Case Report

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

Miltiadis Georgiopoulos¹, Dimitrios Papadakos¹, Pantelis Kraniotis², Spyridon Lygeros³, Vasilios Margaritis³, Dimitrios Karnabatidis², Georgios Gatzounis¹

Departments of ¹Neurosurgery, ²Radiology, ³Otolaryngology, University Hospital of Patras, Patras, Achaia, Greece.

E-mail: *Miltiadis Georgiopoulos - mgeorgiop@gmail.com; Dimitrios Papadakos - dimitriospapadakos@outlook.com; Pantelis Kraniotis - pantelis.kraniotis@gmail.com; Spyridon Lygeros - slygeros@gmail.com; Vasilios Margaritis - vmargaritisent@gmail.com; Dimitrios Karnabatidis - karnaby@upatras.gr; Georgios Gatzounis - g_gatzounis@web.de

*Corresponding author: Miltiadis Georgiopoulos, Neurosurgeon, Department of Neurosurgery, University Hospital of Patras, Patras, Achaia, Greece.

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.
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

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

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Actonel (Risedronate) Dosing, Indications, Interactions, Adverse Effects, and More, Medscape; 2019. Available from: https://www.reference.medscape.com/drug/actonel-risedronate-342835. [Last accessed on 2019 Nov 27].
2. Debkowski MP, Butterworth JF, Moore JE, Kang S, Appelbaum EN, Zuelzer WA. Acute post-operative airway complications following anterior cervical spine surgery and the role for cricothyrotomy. J Spine Surg 2019;5:142-54.
3. Dynastat 40mg Powder for Solution for Injection, EMC; 2019. Available from: https://www.medicines.org.uk/emc/product/1606/smpc. [Last accessed on 2019 Nov 30].
4. Hannoondi F, Sabbagh H. ACE inhibitor-induced angioedema following cervical spine surgery. Case Rep Cardiol 2017;2017:4268962.
5. Kariya N, Nishi S, Minami W, Funao T, Mori M, Nishikawa K, et al. Airway problems related to laryngeal mask airway use associated with an undiagnosed epiglottic cyst. Anaesth Intensive Care 2004;32:268-70.
6. Palumbo MA, Aidlen JP, Daniels AH, Bianco A, Caiati JM. Airway compromise due to laryngopharyngeal edema after anterior cervical spine surgery. J Clin Anesth 2013;25:66-72.
7. Sagi HC, Beutler W, Carroll E, Connolly PJ. Airway complications associated with surgery on the anterior cervical spine. Spine (Phila Pa 1976) 2002;27:949-53.
8. Umerani MS, Alzahrani K, Mostafa GA. Hereditary angioedema: A rare presentation after anterior cervical disectomy and fusion. Asian J Neurosurg 2015;10:253-5.
9. Wilson LA, Zubizarreta N, Bekeris J, Poeran J, Liu J, Fiasconaro M, et al. Risk factors for reintubation after anterior cervical disectomy and fusion surgery: Evaluation of three observational data sets. Can J Anaesth 2020;67:42-56.

How to cite this article: Georgiopoulos M, Papadakos D, Kraniotis P, Lygeros S, Margaritis V, Karnabatidis D, et al. Neck angioedema after anterior cervical disectomy and fusion with coexistent epiglottic cyst. Surg Neurol Int 2020;11:459.