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Invasive tracheal mucormycosis complicated by myiasis following tracheostomy in a diabetic patient: A case report

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ABSTRACT

Invasive tracheal mucormycosis is a rare but fatal opportunistic infection, particularly common in immunocompromised patients. This case report describes a 23-year-old female diabetic patient who developed invasive tracheal mucormycosis following a tracheostomy. The mucormycosis infection was brought under control after antifungal therapy and surgical debridement; however, due to multiple contributing factors, the patient subsequently developed myiasis. This case highlights the importance of early diagnosis, aggressive treatment, and proper tracheostomy management in preventing severe complications.

1. Introduction

Tracheal mucormycosis is a rare but highly destructive fungal infection that primarily occurs in immunocompromised individuals or patients with diabetes [1]. This disease exhibits strong invasiveness, leading to tracheal tissue necrosis, airway collapse, and potentially fatal massive hemorrhage, posing significant clinical challenges and carrying a high mortality rate [2]. Currently, surgery combined with antifungal therapy is considered the first-line treatment for invasive tracheal mucormycosis. However, research on postoperative complications in such patients remains limited. This article reports a rare case of postoperative tracheal mucormycosis complicated by myiasis, detailing its clinical presentation, diagnostic process, and treatment course. Additionally, it explores the possible infection mechanisms and associated risk factors, aiming to provide insights for clinical management and the prevention of complications.

2. Case presentation

A 24-year-old female patient presented to an external hospital with fever, hoarseness, and progressive dyspnea. A tracheostomy was performed on day 0. Comprehensive diagnostic tests were performed, including sputum culture, sputum smear, acid-fast bacilli staining, and SARS-CoV-2 nucleic acid testing, all of which returned negative results. Empirical anti-infective therapy was initiated with cefoperazone sodium and sulbactam sodium (3 g per dose, administered intravenously every 12 hours for 5 days), along with correction of metabolic acidosis and fluid resuscitation. Despite these interventions, the patient continued to have persistent fever, and her hoarseness did not improve postoperatively. She was transferred to our hospital on day 5 for further treatment.The patient had a 6-year history of type 1 diabetes mellitus with poor glycemic control (HbA1c 10.1 %).

Laboratory tests at admission indicated significant metabolic disturbances (random blood glucose: 22.19 mmol/L, β-hydroxybutyrate: 2.36 mmol/L) and systemic inflammatory activation (WBC: 10.26 × 10^9/L, CRP: 72.39 mg/L), along with hypoalbuminemia (28 g/L) (Day 5).Contrast-enhanced neck CT (Day 5) revealed a peritracheal abscess and neck cavity formation (Fig. 1a), while chest CT showed bilateral pulmonary infiltrates, predominantly in the right lung (Fig. 1b). Renal function, electrolyte levels, complete blood count, sputum culture, SARS-CoV-2 nucleic acid test, sputum smear, Mycobacterium tuberculosis DNA, and CD panel results showed no significant abnormalities. Upon admission, the patient was immediately started on antimicrobial therapy with meropenem (1 g per dose, administered intravenously every 8 hours for a total of 4 days). Concurrently, comprehensive supportive treatment was initiated, including mechanical ventilation, intensive insulin therapy for glycemic control, fluid resuscitation, and correction of acidosis. However, the patient continued to experience recurrent fever, with temperatures ranging from 37.8 °C to 38.5 °C.

On Day 6, bronchoscopy detected a suspected tracheal cartilage ring rupture (Fig. 2a) and scattered white plaques in the bronchi (Fig. 2b). On the same day, the patient underwent neck abscess incision and drainage.

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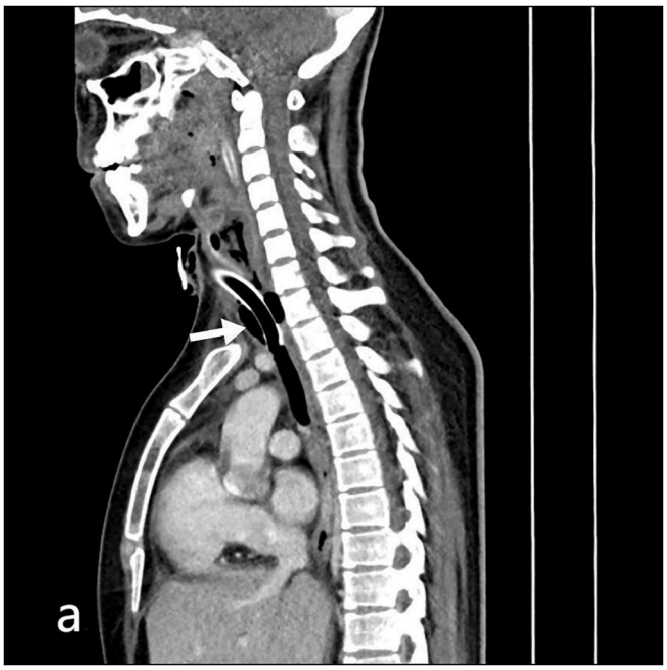

Fig. 1a. A CT scan of the neck showing a cavity formation is observed adjacent to the trachea in the neck, containing a large amount of gas and fluid. Surrounding tissues exhibit enhancement (white arrow).

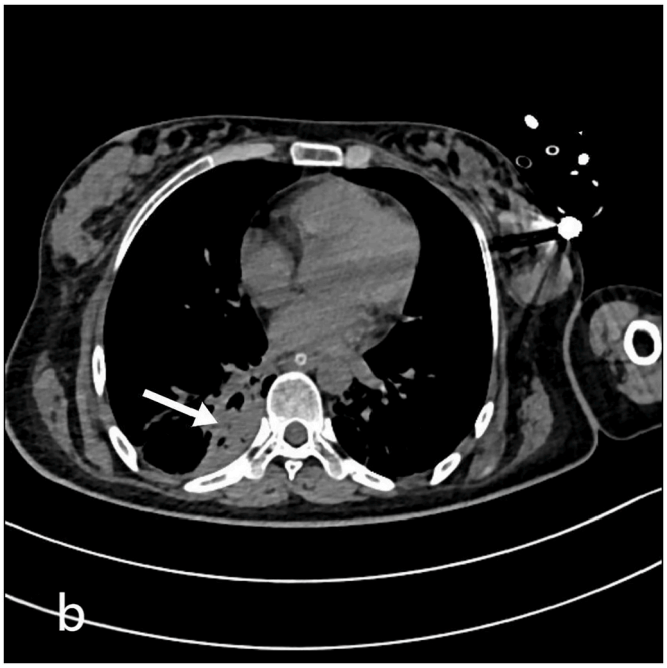

Fig. 1b. Inflammatory infiltration is seen in both lungs, with partial consolidation in the lower lobes, more pronounced in the right lung (white arrow).

Intraoperatively, extensive necrotic tissue was observed around the trachea. Due to the family's refusal of partial tracheal resection, only debridement and drainage were performed.On Day 9, pathological examination confirmed invasive tracheal mucormycosis (HE staining showed broad, non-septate hyphae(Fig. 3), with positive GMS/PAS staining.). Concurrently, no pathogenic organisms were identified in the culture of purulent secretions. As a result, meropenem—previously administered for a total of 4 days—was discontinued, and antifungal therapy with liposomal amphotericin B was initiated (5 mg/kg/day via

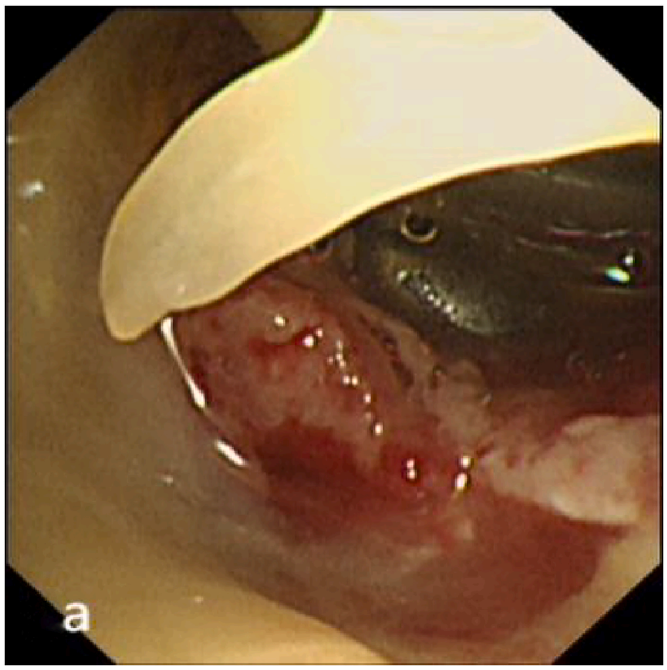

Fig. 2a. Discontinuity of the tracheal mucosa is noted at the proximal end of the tracheostomy tube, suggesting tracheal cartilage ring rupture.

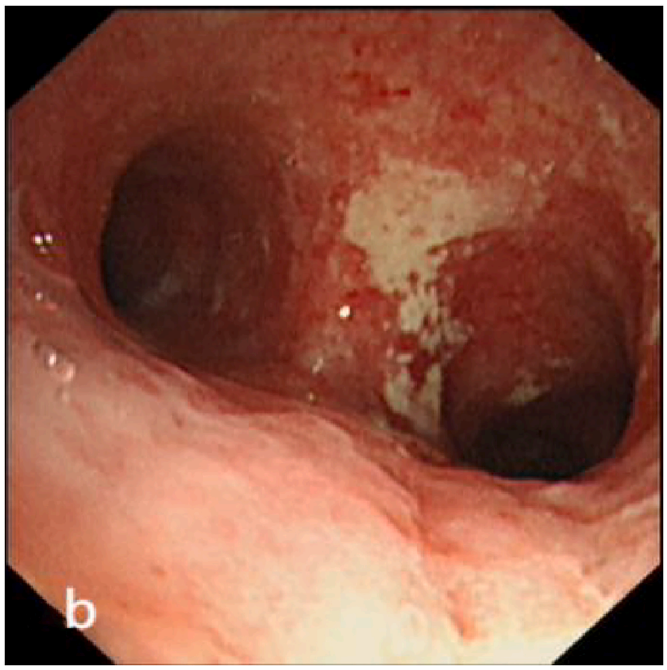

Fig. 2b. Scattered white lesions are visible inside the bronchi.

intravenous infusion, combined with 5 mg nebulized inhalation every 12 hours).

After six days of treatment (Day 15), the patient's condition worsened. Follow-up imaging revealed increased neck emphysema and fluid accumulation (Fig. 4a), along with worsening pulmonary infiltrates and right lung cavitation (Fig. 4b).As a result, a second surgical exploration was performed on Day 15. Intraoperatively, tracheal rupture with extensive necrosis was identified, leading to necrotic tissue resection and tracheostomy. Following discussion with the patient's family on the same day, the antifungal regimen was adjusted to intravenous

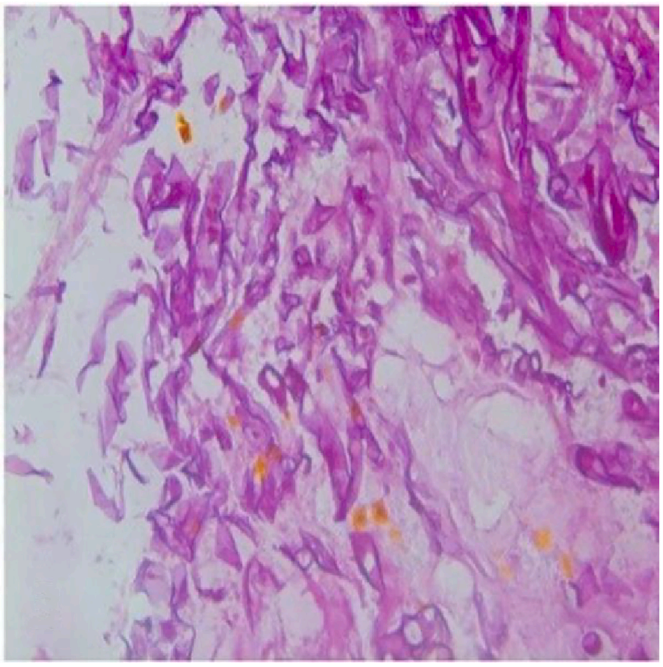

Fig. 3. (HE staining): Broad, non-septate hyphae are observed, consistent with Mucorales infection.

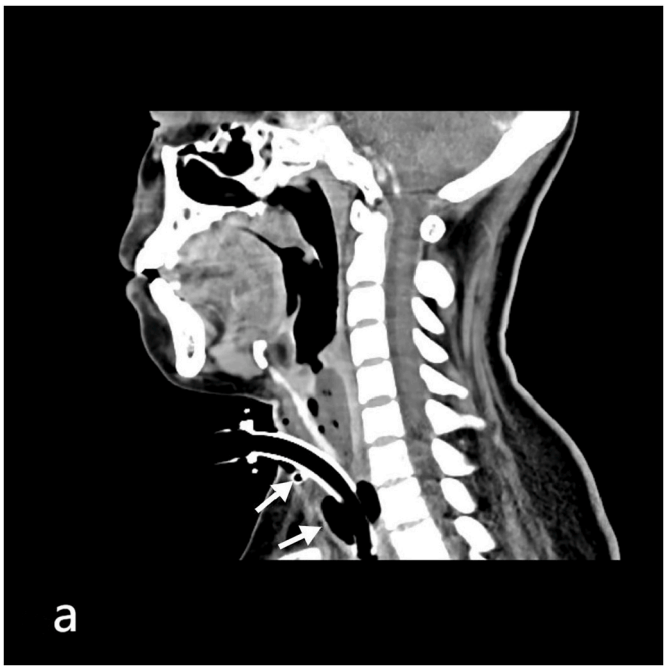

Fig. 4a. The extent of gas and fluid accumulation in the neck has increased compared to previous observations (white arrow).

isavuconazonium sulfate: a loading dose of 200 mg every 8 hours for 2 days, followed by a maintenance dose of 200 mg once daily.On Day 20, the patient showed significant symptom relief and was discharged at the family's request. The tracheostomy tube was retained to maintain airway patency, and she was instructed to continue antifungal consolidation therapy starting from Day 21 with oral isavuconazole capsules (200 mg once daily). During the post-discharge period, regular follow-ups were recommended to monitor infection control, airway healing progress, and potential adverse effects of antifungal treatment.On Day 33, follow-up neck CT showed significant resolution of the lesions

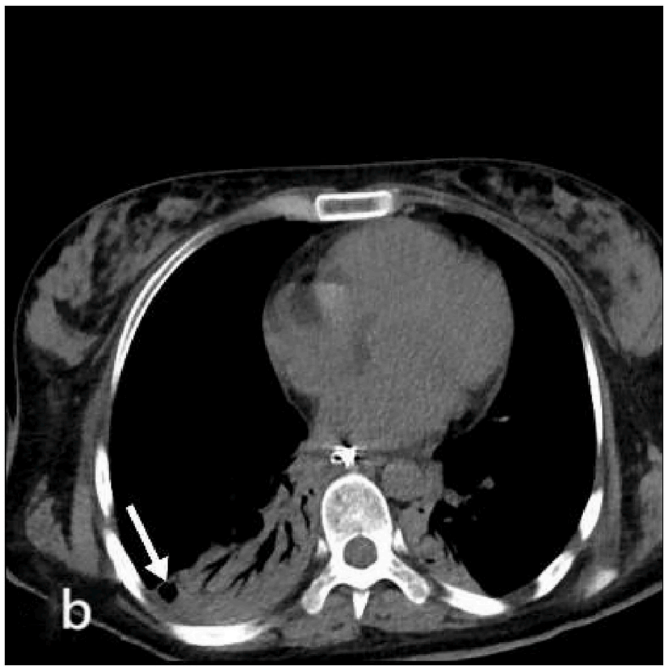

Fig. 4b. Progressive pulmonary infiltration in both lower lobes, with areas of consolidation and new cavitation in the right lower lung (white arrow).

compared to previous imaging, though residual cavities and a small amount of fluid remained (Fig. 5a). Pulmonary imaging revealed residual fibrotic foci (Fig. 5b).The patient continued oral antifungal therapy for another two weeks(total duration of isavuconazole therapy: 33 days, including 6 days of intravenous isavuconazonium sulfate and 27 days of oral isavuconazole capsules)and underwent regular tracheostomy tube replacement and cleaning.

However, on Day 58, the patient was readmitted due to dyspnea, fever (38.0 °C), and tracheostomy site ulceration. Upon further inquiry,

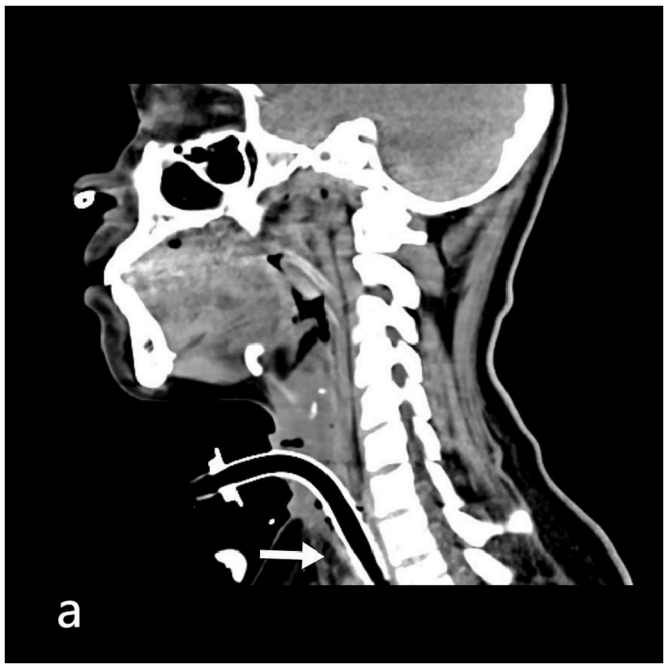

Fig. 5a. The neck lesion appeared more localized compared to previous observations; however, residual cavitation and small amounts of fluid were still present (white arrow).

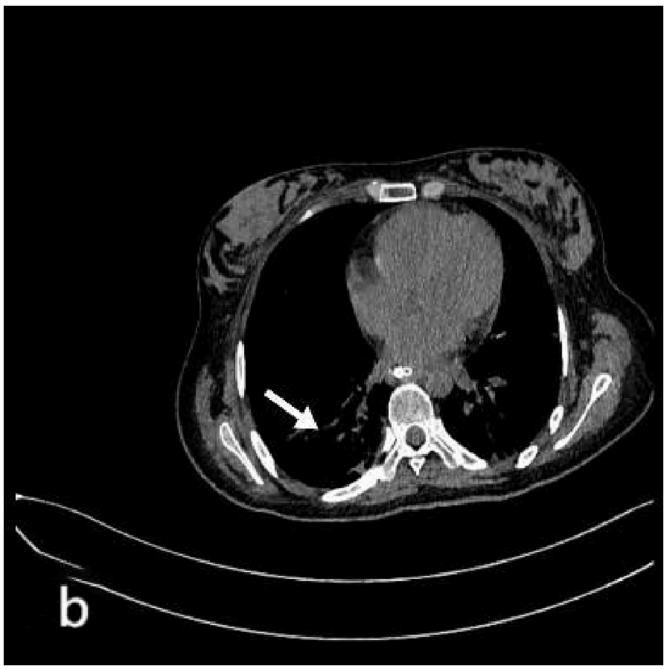

Fig. 5b. Significant absorption of pulmonary infiltration in the lower lobes. The left lung has nearly recovered, and the right lung cavitation has resolved (white arrow).

it was revealed that the patient had not been regularly cleaning or replacing the tracheostomy tube after discharge. Physical examination showed partial dislodgement of the tracheostomy tube with purulent, foul-smelling secretions. Laboratory tests indicated an elevated neutrophil ratio (85.2 %), CRP of 7.04 mg/L, and persistent hyperglycemia (random blood glucose: 14.53 mmol/L).On Day 59, follow-up neck CT revealed cavity formation at the tracheostomy site, accompanied by an elliptical high-density shadow and worm-eaten changes (Fig. 6).

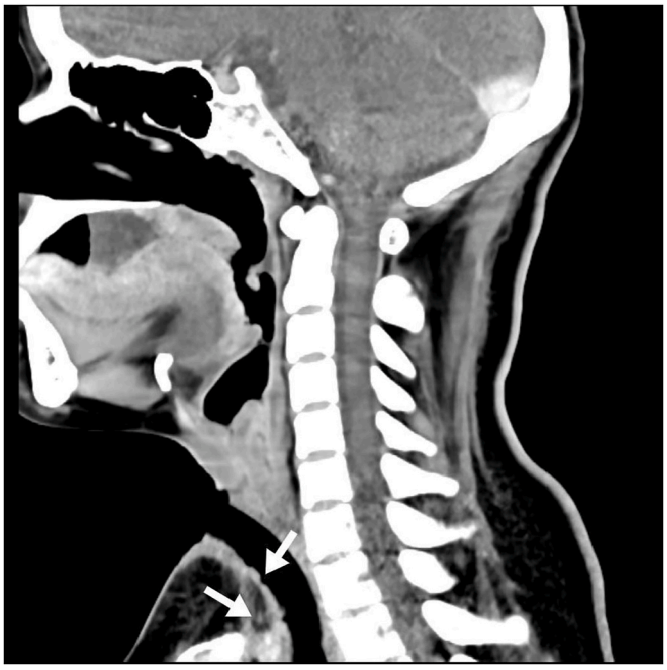

Fig. 6. A cavity formation is detected at the tracheostomy site, containing an oval high-density shadow, with worm-eaten-like destruction of the surrounding structures (white arrow).

Fiberoptic nasopharyngoscopy showed smooth tracheobronchial mucosa without congestion, edema, or white plaques (Fig. 7a).After tracheostomy tube removal, a sinus tract was identified at the distal end (Fig. 7b). The sinus tract was dissected (Fig. 7c), and three maggots were extracted from the tracheostomy site (Fig. 7d–e).The patient received intravenous cefotaxime sodium (1 g per dose, administered every 12 hours for 5 days) for anti-infective therapy, along with esomeprazole for acid suppression and metabolic support. Her symptoms significantly improved, and she remained afebrile, leading to discharge.

On Day 231, a telephone follow-up revealed that the patient had undergone repeat neck CT and fiberoptic bronchoscopy at an external hospital, with no significant abnormalities detected. She continues to live with a tracheostomy tube and is scheduled for tracheal anastomosis surgery in the future.

3. Discussion

Invasive mucormycosis is a highly lethal and fulminant opportunistic infection. Classic risk factors include immunosuppressive conditions such as diabetic ketoacidosis, hematologic malignancies, and solid organ transplantation [3]. In recent years, the COVID-19 pandemic has further increased the risk of secondary mucormycosis infection in susceptible patients [4].The present case involves a 24-year-old female with type 1 diabetes mellitus who had been experiencing chronic uncontrolled hyperglycemia (HbA1c 10.1 %, random blood glucose >14 mmol/L) accompanied by persistent ketosis (β-hydroxybutyrate 2.36 mmol/L). The diabetic environment provides a unique niche for mucormycosis, with underlying mechanisms including: (1) Diabetes-induced neutrophil dysfunction, characterized by impaired chemotaxis, reduced phagocytic index, and decreased NETosis formation, which collectively weaken host immune surveillance [5,6].(2) In diabetic patients, especially those with diabetic ketoacidosis (DKA) or other forms of acidosis, elevated blood glucose levels and decreased blood pH can impair the iron-binding capacity of ferritin, lactoferrin, and transferrin, leading to increased serum free iron concentrations. The elevated free iron levels promote the growth and invasive infection of mucormycosis [3].Additionally, tracheostomy-induced disruption of the mucosal barrier alters the invasion pathway of mucormycosis,

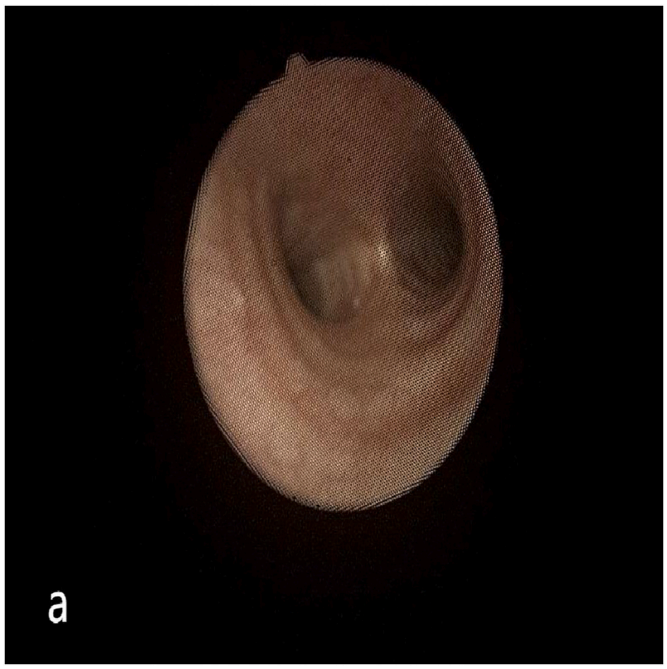

Fig. 7a. Flexible nasopharyngoscopy reveals a smooth trachea and bronchi, with no signs of invasive lesions.

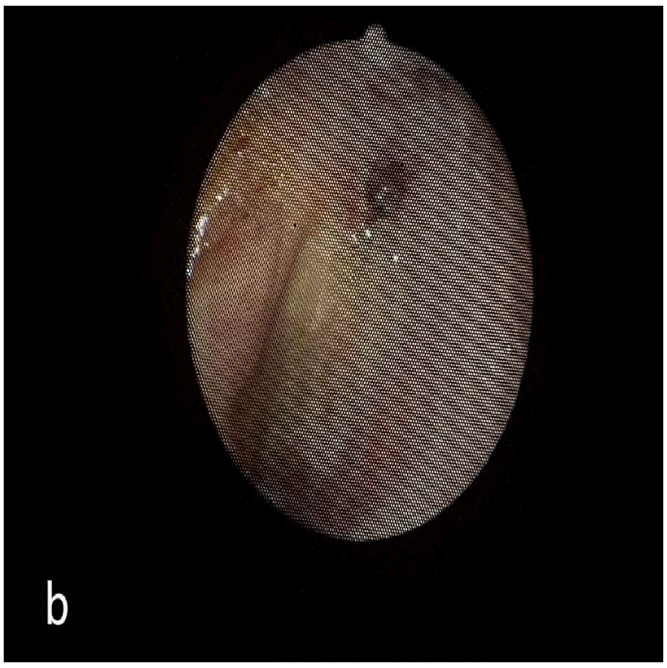

Fig. 7b. After tracheostomy tube removal, a sinus tract is observed at the anterior end of the tracheostomy site.

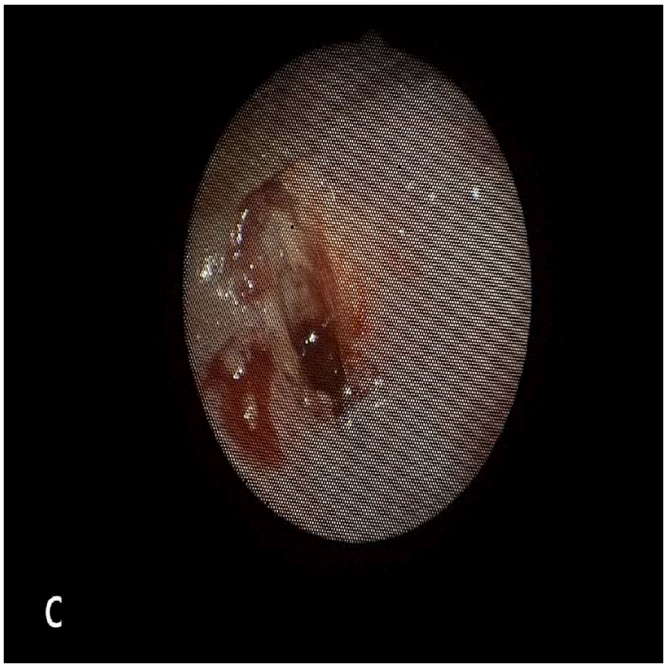

Fig. 7c. Dissection reveals the sinus tract at the anterior end of the tracheostomy site.

potentially leading to primary colonization in the trachea-mediastinum region rather than the classic rhino-orbital-cerebral site.

Tracheobronchial mucormycosis is a rare form of invasive pulmonary mucormycosis that involves the tracheobronchial tree. The most common symptoms include fever, cough, dyspnea, and hemoptysis [7]. Its atypical anatomical localization increases the risk of misdiagnosis, as it may be mistaken for bacterial mediastinitis or tuberculous granuloma, leading to delays in antifungal treatment.In this case, postoperative histopathological examination (HE staining) revealed characteristic non-septate, right-angle branching hyphae, confirming the diagnosis of invasive tracheal mucormycosis [8]. In some studies, the characteristic imaging finding known as the "reverse halo sign" (central necrosis with surrounding ground-glass opacity) has been reported to have a positive predictive value of up to 94 % for mucormycosis lesions [9]. In this patient, contrast-enhanced CT showed a peritracheal abscess with ring-like mediastinal enhancement, providing important diagnostic clues for mucormycosis.However, postoperative debridement may lead to an apparent "pseudo-improvement" on imaging, potentially delaying treatment escalation. This highlights that relying solely on imaging may underestimate fungal burden, necessitating a comprehensive evaluation integrating clinical and pathological evidence.

Studies have shown that delayed treatment is closely associated with the high mortality rate of invasive tracheal mucormycosis [10]. In this case, the patient initially received liposomal amphotericin B (L-AmB) at 5 mg/kg/day combined with nebulized inhalation therapy. Although this regimen helps reduce nephrotoxicity risk, follow-up chest CT revealed cavitation in the lungs, indicating further spread of the infection. Given the disease progression, the treatment was switched to isavuconazole (loading dose: 200 mg every 8 hours, maintenance dose: 200 mg once daily), leading to rapid clinical improvement.Compared with amphotericin B, isavuconazole offers several pharmacokinetic advantages: (1) Its long half-life supports once-daily dosing, improving patient compliance; (2) The bioavailability of oral and intravenous formulations is nearly complete (98 %), and it does not require lipid carriers, thereby reducing the risk of infusion-related toxicity; (3) Rapid achievement of steady-state plasma concentration; (4) High volume of tissue distribution, allowing for greater local drug concentration within necrotic tracheal walls [11].Therefore, isavuconazole has become the preferred option for salvage therapy and step-down treatment in mucormycosis management [12].

Furthermore, when conditions permit, a combination of antifungal therapy and early surgical debridement remains the mainstay of mucormycosis treatment [12]. Studies have shown that tracheal resection combined with antifungal therapy can significantly reduce early mortality caused by acute asphyxia or massive hemorrhage [13]. However, the procedure itself is not without risks, with major complications including airway stenosis, postoperative infection, and long-term tracheostomy tube-related issues, which may affect the patient's quality of life and long-term prognosis [14]. Prolonged tracheostomy can lead to a range of local complications, including vocal cord paralysis, mucosal ulcers, airway stenosis, and granuloma formation [15]. Mechanical failures, such as tracheostomy tube obstruction, displacement, or cuff rupture, can result in acute airway emergencies. Therefore, consistent humidification care and regular replacement of the tracheostomy tube are essential to prevent complications and ensure optimal patient outcomes.Infection remains one of the most severe complications after tracheal resection. The primary pathogens responsible for local incision infections include *Staphylococcus aureus* and *Pseudomonas aeruginosa* [16], while lower respiratory tract infections are predominantly caused by multidrug-resistant Gram-negative bacteria and fungi (such as *Candida* and *Aspergillus*), particularly in immunosuppressed patients or those receiving prolonged broad-spectrum antibiotic therapy [17].Notably, cases of myiasis following tracheal resection have been reported in recent years [17]. A similar occurrence was observed in this case, highlighting deficiencies in perioperative care and long-term postoperative management.

Myiasis is typically caused by flies laying eggs in open wounds or body cavities, where the hatched larvae feed on host tissue or secretions, leading to local tissue necrosis, secondary infections, and potentially triggering a systemic inflammatory response [18]. Moreover, mechanical damage to maggots and disruption of their surface biofilm can further exacerbate the infection response, potentially intensifying the host's inflammatory reaction and complicating the healing process [19]. In this case, the patient's neutrophil ratio rebounded to 85.2 %, providing further evidence of this vicious cycle. A comprehensive analysis suggests that the possible predisposing factors for myiasis in this

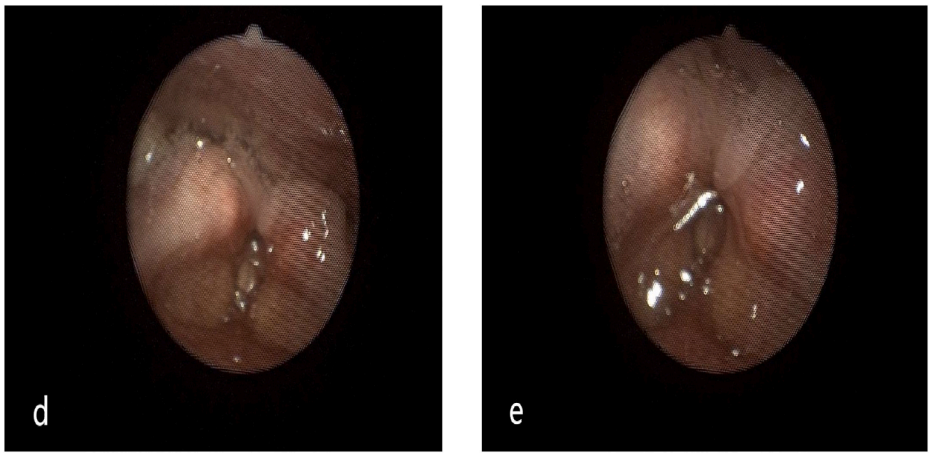

Fig. 7d–e. Larvae are extracted from the tracheostomy site.

patient include the following:(1) The purulent secretions at the tracheal incision site release cadaverine and putrescine, volatile amines that strongly attract blowflies to lay eggs.(2) Improper home care creates an ideal environment for the proliferation of maggots.(3) The patient's long-standing diabetes and immunosuppression reduce resistance to parasitic infections, facilitating maggot infestation.(4) Mucormycosis-induced necrotic lesions provide a rich nutrient source for maggot growth.In recent years, the incidence of mucormycosis has been steadily rising [20], and cases of tracheal mucormycosis may also become increasingly common. For patients requiring long-term tracheostomy following tracheal mucormycosis surgery, management and treatment strategies should be further optimized to reduce the risks of postoperative infection and myiasis.For such patients, the following management measures are recommended:(1) Thorough surgical debridement with regular postoperative imaging follow-ups to prevent cavity formation and fluid accumulation.(2) Optimized discharge education, providing clear guidance to family members on daily cleaning and disinfection of the tracheostomy tube to prevent fly exposure to open wounds.(3) Enhanced community care management, recommending regular follow-ups to dynamically assess the tracheal incision and systemic infection status, thereby preventing severe complications due to inadequate care.(4) Improved nursing quality, with strengthened training for both hospital and community healthcare providers to ensure standardized long-term management of patients undergoing tracheal resection.The implementation of these comprehensive measures is expected to effectively reduce the incidence of postoperative infections and myiasis in tracheal mucormycosis patients, ultimately improving their quality of life and overall prognosis.

In conclusion, while tracheal resection combined with antifungal therapy is an effective treatment for mucormycosis, alleviating airway obstruction and the risk of asphyxia, postoperative management is crucial. Beyond traditional infection prevention measures, special attention should be given to rare but severe complications such as myiasis. A multifaceted approach—including patient education, community nursing care, and improvements in medical devices—should be adopted to optimize perioperative management and enhance long-term quality of life.

CRediT authorship contribution statement

Yuanjiang Zheng: Writing – review & editing, Writing – original draft, Formal analysis, Data curation, Conceptualization. **Li Yang:** Investigation, Formal analysis. **Jianglin Yu:** Investigation, Conceptualization. **Shanyu Wang:** Investigation. **Xianwei Ye:** Writing – review & editing, Project administration, Funding acquisition.

Ethical form

The authors declare no conflicts of interest. Written informed consent for the publication of this case report was obtained from the patient or their legal guardian.

Declaration of competing interest

None.

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References

[1] M.G. Kaliya, M.C. Rathod, K.K. Deokar, S. Singhal, Silent choker: unveiling tracheal stenosis as an atypical manifestation of mucormycosis, Lung India 41 (1) (2024) 63–66.
[2] Q. He, X. Huang, Fatal tracheal and bronchial destruction due to pulmonary mucormycosis in a 20-year-old with diabetic ketoacidosis, Infect. Drug Resist. 17 (2024) 5429–5437.
[3] A. Alqarihi, D.P. Kontoyiannis, A.S. Ibrahim, Mucormycosis in 2023: an update on pathogenesis and management, Front. Cell. Infect. Microbiol. 13 (2023) 1254919.
[4] L. Özbek, U. Topçu, M. Manay, B.H. Esen, S.N. Bektas, S. Aydın, et al., COVID-19-associated mucormycosis: a systematic review and meta-analysis of 958 cases, Clin. Microbiol. Infect. 29 (6) (2023 Jun) 722–731.
[5] S.M. Clayton, S.H. Shafikhani, A.M. Soulika, Macrophage and neutrophil dysfunction in diabetic wounds, Adv. Wound Care 13 (9) (2024) 463–484.
[6] Y. Zhu, X. Xia, Q. He, Q.A. Xiao, D. Wang, M. Huang, et al., Diabetes-associated neutrophil NETosis: pathogenesis and interventional target of diabetic complications, Front. Endocrinol. 14 (2023 Aug 3) 1202463.
[7] V. Damaraju, R. Agarwal, S. Dhooria, I.S. Sehgal, K.T. Prasad, K. Gupta, et al., Isolated tracheobronchial mucormycosis: report of a case and systematic review of literature, Mycoses 66 (1) (2023) 5–12.
[8] J. Safiia, M.A. Díaz, H. Alshaker, C.J. Atallah, P. Sakr, D.G. Moshovitis, et al., Recent advances in diagnostic approaches for mucormycosis, J Fungi (Basel) 10 (10) (2024) 727.
[9] C. Legouge, D. Caillot, M.L. Chrétien, I. Lafon, E. Ferrant, S. Audia, et al., The reversed halo sign: pathognomonic pattern of pulmonary mucormycosis in leukemic patients with neutropenia? Clin. Infect. Dis. 58 (5) (2014) 672–678.
[10] M.M. Czech, J. Cuellar-Rodriguez, Mucormycosis, Infect. Dis. Clin. N. Am. 39 (1) (2025) 121–144.
[11] M.C. Arendrup, R.H. Jensen, J. Meletiadis, In vitro activity of isavuconazole and comparators against clinical isolates of the Mucorales order, Antimicrob. Agents Chemother. 59 (12) (2015) 7735–7742.
[12] O.A. Cornely, A. Alastruey-Izquierdo, D. Arenz, S.C.A. Chen, E. Dannaoui, B. Hochhegger, et al., Global guideline for the diagnosis and management of mucormycosis: an initiative of the European confederation of medical Mycology in cooperation with the mycoses study group education and research consortium, Lancet Infect. Dis. 19 (12) (2019) e405–e421.

[13] R. He, C. Hu, Y. Tang, H. Yang, L. Cao, R. Niu, Report of 12 cases with tracheobronchial mucormycosis and a review, Clin. Res. J 12 (4) (2018) 1651–1660.
[14] S. Kumar, K. Biradar, A. Patra, Tracheostomy complications: insights from a two-year, 15-case analysis at a tertiary care facility, Indian J. Otolaryngol. Head Neck Surg. 76 (1) (2024) 1470–1475.
[15] J.R. Lechien, S. Hans, T. Mau, Management of bilateral vocal fold paralysis: a systematic review, Otolaryngol. Head Neck Surg. 170 (3) (2024) 724–735.
[16] C. Grønhøj, B. Charabi, C.V. Buchwald, T. Hjuler, Indications, risk of lower airway infection, and complications to pediatric tracheotomy: report from a tertiary referral center, Acta Otolaryngol. 137 (8) (2017) 868–871.
[17] W. Huang, C. Zeng, W. Song, P. Xu, Intratracheal myiasis followed by tracheal-esophageal fistula: report of a rare case and literature review, BMC Infect. Dis. 19 (1) (2019) 1060. Published 2019 Dec 17.
[18] F. Francesconi, O. Lupi, Myiasis, Clin. Microbiol. Rev. 25 (1) (2012) 79–105.
[19] T.A. McGraw, G.W. Turiansky, Cutaneous myiasis, J. Am. Acad. Dermatol. 58 (6) (2008) 907–929.
[20] F. Danion, A. Coste, C. Le Hyaric, C. Melenotte, F. Lamoth, T. Calandra, et al., What is new in pulmonary mucormycosis? J Fungi (Basel) 9 (3) (2023) 307.