



Case Report

Neck angioedema after anterior cervical discectomy and fusion with coexistent epiglottic cyst

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ABSTRACT

Background: We present a case and reviewed the literature regarding airway obstruction and angioedema after an anterior cervical discectomy and fusion (ACDF).

Case Description: A 60-year-old female with degenerative cervical myelopathy and a previously undiagnosed epiglottic cyst underwent a C5–C6 ACDF; notably, the anesthesiologist found an epiglottic cyst when the patient was first intubated. Two hours postoperatively, the patient acutely developed severe neck swelling with airway obstruction due to angioedema. She was immediately treated with hydrocortisone and required a tracheostomy. The edema decreased markedly in the next 12 h and by the 3rd postoperative day it resolved. Three months later, she had no residual medical sequelae.

Conclusion: Patients with epiglottic cysts who need cervical spine surgery should either first have the cyst treated or should be closely monitored postoperatively.

Keywords: Angioedema, Anterior cervical discectomy and fusion, Cervical, Epiglottic, Spine

INTRODUCTION

Postoperative airway obstruction (PAO, partial or complete) after anterior cervical spine surgery (ACSS) occurs in 1.2–6.1% of patients and in up to 14% of cases following multilevel ACSS.^[6,7] PAO may be attributed to edema, hematoma, anterior extrusion of graft/hardware, abscess, swelling, internal jugular vein thrombosis, a major cerebrospinal fluid leak, and angioedema.^[6]

Epiglottic cysts (ECs) are usually asymptomatic lesions caused by mucus retention, appearing mostly in 60-year-old patients.^[5] Typically, these lesions are incidentally found by otolaryngologists, anesthesiologists, and gastrointestinal endoscopists.^[5] Here, we present a 60-year-old female with degenerative cervical myelopathy whose EC was incidentally found by anesthesia on intubation for a C5–C6 anterior cervical discectomy and fusion (ACDF): 2 h later, she acutely developed severe neck swelling and airway obstruction due to angioedema.

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MATERIALS AND METHODS

History and examination

A 60-year-old female presented with a 1-year history of progressive myelopathy attributed to a C5–C6 MR-documented cervical stenosis with cord compression. The modified Japanese Orthopedic Association (mJOA) score was 14. At surgery, during intubation, the anesthesiologist noticed an EC cyst on the lingual surface of the epiglottis. A C5–C6 ACDF was routinely completed utilizing polymethylmethacrylate-based bone cement (Palamed G, Heraeus Medical Ltd., Wehrheim, Germany) containing gentamicin, as a cage was not available at the time. There were no intraoperative surgical or anesthetic complications.

RESULTS

Postoperative course

One hour postoperatively, the patient received one dose of parecoxib (40 mg, Dynastat, Pfizer Ltd., Kent, UK) and one dose of hydrocortisone (100 mg, Solu-Cortef, Pfizer Hellas, Athens, Greece) intravenously (e.g., to prevent edema, due to surgical manipulations with the newly diagnosed EC). Nonetheless, at 2 h postoperatively, she developed severe swelling of the whole right side of the neck/lower face along with difficulty breathing and swallowing. She immediately received 500 mg of intravenous hydrocortisone, while the otolaryngologists performed a fiber-optic endoscopy that showed acute swelling of the EC and edema of the hypopharynx/larynx.

The emergent computed tomography scan revealed massive edema of the right lateral neck affecting the epiglottis/hypopharyngeal wall, resulting in severe airway compromise. To a lesser extent, it involved the rest of the larynx and oropharynx. There was no evidence of rupture of the EC [Figures 1 and 2].

The anesthesiologist emergently intubated the patient, and a tracheotomy performed by the otolaryngologists. The edema decreased markedly in the next 12 h, and by the 3rd postoperative day, it resolved. Her neurologic condition improved postoperatively (mJOA score: 16), and there were no surgical/medical sequelae at 3 postoperative months [Table 1, Figure 3].

DISCUSSION

Literature review

Although we searched the following databases: PubMed, PubMed Central, Scopus, Science Direct, Directory of Open Access Journals, and Google Scholar using multiple

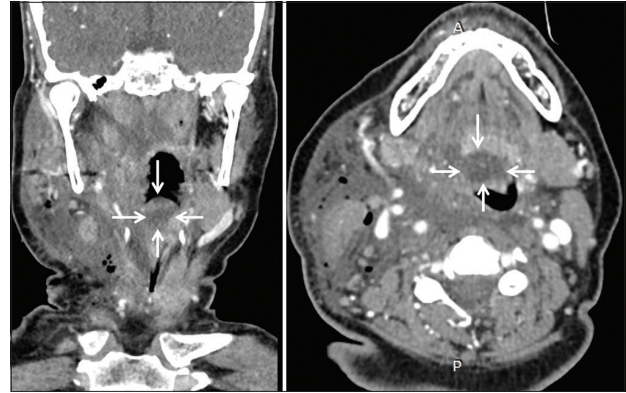


Figure 1: Coronal multiplanar reformat (left image) and axial (right image) postcontrast computed tomography images of the neck at the level of the epiglottis. There is a cystic lesion (1.8 × 1.6 × 1 cm) centered at the tip of the epiglottis, consistent with an epiglottic cyst (arrows). The right vallecula is completely obliterated, while the left vallecula is less affected. Furthermore, there is extensive edema of the soft tissues of the right lateral neck.

Table 1: Timeline of the most important information of the case report.

Timeline

- The patient suffered from moderate DCM and osteoporosis on Risedronate
- The anesthesiologist noticed an epiglottic cyst during the intubation
- The ACDF was completed without any intraoperative complications. For the fusion of the C5–C6 level, bone cement containing gentamicin was used
- ~1 h after the operation, parecoxib and hydrocortisone were administered
- ~2 h after the operation, neck swelling appeared abruptly and deteriorated rapidly
- Administration of larger doses of IV hydrocortisone was initiated.
- The neck CT scan revealed extensive edema, which affected the right side of the neck tissues and reduced the size of the upper airway critically
- There was suspicion of a pseudoaneurysm of a small branch of the external carotid artery in the neck CTA scan, but neither a pseudoaneurysm nor any active bleeding were found with the DSA
- An emergency surgical tracheotomy was performed by the otolaryngologists
- The edema decreased markedly in the next 12 h and by the 3rd postoperative day it resolved, while the patient was still on IV hydrocortisone
- The tracheostomy tube was removed on the 12th postoperative day
- 3 months after the operation, the neurologic condition of the patient associated with DCM was improved, while there were not any other residual medical sequelae

DCM: Degenerative cervical myelopathy, ACDF: Anterior cervical discectomy and fusion, CT: Computed tomography, CTA: Computed tomography angiography, DSA: Digital subtraction angiography, IV: Intravenous

key words, for example, cervical, spine, neck, surgery, angioedema, epiglottic, laryngeal, and cyst (in various combinations), we were unable to find case reports/studies comparable to the one presented.

Reintubation rate after ACSS

According to the literature, reintubation after ACSS occurs in 0.1–2.4% of patients overall and in 1.6–5.2% of cases after multilevel surgery.^[7,9] Postoperative laryngopharyngeal edema is the most common cause of airway compromise (reaching 6% in multilevel surgery) and is typically seen within 12–72 h.^[2,6,7]

Angioedema

Angioedema is one of the earliest causes of postoperative airway compromise after ACSS and typically presents

within 6–12 h. However, due to its rarity, it is often misdiagnosed [Table 2].^[4,8] Further, angioedema is one of the known side effects of both risedronate and parecoxib, and has also been associated with a history of smoking; notably, our patient was a smoker [Table 2].^[1-4,8] It is well known that corticosteroids reduce the risk of reintubation following ACSS.^[9]

EC

ECs are usually undiagnosed because patients may not report any symptoms, but they may be potentially life threatening, requiring emergent tracheostomy.^[5] The EC in our case likely became symptomatic due to the retraction/manipulation of ACDF surgery; the resultant edema then contributed/was responsible for upper airway obstruction.

Table 2: Risk factors associated with airway obstruction and/or reintubation, angioedema, and postoperative edema after ACSS.

Airway obstruction/reintubation	Angioedema	Postoperative edema
Patient factors: Male sex Age >60 years DCM Alcohol abuse Smoking Obesity Pulmonary disease Obstructive sleep apnea Bleeding disorders/anemia Congestive heart failure High comorbidity burden Dependent functional status Use of heparin or >1 anticoagulant Surgical: Multilevel surgery (≥ 3) Exposure of C2–C4 Long operative time (>5 h) Blood loss >300 ml Hematoma ACCF Combined approach (anterior and posterior) Previous ACSS Use of bone morphogenetic protein Anesthetic: Multiple intubation attempts ASA class ≥ 5 Cormack and Lehane system: Grade Ti view Institutional: No 24 h in-house surgical staff or anesthesia care Operations performed at large hospitals Nonelective surgery	Older age Smoking Physical stimuli, including neck surgery Allergens, including latex of surgical gloves History of allergies or autoimmune disease C1 inhibitor deficiency or dysfunction Cardiopulmonary disease Drugs: Bisphosphonate derivatives NSAIDs Including aspirin ACE inhibitors Strong connection) ARBs Preservatives found in drugs or fluid preparations, IV contrast material Anesthetic: ASA class ≥ 3	Patient factors: Male sex Age >65 years Smoking DCM (moderate-severe) Obesity Pulmonary disease Obstructive sleep apnea Surgical: Multilevel surgery (≥ 3) Exposure of C2–4, Long operative time (>5 h) Blood loss >300 ml Prior ACSS Intraoperative blood transfusions (>4) Anesthetic: Multiple intubation attempts

ACCF: Anterior cervical corpectomy and fusion, ACDF: Anterior cervical discectomy and fusion, ACE: Angiotensin-converting enzyme, ACSS: Anterior cervical spine surgery, ARBs: Angiotensin II receptor blockers, ASA: American Society of Anesthesiologists, DCM: Degenerative cervical myelopathy, IV: Intravenous, NSAIDs: Nonsteroidal anti-inflammatory drugs

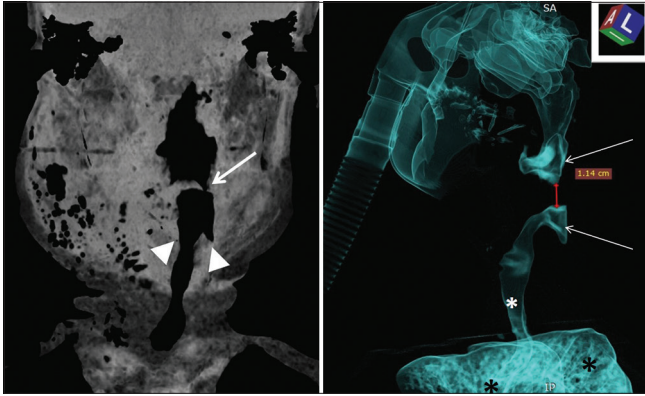


Figure 2: Computed tomography coronal minimum intensity projection image of the neck (left image) and left lateral oblique volume-rendered image reconstructed with airway algorithm (right image). These images show the compromised airway, which measures 1.14 cm in length (depicted between the red calipers in the right image) and includes just a tiny patent part left (arrow in the left image). Furthermore, in the left image, the piriform sinus is obliterated on the right side compared to the left normal side (arrowheads). In the right image, the patent airway above and below the narrowed part are shown with white arrows, the patent trachea with a white asterisk and the lungs with black asterisks.

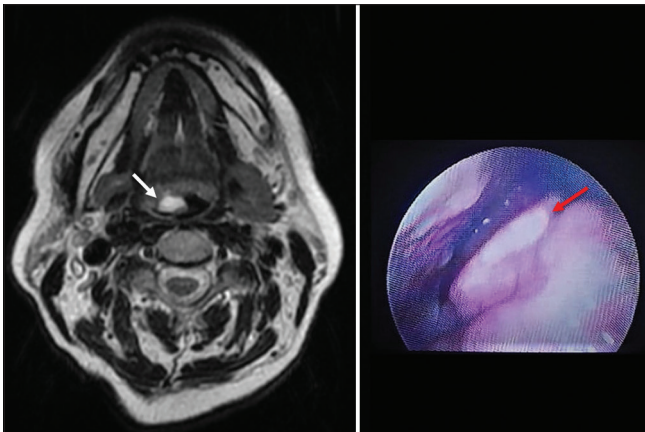


Figure 3: Left image: axial T2 fast spin echo magnetic resonance imaging image of the neck, which shows a high signal cyst next to the right side of the base of the tongue (white arrow) corresponding to the known epiglottic cyst. Right image: endoscopic view of the epiglottic cyst (red arrow) on the lingual surface of the epiglottis.

CONCLUSION

Patients with EC who need ACSS/ACDF neck surgery should either receive treatment of the cyst before an ACDF (e.g., if incidentally found on intubation, surgery should be cancelled and the cyst dealt with) or be closely

monitored postoperatively for signs of increased airway obstruction.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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