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Transient quadriplegia following re-puncture of tracheo-oesophageal fistula

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Abstract
We present a case of a laryngectomized patient who underwent re-puncture of tracheo-oesophageal fistula for speaking valve voice restoration, resulting in a previously unreported inherent complication of this procedure: extradural abscess of the cervical spine with transient quadriplegia.

Key words: Cervical Vertebrae; Epidural Abscess; Larynx, Artificial; Quadriplegia

Case history

A 54-year-old man underwent salvage total laryngectomy for recurrent glottic T2N0M0 squamous cell carcinoma, after radical radiotherapy given two years previously had failed. A primary puncture for tracheo-oesophageal speech restoration was carried out with a standard Provox valve. Owing to persistent leakage around the Provox valve, not responding to purse-string suturing, the fistula was downsized and a 16-FG low-pressure Blom–Singer valve substituted. Histology showed clear resection margins.

Following laryngectomy, the patient developed recurrent benign stenoses of the pharyngeal remnant requiring periodic dilatation. On one such occasion three years later, the valve became extruded and difficult to re-insert due to loss of the fistula, so a re-puncture procedure was performed under general anaesthesia and a new valve (Blom–Singer low pressure 6 mm) was inserted. The procedure was difficult and required a paediatric pharyngoscope to visualize the trocar due to the stricture. There was no evidence of malignancy. The patient was discharged the following day.

Three days later the patient was re-admitted complaining of throat and neck pain, difficulty in swallowing, fever, generalized malaise, and weakness in both arms. The valve was removed and he was treated with antibiotics, with a presumptive diagnosis of peristomal cellulitis. Cervical spine X-ray revealed some narrowing of C5/6 with anterior osteophytes. Contrast swallow showed air in the soft tissues of the neck but no leakage was seen outside the pharyngo-oesophageal segment. An isotope bone scan was reported as normal, with no evidence of bony metastases.

Upon orthopaedic consultation the patient was found to be septic with grade IV weakness of grip in both hands, with no other focal neurological signs. Non-steroidal anti-inflammatories, nil by mouth with nasogastric tube feeding, and a soft cervical collar were instigated. The patient's condition rapidly improved and he was discharged nine days later.

Four weeks following the re-puncture the patient was brought to the ENT clinic with a complaint of very severe pain and limited abduction of the right shoulder (80°). The spinal accessory nerve appeared intact. Urgent orthopaedic consultation was arranged, and evidence of right-sided radiculopathy in a C6/7 distribution was found. X-rays of the cervical spine showed some loss of normal curvature as well as degenerative changes. An urgent magnetic resonance imaging (MRI) scan was scheduled for three days later, but in the meantime the patient's condition rapidly progressed to quadriplegia with urinary retention. Gadolinium-enhanced MRI scan of the cervical spine then revealed an extra-dural abscess with an infected disc at the C6/7level displacing and compressing the spinal cord posteriorly in the spinal canal (Figure 1).

The patient underwent emergency neurosurgical decompression of the cord by a C6/7 anterior cervical discectomy and evacuation of extra-dural empyaema through a vertical incision along the anterior border of the sternomastiod. Following isolation of *Staphylococcus aureus* from the abscess, he was treated with intravenous flucloxacillin and gentamicin for four weeks. The patient's condition slowly improved, with progressive recovery of motor function. A post-operative MRI scan showed resolution of the cord compression, and the patient was discharged home two weeks later.

Discussion

Tracheo-oesophageal puncture and insertion of a speaking valve is known to carry a risk of early and late complications, including submental abscess formation.[1] In our case, this procedure was complicated by the development of discitis and extra-dural abscess of the cervical spine. The most likely mechanism for this was the impingement of the trocar used to re-puncture the tracheo-oesophageal fistula through the posterior oesophageal wall and into the pre-vertebral space. A less likely cause was a perforation arising from the use of the

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rigid pharyngoscope at the site of stenosis. Osteophytes of the cervical vertebral bodies are known to be risk factors for perforation.[2] Inoculation of the retropharyngeal space and prevertebral soft tissues by oropharyngeal flora in this previously irradiated region resulted in pyogenic osteomyelitis and discitis, and consequential epidural abscess with spinal cord compression.

Most cases of cervical osteomyelitis are due to haematogenous spread of pyogenic micro-organisms (including TB) from a distant infective source.[3] There are cases of haematogenous spread following head and neck procedures, e.g. tonsillectomy,[4] adenoidectomy,[5] dental extraction[6] and mandibular fracture.[7] Given the circumstances of our case, haematogenous spread would be unlikely. Direct inoculation from an oesophageal perforation is a rarer cause of cervical osteomyelitis. Typically, the perforation results from penetrating trauma,[8] a foreign body or iatrogenic injury. The latter includes rigid oesophagoscopy,[2,9] anterior spinal surgery,[10] perforation of pharyngeal pouch by a nasogastric tube[11] and percutaneous emergency cricothyriodotomy.[3] One case of fatal cervical osteomyelitis was reported following tonsillectomy in which infiltration of local anaesthesia for glossopharyngeal nerve block was performed.[12] Blunt trauma has also been associated with oesophageal perforation and cervical spinal fractures.[13] Risk factors for progression to osteomyelitis are previous radiotherapy, residual malignancy, steroids, diabetes mellitus, immunopathy, intravenous drug abuse and age.[11]

As far as we are aware from our *Medline* and *PubMed* search, there is only one reported case in the literature of a retropharyngeal abscess and cervical spinal osteomyelitis following secondary tracheo-oesophageal puncture for insertion of a speaking valve.[14] In this case there were no reported neurological complications.

FIG. 1

Sagittal T1 weighted magnetic resonance imaging scan of the cervical spine with gadolinium enhancement, revealing an anterior extra-dural abscess with an infected disc at C6/7 displacing and compressing the spinal cord posteriorly.

In their review of 22 patients with cervical osteomyelitis or neck abcesses, Gaudinez *et al.*[10] reported that the micro-organisms involved were alpha *Streptococcus*, *Staphylococcus aureus*, *Psuedomonas aeruginosa*, *Bacteroides* species and anaerobic Gram-positive cocci.

- **A laryngectomized patient underwent re-puncture of a tracheo-oesophageal fistula for speaking valve voice restoration**
- **This resulted in the complication of an extradural abscess of the cervical spine, with transient quadriplegia**

From the literature, there appears to be a delay of a few weeks to months between the perforation and manifestation or diagnosis of the osteomyelitis. Our case presented relatively early owing to the neurological features.

The diagnosis is made on clinical assessment, and MRI scan appears to be the best imaging modality, with a reported 96 per cent sensitivity and 92 per cent specificity.[15] There is high intensity on T2 weighted image, and a low intensity on T1 weighted image which enhances with gadolinium.[3,11] Initial plain lateral cervical spine X-rays can be normal,[2,12,16] but a few months later may show obliteration of intervertebral spaces.[11] Computerized tomography may reveal surgical emphysema,[17] osteogenesis in the anterior cervical prevertebral space and, if contrast is given, a space-occupying lesion of the epidural space.[11] Looking for an oesophageal perforation by means of a contrast study or endoscopic evalution carries the risk of a false negative result,[10] as highlighted by our case, possibly because the perforation is small or heals before resolution of the cervical infection. The erythrocyte sedimentation rate and serial MRI scans can be used to monitor the course of the osteomyelitis.

The principles of management are to drain the abscess, debride necrotic bone and deal with any perforation (either conservatively or surgically). Long-term antibiotics are also recommended. Where appropriate, the cervical spine is stabilized.

The long-term outcome of spinal osteomyelitis is vertebral fusion, spinal deformity, sclerosis and limitation of movement.[11]

Conclusions

Surgeons performing tracheo-oesophageal puncture for voice rehabilitation following laryngectomy should be aware that the procedure carries a risk of cervical osteomyelitis and sub-dural abscess with neurological deficit. These typically present a few weeks following the procedure.

Osteomyelitis of the cervical spine presents insidiously with a varied clinical picture which is often overlooked. An early high index of suspicion, and early diagnosis and treatment, are the keys to a successful outcome.

There may be a role for pre-operative lateral cervical spine X-rays to assess curvature and osteophyte formation, known risk factors in this procedure.

It is now our practice to cover all secondary valve puncture procedures with a single dose of intravenous gentamicin, although we accept there is no evidence in favour of this step.

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