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CASE REPORT

Neisseria meningitidis sepsis in a patient with acute epiglottitis and respiratory failure

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SUMMARY

A 78-year-old female patient presented to our emergency department with a 5-day history of throat pain, hoarseness and a progressive and dolorous submandibular swelling. Due to non-conclusive clinical examination and the stable but visibly affected patient, we performed a neck CT scan with intravenous contrast, which showed the thumbprint sign typical for acute epiglottitis. Within minutes, the patient's condition deteriorated and the patient was close to respiratory exhaustion. As intubation was severely complicated by massive swelling of the supraglottic soft tissue, the patient went into hypoxaemia and eventually cardiac arrest. After initiating reanimation protocol, eventually the airway was secured and Return of spontaneous circulation (ROSC) achieved after around 5 min. The initially sampled blood cultures revealed *Neisseria meningitidis* bacteraemia and the patient was successfully treated accordingly. In patients with swelling of the upper airway, rapid clinical deterioration is possible. Diagnostics should not delay therapy, including administration of empiric antibiotics, steroids and intubation.

BACKGROUND

Whenever evidence for swelling of the upper airway exists, clinicians should be aware of possibly rapid clinical deterioration. Intubation should be evaluated early aiming at avoiding complications such as severe hypoxaemia. *Neisseria meningitidis* is a rare cause of epiglottitis/supraglottitis, usually responsive to most recommended antibiotic regimens. Steroid treatment is controversially discussed in the treatment of epiglottitis, but can be taken into consideration for the clinical presentation of bacterial sepsis in the sense of sepsis bundle treatment.

CASE PRESENTATION

A 78-year-old woman with a known history of hypertensive cardiomyopathy and paroxysmal benign positional vertigo presented to the emergency department with a 5-day history of throat pain and hoarseness, as well as a progressive and dolorous swelling in the submandibular area. She further suffered from progressive coughing and dysphagia for 2 days and ultimately breathing difficulties. The body temperature measured at the emergency department was 37.7 °C (100°F). Haemodynamically, the patient was stable with a slightly hypertensive blood pressure (150/80 mm Hg) and tachycardia (118/min) as well as a room-air oxygen

saturation of above 90%. Blood analysis showed leucocytosis of 17.2 g/L and elevated C-reactive Protein (CRP) of 125 mg/L.

INVESTIGATIONS

The initial clinical examination confirmed the submandibular swelling; the enoral examination was severely limited by a restricted mouth opening. Otolaryngoscopic examination was impossible due to cerumen occlusion. Meningism was ruled out. Due to a non-conclusive clinical examination and the, although stable, visibly affected patient, a neck CT scan performed with intravenous contrast, which showed the thumbprint sign typical for acute epiglottitis as well as extensive supraglottic and subglottic swelling resulting in stenosis (figure 1).

TREATMENT

After returning from the CT scan, the patient's condition deteriorated within minutes. The oxygen saturation dropped to about 70% and did not recover even with 10 L of O₂ administered via reservoir mask. Respiratory exhaustion was imminent. While intravenous Solu-Medrol and Co-Amoxicillin were administered, intubation was initiated, which proved to be extremely difficult due to swelling of the supraglottic soft tissue, although video-guided GlideScope was used. Consequently, the patient went into hypoxaemia and eventually into cardiac arrest. Reanimation protocol was immediately initiated. After even direct tracheal airway access failed, another attempt was made to orally intubate the patient, this time with success. After around 5 min of mechanical reanimation and a total of 2 mg intravenous adrenalin, ROSC was achieved. During the course of hospitalisation, the initially sampled blood cultures revealed *N. meningitidis* bacteraemia. The antibiotic regimen was changed from Co-Amoxicillin 2.2 g every 8 hours to ceftriaxone 2 g every 12 hours. Intravenous steroid application was continued.

OUTCOME AND FOLLOW-UP

The patient remained intubated for a week when a follow-up CT scan showed decreased swelling of the epiglottis while swelling of the larynx and subglottic area remained grossly unchanged. The swelling was furthermore monitored by regular direct laryngoscopy. In time, the decision was made to perform a surgical tracheotomy for adequate long-term ventilation.



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Figure 1 CT scan showing thumbprint sign (arrow) in a patient with acute epiglottitis.

After 11 days, the respiratory situation had improved enough to cease mechanical ventilation and eventually tracheostomy was reversed. The patient was discharged into a neurorehabilitation facility to properly treat the remaining neurological impairments caused by the initial hypoxic event.

About 6 months after discharge, the patient was interviewed via telephone. All temporary neurological impairments were completely regressive over time. The patient though reported that delayed, she started suffering from panic attacks and states of anxiety regarding the incident.

DISCUSSION

Typically, patients with obstruction of the upper airway present with clinical features of stridor and respiratory distress. The differential diagnoses include infections, foreign-body ingestion, trauma, anaphylaxis and spasms. However, even without classical warning symptoms, swelling in the head-and-neck area should always trigger further investigation to exclude acute impairment of airway. As shown in this case, even oligosymptomatic patients with swelling of the neck area can deteriorate unexpectedly fast and hard. We therefore recommend a liberal use of imaging via CT scan, when airway obstruction might be suspected even without obvious reason, to secure diagnosis and prompt timely treatment. Besides the diagnostic pathway, intubation should be evaluated early, especially if there are signs of respiratory decline like a drop in oxygen saturation, visibly strained breathing or an increase in respiratory rate. In our case, the patient was initially in a stable clinical state; we therefore chose to push on with diagnostic measures and not to choose the therapeutic pathway of early intubation. In retrospect, early extended airway management might have prevented the hypoxic cardiac arrest. Since initially there were no signs for respiratory distress, we believe that at the time, the chosen approach was nevertheless correct. As intubation is mostly severely complicated by a swelling of the upper airway, appropriate preparations have to be made. If possible, intubation should be performed by experienced medical professionals with all the necessary auxiliaries at hand (GlideScope, fibre optics etc.), and surgical back up should be in place.

In our case, intubation had to be performed as an emergency procedure and only a GlideScope could be arranged, which unfortunately turned out not to suffice in this case, leading to a prolonged intubation process, hypoxaemia and subsequently cardiac arrest. Concerning extubation procedure, some rules apply: Given the circumstances and the possible complications of a premature extubation, extubation should not be rushed. In our case, we performed tracheostomy for long-term ventilation, as the local situation did not significantly improve in the first couple of days. The swelling was monitored by follow-up CT scans and fibre-optic examination. Once we were content with the development of the swelling, the infection was under control and ventilation went smoothly, we were able to taper mechanical ventilation and eventually reverse tracheostomy. When epiglottitis is suspected, early antibiotic therapy is imperative. It is generally recommended to start an empiric regimen after sampling and eventually adapt the regimen according to microbiological findings. The recommended empiric regimen is ceftriaxone for 7–10 days, which is able to treat the most likely organisms (*Haemophilus influenzae*, *Streptococcus pneumoniae*, Group A *Streptococcus* and *Staphylococcus aureus*). Depending on local circumstance, guidelines may include a standard use of vancomycin against MRSA. In our case, the cause of the airway obstruction was caused by epiglottitis/supraglottitis due to infection with *N. meningitidis*. Although several similar cases have been published previously, *N. meningitidis* remains a rare cause of such a clinical manifestation. Steroid treatment is controversially discussed in the treatment of isolated epiglottitis, but can be taken into consideration for the clinical presentation of sepsis in the sense of sepsis bundle treatment. In this case, we decided to administer prednisone 0.5 mg/kg for 2 weeks.

Learning points

- ▶ Whenever evidence for swelling of the upper airway exists, clinicians should be aware of possibly rapid clinical deterioration. Intubation should be evaluated early, aiming at avoiding complications such as severe hypoxaemia.
- ▶ If in doubt, rare causes of airway swelling cannot be dismissed and should always be part of the differential diagnoses.
- ▶ Early administration of antibiotic therapy when suspecting an infectious origin can save a patient's life and should not be delayed.

Contributors TZ and CC were both involved in the emergency department treatment of the patient, TZ as an intern and CC as a senior physician. TZ took the initiative for preparing the case report and wrote the draft of the manuscript. TZ conducted the telephone follow-up with the patient and acquired the informed consent. CC supported TZ in the draft conception and design, corrected the manuscript and adapted it to the journal's requirements. TZ approved of the final version of the manuscript.

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