Intratracheal myiasis followed by tracheal-esophageal fistula: report of a rare case and literature review

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Abstract

Background: To enhance awareness of the clinical features and prevention of endotracheal myiasis.

Case presentation: A case of intratracheal myiasis is reported. A 61-year-old male patient with a history of laryngectomy was admitted to hospital due to tracheostomal hemorrhage of 3 h duration. Intratracheal myiasis was confirmed by bronchoscopy, and the patient underwent bronchoscopic intervention, which was complicated by a tracheal-esophageal fistula and resolved by endotracheal stenting. Twenty months after stent placement, the fistula had not healed.

Conclusion: Intratracheal myiasis has serious complications and is difficult to treat. For post-tracheostomy patients, healthcare providers and caregivers should pay attention to the care and monitoring of wounds and maintenance of a tidy, clean living environment to prevent intratracheal myiasis.

Background

Myiasis is the invasion of dipterous insect larvae into tissues. The larvae can invade human skin, gangrenous tissue, and natural cavities. Myiasis of the trachea is very rare with only a few cases previously reported. Furthermore, intratracheal myiasis followed by a tracheal-esophageal fistula has not, to the best of our knowledge, been reported previously [1–5].

Case presentation

A 61-year-old man from Shenzhen presented with tracheostomal hemorrhage of 3 h duration. One year previously, the patient had been diagnosed with laryngeal squamous cell carcinoma by electronic laryngoscopy, and had undergone partial laryngectomy and tracheotomy under general anesthesia. Following these procedures, he was discharged and recovered well. The tracheotomy was maintained after discharge. The patient and his family cleaned the tracheal cannula themselves which may have resulted in inadequate care. The patient visited our hospital due to tracheostomal hemorrhage of 3 h duration.

Physical examination at admission showed peri-tracheostomal redness and correct positioning of the tracheal cannula. Bloody secretions were seen in the dressing. Electronic laryngoscopy showed postoperative changes in the glottic area, with scar formation; the airway was patent. The results of a complete blood count were 9.58 × 10⁹/L white blood cells, 74.4% neutrophils, and 1.0% eosinophils. After admission, the tracheal cannula was removed, and the surface of the cannula was dirty. The stomal tissue was red and swollen. On the day after removal of the tracheal cannula, the patient coughed out maggot larvae through the tracheostomy, and several larvae were found in the peri-stomal area.

Computed tomography of the chest showed a hyperdense mass involving the trachea (Fig. 1a, arrow). The peri-esophageal and peri-tracheal fat spaces were blurred (Fig. 1b, arrow). Bronchoscopy confirmed the presence of live larvae in the left lower tracheal segment, with fibrous granulation and tracheal stenosis. Furthermore, heavy granulation was found in the upper and middle thirds of the trachea. A tracheosthenosis with a smallest diameter of about 0.6 cm was noted (Fig. 1c); it was resolved using electrocautery to burn the larvae (Fig. 1d),
and by making linear and circular incisions in the larval burrow and removing the larvae using cryotherapy and forceps (Fig. 1e) and clearing the basal granulation. This procedure resulted in improvement of the tracheal stenosis. Bronchoscopy showed that the left main bronchus and their upper and lower segments were patent. One week later, follow-up fiberoptic bronchoscopy showed granulation and membranous necrosis in the upper and middle trachea. The larval burrow was absent from the left lower trachea, but the site was covered with granulation and necrotic tissue. The trachea was restored to three-quarters of its normal size. We removed the necrotic tissue and granulation using forceps and freezing (Fig. 1f).

After the procedure, the patient experienced cough after taking food. Follow-up bronchoscopy showed a tracheoesophageal fistula of about 1.5 cm (Fig. 1g and h) in the lower trachea, 2 cm above the carina, so an indwelling gastric tube was placed which was visible on bronchoscopy passing through the tracheoesophageal fistula. The thoracic surgeon recommended conservative medical treatment. However, the patient continued to cough up food so bronchoscopy was performed 1 month later which showed that the tracheal-esophageal fistula had not properly healed. Next, a Y-type tracheal silicone stent was placed. The patient’s condition has been stable since that time. Follow-up gastroscopy performed 12 months after the procedure showed that the tracheal-esophageal fistula had not healed and covered the Y-type tracheal silicone stent (Fig. 1i), for which we suggested surgical repair.

**Discussion and conclusions**

In this case, intratracheal myiasis resulted from invasion of the trachea by dipterous larvae. Few cases of bronchial myiasis have been reported [1]. A search of PubMed using the keywords ‘human intratracheal myiasis’ or ‘tracheopulmonary myiasis,’ or ‘bronchial myiasis’ revealed five reports of five cases published up to July 2018 (Table 1). The cases were sporadic, without age or sex predilection, and all occurred in tropical and subtropical regions where flies are prevalent following endotracheal intubation or tracheotomy [3–7].
In conclusion, tracheal-esophageal fistula is a serious complication of endotracheal myiasis and is difficult to treat. Tracheal stenting can improve the resulting reflux but cannot treat the condition. For post-tracheostomy patients, healthcare providers and caregivers should pay attention to the care and monitoring of wounds and maintenance of a tidy, clean living environment to prevent intratracheal myiasis.

**Abbreviations**
cm: centimetre; g: gramma; L: Liter

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**Authors’ contributions**
Data acquisition, analysis and interpretation: PX, WDH, CZ, WSD. Writing the article or substantial involvement in its revision: WDH, PX. All authors have read and approved the manuscript, and ensure that this is the case.

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**Table 1** Reported cases of intratracheal myiasis

| Authors and year | Patient sex/age | Country | Concomitant diseases | Involved areas | Confirmative evidence | Clinical manifestations |
|------------------|-----------------|---------|----------------------|----------------|-----------------------|------------------------|
| Cornet et al. 2002 | Female/60 years | US      | No                   | Intratracheal  | Cough out             | Cough, haemoptysis     |
| Cecchini et al. 2012 | Male/47 years | France  | Diabetes, septic shock | Left main bronchus | Bronchoscopy    | No                     |
| Arindom et al. 2015 | Male/13 years | Oman    | No                   | Right upper bronchus | Bronchoscopy | Cough, dyspnoea        |
| Ahadizadeh et al. 2015 | Male/53 years | US      | Cardiac arrest       | Intratracheal  | Inside tracheal catheter | No                  |
| Ince et al. 2015   | Female/8 months | Turkey  | N/A                  | Intratracheal  | N/A                   | N/A                   |

N/A not accessible
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Availability of data and materials
The materials described in the manuscript are available from the corresponding author on reasonable request.

Ethics approval and consent to participate
The study was approved by the Ethics Committee of Peking university Shenzhen Hospital. Informed written consent was obtained from the patient for publication of this case report and accompanying images.

Consent for publication
All presentations of case reports have consent to publish. Informed consent for publication of images or other personal or clinical details was obtained from the patient.

Competing interests
The authors declare that they have no competing interests.

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