CASE REPORT

Laryngopyocoele in an adult female mimicking an infected branchial cyst

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SUMMARY

The incidence of laryngopyocoele is very unusual and only 41 cases have been documented in the world literature. Although hoarseness and stridor are the predominant symptoms associated with laryngopyocoele, patient can present with a recurrent neck abscess misleading to an infected branchial cyst. Complete excision of the cyst can be effectively achieved by the transcervical approach, especially in case of a large lateral cyst in a stable patient.

BACKGROUND

When the neck of an internal laryngocoele gets obstructed, the mucous accumulates in a sacculus called as laryngomucocoele. Very rarely it gets infected to form a pus-filled cavity, called as laryngopyocoele. The occurrence of laryngopyocoele is very unusual and only 41 cases have been documented in the world literature.¹⁻⁵ Clinical presentation depends on the extension of the lesion into the endolarynx including foreign body sensation in the throat, hoarseness, dysphagia and stridor. Very rarely patients can have a neck swelling above the level of thyroid lamina and often gets confused with a branchial cyst, especially when there is a history of the recurrent infection. Flexible laryngoscopy and contrast-enhanced CT scan are the mainstay investigations performed to look for the extension of the lesion and to rule out the airway obstruction. Diagnosis mainly depends on the clinical suspicion of the disease along with the accurate radiological correlation. Although various surgical approaches have been recommended for the excision of the laryngopyocoele, the transcervical approach is a safe and effective surgical method for the complete excision of the sac with preservation of the laryngeal mucosa. Patients need close follow-up for endoscopic evaluation of the larynx to look for the recurrence of the disease. We present an atypical case of laryngopyocoele in an adult woman which was misleading clinically to an infected branchial cyst, successfully managed through a transcervical excision.

CASE PRESENTATION

A 40-year-old female patient presented to the outpatient department with intermittent pain and swelling over the right side of the neck just above the level of the thyroid lamina for 5 years and fever for the 7 days. The general physical examination was found to be normal. She had a history of similar episodes in the past 5 years at the same site for which she had undergone repeated incision

and drainage in different peripheral hospitals. There was no history of dysphagia or hoarseness to the patient. She had no history of vocal abuse or laryngeal trauma in the past. On examination, a round to oval swelling of 3cm×3 cm was found above thyroid lamina on the right side of the neck, which was tender, mobile and the swelling did not move with deglutition. Keeping in mind a neck abscess, broad spectrum intravenous antibiotics (amoxicillin/clavulanate 40 mg/kg/day and metronidazole 1500 mg/day) were started and continued for 7 days.

INVESTIGATIONS

Flexible fibre-optic laryngoscopy revealed a smooth mucosal bulge found just over the false cord on the right side partially obstructing the laryngeal inlet. Transcervical aspiration of the swelling revealed the presence of mucopus and culture sensitivity of the aspirate was found negative for the growth of the microorganism. A contrast-enhanced CT scan revealed a well-defined hypodense mass found at the lateral aspect of the right false vocal extending superiorly till the hyoid bone with minimal obstruction of the airway (figure 1).

DIFFERENTIAL DIAGNOSIS

Based on the clinical and radiological findings, the patient was diagnosed as laryngopyocoele.

Figure 1 Contrast-enhanced CT scan revealed a well-defined hypodense mass found at the lateral aspect of the right false vocal extending superiorly till the hyoid bone with minimal obstruction of the airway.

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Figure 2 The cyst was completely delineated from the surrounding structures and was excised with gentle dissection, preserving the laryngeal mucosa.

TREATMENT

After the written informed consent, the patient was planned for excision of the cyst under general anaesthesia. Orotracheal intubation was carried out with the help of flexible bronchoscope. A transverse skin incision was made in the skin crease over the thyrohyoid membrane, from the anterior border of the sternocleidomastoid to the midline of the neck (figure 1). The subplatysmal flap was elevated exposing the submandibular gland superiorly, omohyoid muscle anteriorly and sternocleidomastoid muscle posteriorly. The cyst was exposed all around the soft tissue along with the carotid sheath with the blunt dissection. The superior laryngeal nerve and vessels were identified and later needed ligation intraoperatively for a better exposure. The cyst was found extending inferomedially along the thyrohyoid membrane into the right paraglottic space. It

was completely released from the surrounding structures and was excised with gentle dissection preserving the laryngeal mucosa (figure 2) which was confirmed intraoperatively by rigid laryngoscopy. Histopathology revealed a benign cyst in direct continuity with the airway tract, lined predominantly by flattened cuboidal lining epithelium and respiratory lining at the zone of transition to the native airway. The cyst wall was composed of fibrocollagenous tissue and did not contain any submucous gland unlike the part of native airway (figure 3a and b). The patient was put on intravenous steroid and broad spectrum antibiotics for 1 week in the postoperative period. Ryle's tube feeding was continued for 7 days in the postoperative period to avoid the stretch injury to the laryngeal mucosa. There were no significant intraoperative and postoperative complications noted in the patient.

OUTCOME AND FOLLOW-UP

She is on regular follow-up in the voice clinic for the past 6 months with normal speech without the recurrence of the disease.

DISCUSSION

Laryngocoele is the herniation of the air-filled sac from the laryngeal ventricle communicating with the laryngeal lumen. Its incidence is approximately one per 2.5 million population per year in UK.⁴ Sometimes the neck of the laryngocoele gets obstructed producing a closed mucous-filled cavity called as laryngomucocoele and on secondary infection, laryngopyocoele is formed although later is very rare.^{2 6 7} Till date, only 41 cases of laryngopyocoele have been documented in the world literature.¹⁻⁵ Although various predisposing factors have been proposed, people with professional trumpet playing, glass blowing, singing, straining at passing of the stools, weight lifting and malignancy of the larynx are more prone for the disease.⁸⁻¹⁰ Males (M: F, 5:1) are predominantly affected by with the disease and the most common age is the sixth decade of life^{2 4} unlike in the present case where the patient was a 40-year-old woman. Symptoms vary from the foreign body sensation in the throat to the frank stridor depending on the extension of the cyst into the laryngeal lumen and features of sepsis may be detected due to the secondary infection.^{6 11} Unlike in the present case, there were no definitive laryngeal

Figure 3 (A) Photomicrograph showing a benign cyst (red arrow) in direct continuity with the native airway (black arrow), the latter containing submucous glands whereas the former having a fibrocollagenous tissue (H&E; 20×). (B) The cyst had flat cuboidal lining and fibrocollagenous cyst wall (H&E; 100×).

symptoms, rather the presence of the neck swelling with fever had misled the diagnosis to an infected branchial cyst. Again, flexible laryngoscopy and contrast CT scan had no significant abnormality except mild lateral bulge over the false vocal cord. Keeping in mind an infected branchial cyst which could have a medial extension, the patient had undergone repeated abscess drainage in different peripheral hospitals. The occurrence of the laryngopyocoele is very uncommon in the clinical practice and laryngopyocoele is still very rare (8% cases) and it is mostly associated with the combined laryngocoele.⁴ The common organisms isolated from the pyocoele are the *Escherichia coli*, haemolytic *Streptococcus B*, *Staphylococcus aureus* and *Pseudomonas aeruginosa*. In contrast, no microbial growth was isolated in our case which could be due to the previous antibiotic treatment.³ Contrast-enhanced CT scan is the mainstay radiological test advised in each case of laryngopyocoele which not only defines the content but also assesses the relationship with the inner laryngeal structure and most importantly to rule out the coexisting carcinoma.^{12–14} It is always necessary to rule out the parapharyngeal cyst and tumours which may exist in the similar location producing similar clinical and radiological features.^{2 13 15} The most life-threatening complication is the rupture of the cyst into the respiratory passage causing damage to the lung tissue but fortunately, we did not encounter any such complications which could be due to the prompt medical management and meticulous follow-up with serial flexible endoscopy. Although the recommended

treatment for laryngopyocoele is endoscopic drainage of the cyst,^{13 16} we preferred for an external approach due to the extensive lateral extension of the cyst and there was no acute obstruction of the airway. Although recurrence is very uncommon after a complete excision as observed in our case, patients need a close follow-up with flexible laryngoscopy to rule out the laryngeal oedema and superior laryngeal nerve injury which may not be prominent in the immediate postoperative period.

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Learning points

- The incidence of laryngopyocoele is rare in the clinical practice.
- Patients can present with recurrent lateral neck swelling which may mislead the diagnosis to an infected branchial cyst.
- Although flexible laryngoscopy and CT scan of the neck are thought to be the mainstay of diagnosis for laryngopyocoele, it is often a challenge to have a preoperative confirmation of the diagnosis.
- The external cervical approach is an ideal surgical technique for complete excision of the cyst without any significant complication.

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