

Respiratory tract obstruction due to intraluminal tracheal schwannoma. The first report in Indonesia

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Abstract

Objective: To enrich the literature regarding tracheal schwannoma (TS), which can cause airway obstruction and has a high mortality risk. The authors will also discuss the management strategies for this case in a clinical setting.

Design: This article was a rare case report.

Setting: The case was identified and managed at a tertiary referral hospital in West Nusa Tenggara Province, Indonesia.

Patient and participant: A 34-year-old man presented with symptoms of airway obstruction and was examined using a computerized tomography (CT) scan and bronchoscopy. According to all in-

vestigations, the diagnosis was an intraluminal tracheal tumor.

Intervention: Complete tumor resection was successfully performed using an endoscopic approach in this patient.

Measurement and result: Post-operative evaluation revealed no symptoms or tracheal mass. The histopathology examination after surgery confirmed that the tumor was a schwannoma.

Conclusion: Intraluminal TS can lead to airway obstruction, increasing the risk of mortality. Complete resection should be performed to achieve excellent treatment outcomes.

Keywords: Tracheal tumor, tracheal schwannoma, endoscopic resection, airway obstruction.

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Background

Primary tracheal tumors are rare cases. The incidences of tracheal tumors are dominated by the malignant type. (1) One of the benign tumors of the trachea is tracheal schwannoma (TS). TS is a benign tumor that originates from the peripheral nerve sheath. The etiology of schwannomas is attributed to the malfunction of the mutations in the neurofibromatosis type 2 (NF2) tumor suppressor gene. (2)

The symptoms of TS vary depending on the site, size, and extent of the tumor's obstruction. (3) In the early stage, TS can present with asthma-like symptoms or upper airway obstruction, and less frequently with hemoptysis and hoarseness. Therefore, the diagnosis of TS is challenging due to its similarity in presentation to other diseases, such as asthma and chronic obstructive pulmonary disease. (4)

In the advanced stages, TS can cause upper airway

obstruction by wholly or partially occluding the trachea, leading to compromised ventilation. Therefore, it may increase the risk of mortality when the diagnosis and treatment are not carried out correctly. Proper diagnosis and treatment can improve patient outcomes. Furthermore, reports of TS in Indonesia are minimal. The aim of this report was to enrich the literature canon on TS that may cause airway obstruction and carry a high mortality. The authors would also like to discuss the strategy for managing this case in the clinical setting.

Case presentation

A 34-year-old man was referred from a secondary hospital in Mataram City after four days of treatment in the intensive care unit. The patient came to the hospital due to a severe shortness of breath for 2 days before entering the ward. Two weeks before the latest hospitalization, the patient was also consulted at the outpatient clinic at a similar hospital with a complaint of bloody cough with a volume of about 3 ml. The patient has also been feeling chest pain with dysphagia and odynophagia for 1 week before being admitted. He had no history of fever, heartburn, or vomiting, experienced appetite dysfunction, and weight loss. However, he did not record his weight precisely. This gentleman also presented with intermittent shortness of breath for the past 7 months, accompanied by a dry cough. Due to those symptoms, he underwent a molecular rapid test on the sputum 4 weeks ago, and the result was negative. The patient had a history of mild hypertension and had been working in the paint industry for 10 years.

Physical examination revealed mild hypertension, wheezing, and ronchi in auscultation. Laboratory tests were within normal limits, except for an elevated white blood cell count. A bronchoscopy test was performed at the referral hospital. During bronchoscopy, an intraluminal tracheal mass was noted, solid and easily bleeding, leading to tracheal stenosis (**Figure 1**). Computerized tomography (CT) thorax without and with contrast revealed a mass in the intraluminal space of the trachea (at the level of Th 3–4) with a size of 2.1 x 1.35 x 1.13 cm (**Figure 2**). The patient was finally diagnosed with an intraluminal tracheal tumor. The Tumor Board was scheduled to discuss the treatment that would be given to this patient while he was observed in the intensive care unit (ICU) after the attack of severe intermittent airway obstruction.

Considering the patient could not be intubated or undergo tracheostomy before surgery, the patient in this case underwent endoscopic surgery with the back-up of a thoracic surgeon. The patient planned

to undergo general anesthesia, then undergo an endoscopic incision and tracheal evaluation; however, if the procedure failed, then an external approach would be done.

During the procedure, there was an intraluminal mass that filled almost the entire trachea with attachment to the right wall of the trachea (**Figure 3**). Tumor resection was done successfully, and tumor samples (**Figure 4**) were sent for histopathological examination. The result showed a spindle cell neoplasm typical of schwannoma tumors (**Figure 5**). After the procedure, the patient was observed in the ICU for a day. The condition was improving, as observed in the ward before discharge in 2 days. A week later, the patient returned to the outpatient clinic and reported no complaints. A CT scan was done; the result showed no mass in the trachea.

Discussion

TS originates from Schwann cells of the intraluminal sheath. The first case of tracheal schwannoma was reported in 1951. (5) Tracheal schwannoma is a benign tumor with a predilection for the upper one-third and middle one-third of the trachea. (3) The etiology of TS remains unclear; however, the tumor manifests more frequently in young adults and females gender. Recent studies have demonstrated a correlation between the pathophysiology of TS and genomic alterations in neurofibromatosis type 2 (NF2), adenine and thymine-rich interaction domain 1A (ARID1A), adenine and thymine-rich interaction domain 1B (ARID1B), and discoidin domain receptor 1 (DDR1) genes. Mutations in these genes lead to cell proliferation and tumorigenesis. (2,3,5)

Tracheal innervation arises from nerves in the tracheal wall. The innervation of the trachea, which is responsible for sensory and motor functions, originates from the branches of the vagus nerve. The superior laryngeal nerve arises from the vagus nerve. This nerve has two branches: the internal branch and the external branch. The internal branch innervates the upper and lower parts of the vocal cord, while the external branch innervates the cricothyroid muscle. (6) The right and left recurrent laryngeal nerves are branches of the vagus nerve. Both recurrent laryngeal nerves sensation and motor innervation from the lower vocal cord to the laryngo-tracheal supply parasympathetic (secretory glands). (6,7)

Risk factors that may be associated with this case include exposure to chemicals used in the paint industry. Several chemical substances have been identified as risk factors for airway symptoms among paint industry workers. The chemical substances related to the paint industry, called volatile organic

compounds, consist of benzene, ethyl benzene, toluene, xylene, styrene, N-butyl acetate, isobutyl acetate, acetone, ethanol, etc. (8) Furthermore, paint industry workers are also at risk of lung cancer. (9)

The patient in our case showed symptoms of airway obstruction. This was indicated by complaints of shortness of breath and cough. Auscultation of the lungs found wheezing and ronchi. The clinical presentation of TS depends on the tumor's size, site, and degree of airway obstruction. (4,10) Therefore, patients with TS present with nonspecific symptoms. In the early stages, it can be asymptomatic or with shortness of breath and a dry cough. In severe cases, breathing difficulties will occur and lead to fatal consequences. Other rare but possible symptoms include hoarseness, chest pain, and hemoptysis. (2,4,10,11)

Imaging studies have been shown to contribute to the initial diagnosis and subsequent surgical planning in TS cases. Chest X-rays are considered normal in 75% of cases; they offer no diagnostic value. (3) CT scanning plays a vital role in diagnosing these cases. It can be used to evaluate the lumen, presence of a mass, its size and site, the degree of airway obstruction, and its extratracheal extension. (2,3,10,11) Bronchoscopy is a crucial examination for determining these cases. This examination is used to both visualize the mass and obtain a biopsy. (3,5,12)

Histopathological examination of the TS showed a tissue section lined with fibrous connective tissue, demonstrating a benign tumor composed of dense areas (Antoni A) characterized by spindle-shaped cells, some appearing obliquely arranged in fascicles, and loose, microcystic areas (Antoni B) with oval to spindle-shaped cells embedded within a fi-

brocollagenous stroma. (2,5) In the present case, a specific appearance of TS was found.

As to the present case, the history-taking should be carefully conducted to identify the risk factors. Physical examination may reveal signs of airway obstruction and should be confirmed with bronchoscopy and X-ray imaging, which will guide the physician in distinguishing the diagnosis (**Figure 6**). Histopathology work-up is the gold standard for determining the final diagnosis.

The treatment recommendation for TS is a complete resection. (5) Treatment of TS includes tracheal resection of the tumor and various endoscopic procedures, including cryotherapy, laser, electrocautery snaring, argon plasma coagulation, and microdebridement. (11) The selection of therapy in these tumor cases is based on clinical manifestations, such as pedunculated vs sessile, the risk of tracheal resection, and the presence or absence of an extratracheal component. (12) In our patient, endoscopic resection was done successfully. Endoscopic resection is indicated if the tumor size is up to 2 cm with a pedunculated mass, no extratracheal extension, or if the patient has poor cardiopulmonary function. (3) The prognosis for patients with TS, either with surgical removal or by endoscopic treatment methods, is excellent. (12)

Conclusion

An obstructing tracheal schwannoma can cause partial or complete airway narrowing, leading to ventilation impairment. Complete resection is the gold standard for the management of these tumors, with various methods tailored to clinical tumor characteristics.

Figure 1. Endoscopic view found a mass intraluminal tracheal (blue arrow), and the carina cannot be seen

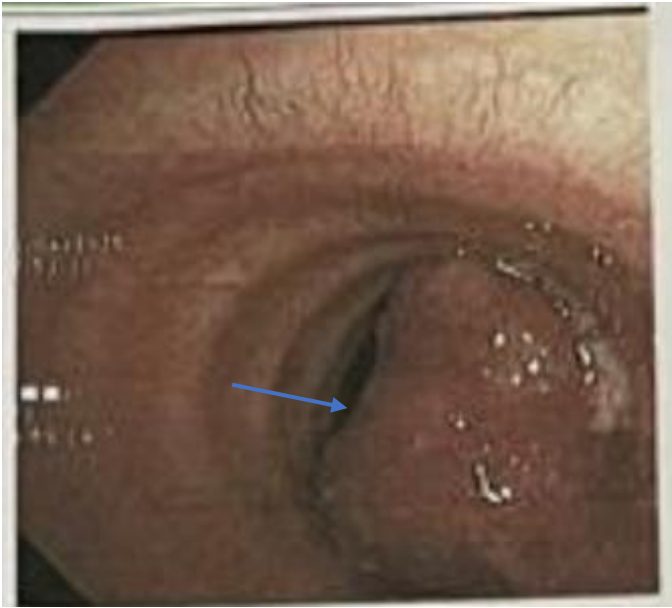


Figure 2. The computerized tomography scan showed an intraluminal mass (blue arrow)

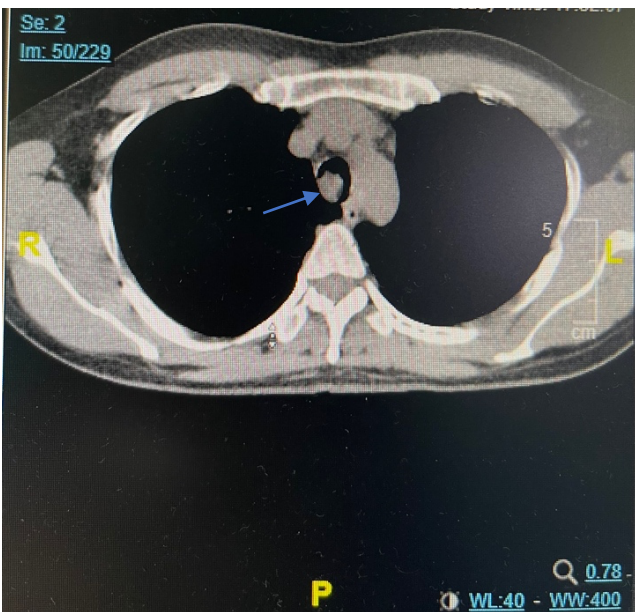


Figure 3. Endoscopic view after mass resection (blue arrow is carina)

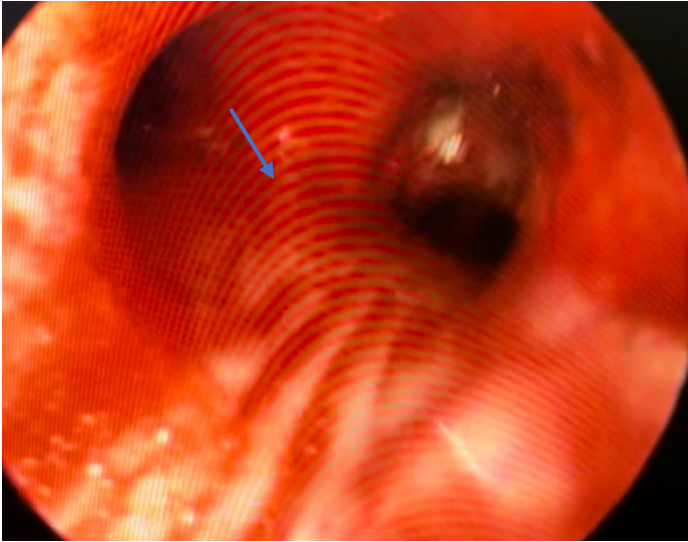


Figure 4. Resected intraluminal mass with size 2x1.5x1 cm



Figure 5. Histopathological figure of tracheal schwannoma (400x magnification) (the red arrow shows spindle cells)

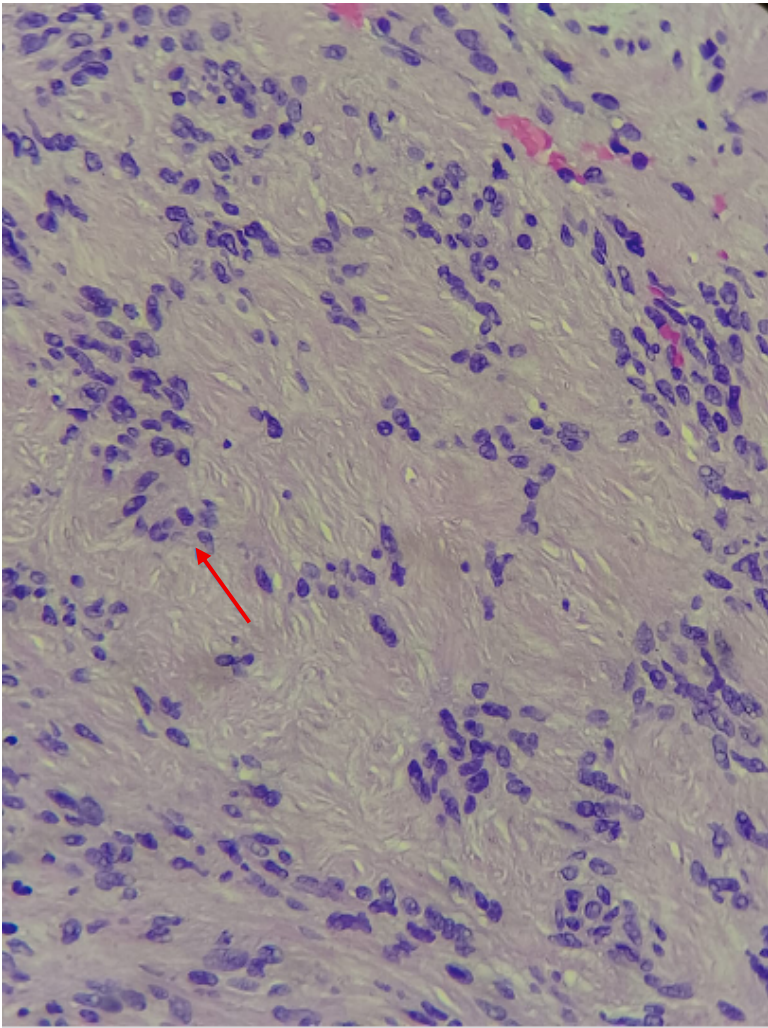
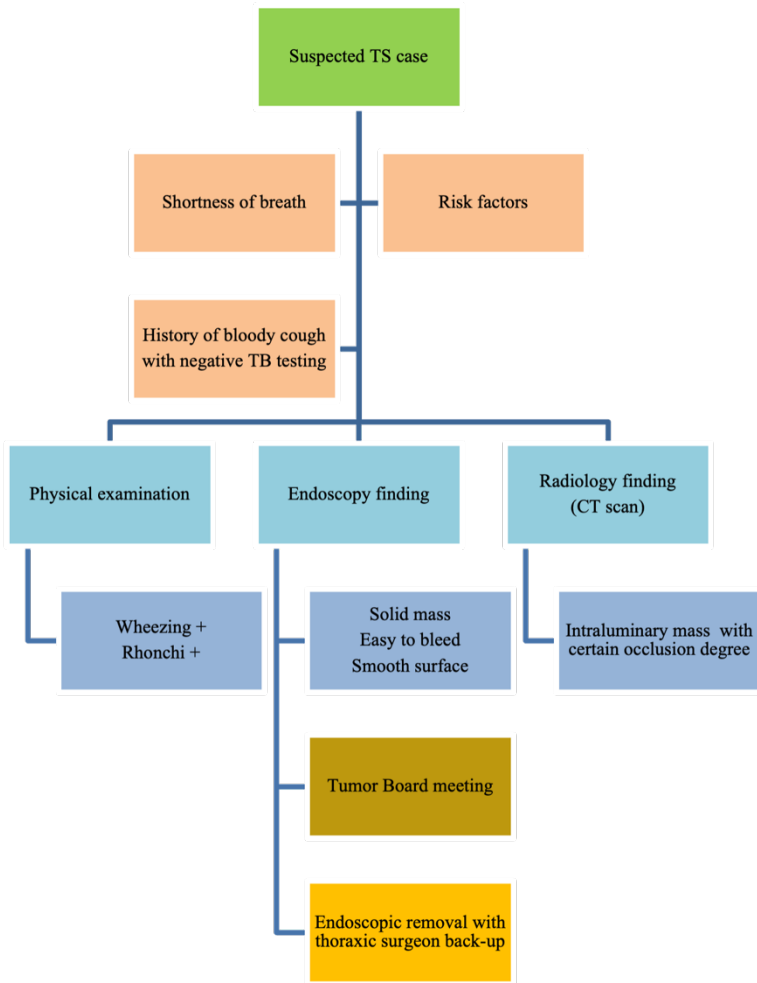


Figure 6. Management of intraluminal TS



Legend: TS=tracheal schwannoma; TB=tuberculosis; CT=computerized tomography.

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