

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