CASE REPORT

Pin Point Trachea: A Case Report

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ABSTRACT

Introduction: Pin point trachea is a rare case. It is caused by tuberculosis (TB) and trauma due to intubation procedure. Main complication of this abnormality is respiratory failure. It can be diagnosed through bronchoscopy. Managements of this abnormality are interventional bronchoscopy and treating the etiology. Surgery is considered if interventional bronchoscopy failed or cannot be performed.
Case: A 29-year-old woman came to the emergency room complaining shortness of breath and hoarseness since two months before hospitalized. The patient also experienced cough, decreased body weight and appetite since 7 months earlier. The patient was diagnosed with bacteriologically confirmed TB and anti-TB drug was given. Cough symptom was decreasing but shortness of breath and hoarseness remained after treatment. Physical examination showed decreased vesicular sound and stridor. Bronchoscopy revealed narrow tracheal lumen (pin point) with fibrosis. Thoracic computed tomography (CT) scan showed severe narrowing of tracheal lumen at thoracic vertebrae 1-2. Surgery was performed to cut the fibrotic tissue and tracheostomy was placed at stenosis area.
Conclusion: Pin point trachea is a rare case. One of the causes is tracheobronchial TB. The main managements are optimal administration of anti-TB drugs and interventional bronchoscopy or surgery.

INTRODUCTION

The respiratory tract is one of the targets of various diseases. Central airway obstruction (CAO) is generally defined as occlusion of >50% of the trachea, main stem bronchi, bronchus intermedius or a lobar bronchi.[1] The etiology can be divided into neoplastic and non-neoplastic. Non-neoplastic causes of central airway disorders include congenital abnormalities, infection, iatrogenic trauma, and systemic inflammatory disease.[2]

Pin point trachea (tracheal fibro-stenosis) is a rare central airway disorder. Its causes include trauma due to intubation and tracheostomy procedures (10-22%), systemic inflammation, and infection. The most common infection which causes tracheal fibro-stenosis is tuberculosis (TB).[3,4]

Tracheobronchial TB is a TB infection of the tracheobronchial tree with microbial and histopathological evidence. It is difficult to diagnose because it may not appear normal on chest X-ray unless there is significant airway obstruction leading to atelectasis or concurrent parenchymal or pleural disease. This causes delays in diagnosis and treatment.[5,6] Endobronchial TB tends to affect the right upper and right main bronchus.[1] Su, *et al*. reported about 23.9% of pulmonary TB patients underwent bronchoscopy were presented with tracheobronchial TB.[7] Jung, *et al*. reported that 54.3% of patients with pulmonary TB also had tracheobronchial tuberculous lesions, with female gender and duration of symptoms above 4 weeks being the main predictors. Tracheobronchial fibro-stenosis

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develops 4-6 months from disease onset in 68% of patients with tracheobronchial TB and it can result in significant morbidity.[8] Su, *et al*. suggested young and middle-aged females with symptoms persisting for ≥4 weeks (the main predictors of trachea bronchial TB and related tracheobronchial stenosis) should receive bronchoscopy immediately when diagnosed with pulmonary TB.[7] Patients with pulmonary TB and continuous respiratory symptoms during or after treatment should be assessed for tracheobronchial involvement.[9]

The clinical feature of patients with tracheal fibro-stenosis varies depending on the cause, location of the lesion, degree of luminal narrowing, and progression of the lesion. The general condition of the patient also plays an important role. Respiratory distress is experienced by approximately 54% of patients with tracheal fibrostenosis.[4]

Bronchoscopy is the main method to diagnose the presence of tracheal fibro-stenosis and tracheobronchial TB. Bronchoscopy is performed when tracheobronchial TB is suspected, usually in patients complaining severe cough, hemoptysis, wheezing, or a positive acid-fast bacillus sputum smear with normal chest X-ray.[8] Interventional approach is the main management of tracheal fibro-stenosis, including interventional bronchoscopy with balloon dilation, stent placement after balloon dilatation, laser photo-resection, argon plasma coagulation, and cryotherapy. Persistent airway stenosis after balloon dilatation may occur, especially if there is active inflammation, calcification, and malacia. The incidence of restenosis after balloon dilatation was 37.5%. Other alternative management is surgery.[1,3]

CASE

A 29-year-old female patient came to the emergency room complaining shortness of breath and hoarseness since two months before hospitalized, coughing up phlegm since seven months prior, accompanied by weight loss and appetite. The patient had been diagnosed with bacteriologically confirmed TB four months earlier. She was given anti-TB drug containing isoniazid, rifampicin, pyrazinamide, and ethambutol every day for two months, followed by isoniazid and rifampicin three times a week for the next two months. After receiving anti-TB drug, the cough with phlegm decreased, but the shortness of breath and stridor persisted. Acid fast bacilli sputum smear examination after two months of intensive phase showed negative results. Vital sign revealed an afebrile patient with a blood pressure of 110/70 mmHg, pulse 104 times per minute, respiration 22 times per minute, and oxygen saturation of 99% with nasal cannula oxygenation of 2 liters per minute. Physical examination of the lungs, breath sounds were decreased in both lung fields and stridor. Complete blood count showed leukocytosis 12.73 10µ/µL with neutrophils 48.07% and lymphocytes 44.91%. A compensated respiratory acidosis with arterial partial pressure of CO2 54.1 mmHg was found from the blood gas analysis. Chest X-ray examination showed opacity at the apex of the right lung as shown in Figure 1. Computed tomography (CT) scan of the neck and thorax showed severe narrowing of tracheal lumen at thoracic vertebrae 1-2 and opacity of the apex of the right lung (Figure 2, 3). Bronchoscopy result showed narrowed tracheal lumen (pin point) with fibrosis (Figure 4).

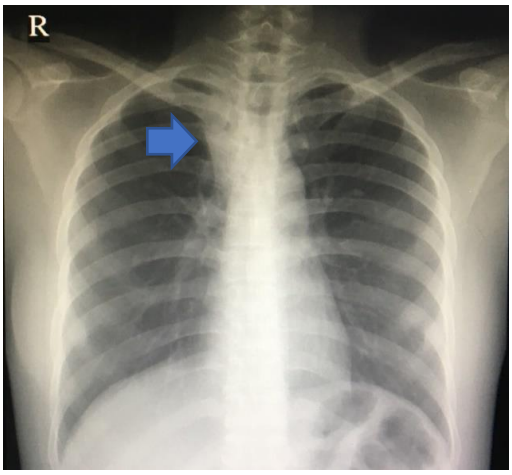

Figure 1. Chest X-ray showed an opacity at the apex of the right lung

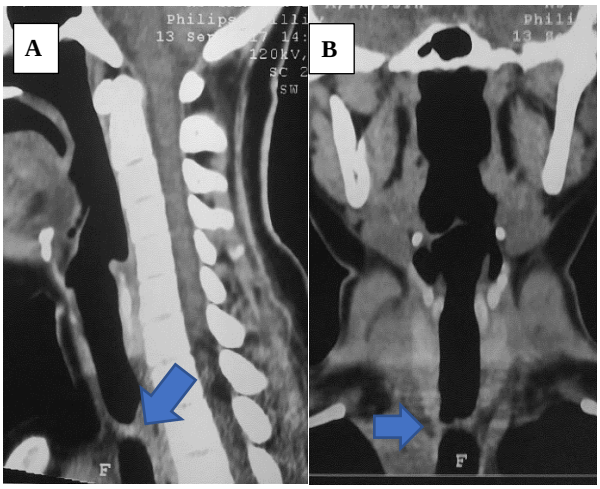

Figure 2. CT scan of the neck **A)** sagittal; **B)** coronal slices showed narrowing of tracheal lumen at thoracic vertebrae 1-2

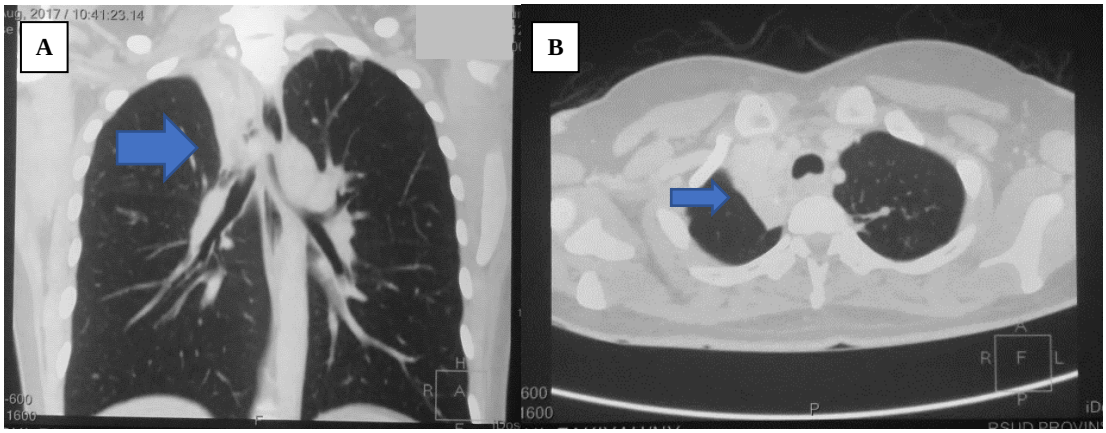

Figure 3. Thoracic CT scan **A)** coronal; **B**) axial slices showed narrowing of tracheal lumen at thoracic vertebrae 1-2 and opacity of the apex of the right lung

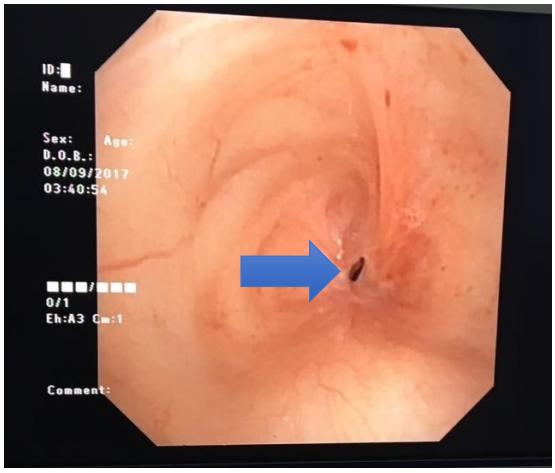

Figure 4. Bronchoscopy showed a narrowed (pin point) tracheal lumen due to fibrosis

The patient was consulted to cardiothoracic surgeon and surgery. From that consulted was performed with excision of fibrotic tissue at the 5th-6th tracheal rings, release stenosis, and tracheostomy in the area of stenosis (Figure 5).

Tracheostomy can only be performed with a 6F cannula, thus a follow-up bronchoscopy was performed to evaluate the distal airway. Follow-up bronchoscopy showed an intraluminal bulge in the distal trachea and right main bronchus (Figure 6).

The results of histopathological examination of the resected tissue revealed a fibrotic tissue with non-specific inflammation.

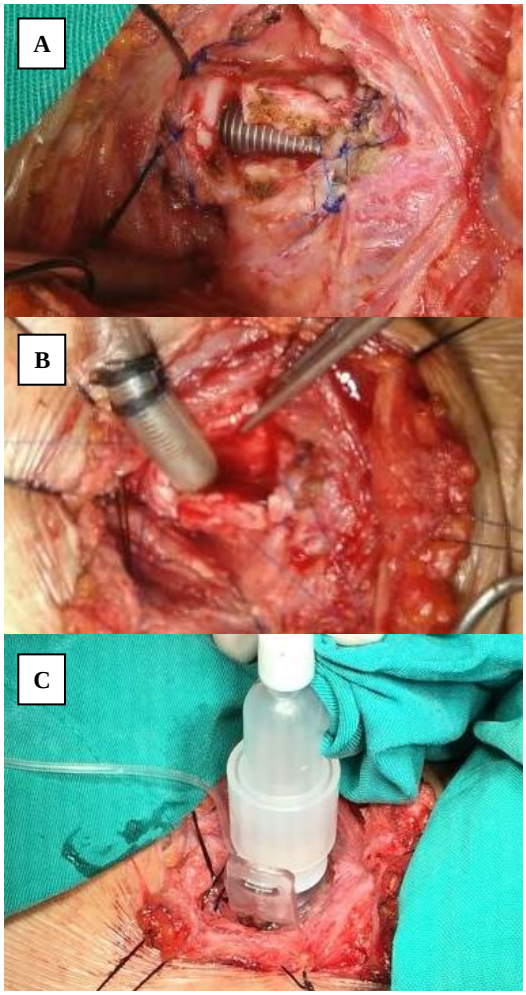

Figure 5. A) and **B)** Excision of fibrotic tissue and release of stenosis at the level of the 5th-6th tracheal ring; **C)**Tracheostomy performed at the site of stenosis with a 6F cannula

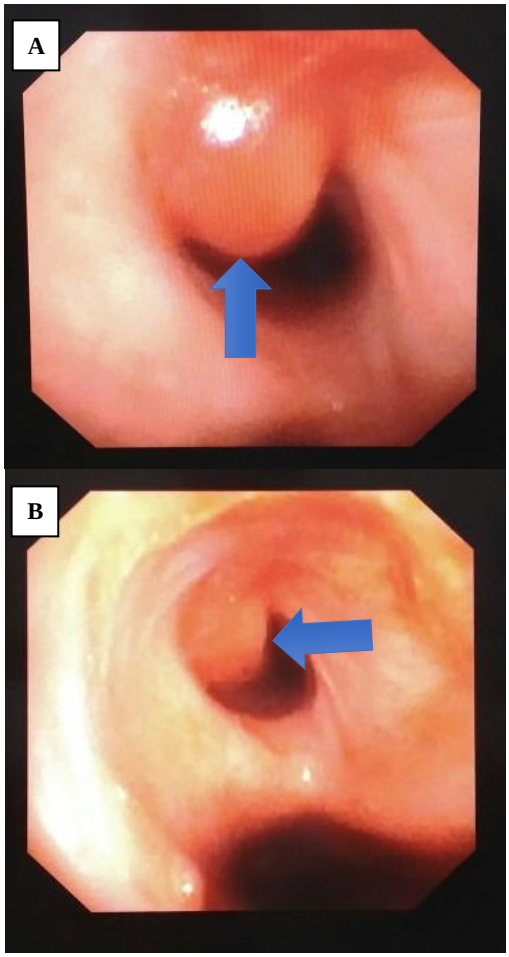

Figure 6. A) Bronchoscopy image of intraluminal bulge at distal trachea; **B)** Bronchoscopy image of intraluminal bulge on the right main bronchus

DISCUSSION

Pin point trachea (tracheal fibro-stenosis) is a rare case. It has various causes, including trauma due to intubation and tracheostomy procedures, malignancies, systemic inflammatory diseases, and infections. Tracheal fibro-stenosis is one of the long term complications of tracheobronchial TB.[3]

Tracheobronchial TB is TB infection of the tracheobronchial tree with microbial and histopathological evidence. It is difficult to diagnose because it may appear normal on chest x-ray and causes delays in treatment.[5,6]

Jung, *et al*. reported that 54.3% of patients with pulmonary TB also had tracheobronchial tuberculous lesions, with female gender and duration of symptoms above 4 weeks being the main predictors.[8] Although the reasons for the female predominance are unclear, some studies have pointed that since females have smaller tracheobronchial lumens, they have weaker expectoration capacity than males, which possibly make females prone to direct implantation of *Mycobacterium tuberculosis* (M.Tb) within the expectorated phlegm from pulmonary focus.[10] The high incidence of endobronchial TB in women may also be caused by sociocultural factors which expect women not to expect excessive phlegm, hence the exposure to TB germs in the bronchi becomes longer.[11] Su, *et al*. supposed that symptoms duration might result in longer exposure to MTB and contribute to the development of tracheobronchial TB and related tracheobronchial stenosis.[7] Tracheal fibro-stenosis develops 4-6 months from disease onset in 68% of patients with tracheobronchial TB.[6] Patients with pulmonary TB and continuous respiratory symptoms during or after treatment should be assessed for tracheobronchial involvement.[9] This finding is in line with this study. The patient was a woman with symptom duration above 4 weeks, shortness of breath and stridor occurred after 5 months from symptom onset.

Tracheobronchial TB occurs in several ways, including direct spread of TB bacteria from parenchyma or cavity lesions, dissemination from the peri bronchial lymphatic pathways, spread through the mediastinal lymph nodes to the bronchial mucosa, which sometimes causes broncho-nodal fistulas (more common in pediatric patients), and direct implantation of inhaled bacteria on the airway mucosa.[6]

Tracheobronchial TB can affect any part of the tracheobronchial tree, the most commons are the main bronchus, bilateral superior lobe bronchus, and right middle lobe bronchus.[5] This finding is different from this study, in which the lesion was located in the trachea, causing stridor and respiratory distress.

TB infection in the tracheobronchial tree generally improves if the primary disease has been controlled, but if the inflammation extends deeper into the bronchial mucosa, ulceration and necrosis will occur, then in healing process fibrosis occurs, and eventually bronchial stenosis is formed.[6] Pathologically, tracheobronchial TB can invade any layer of the tracheobronchial wall, including the lamina muscularis and cartilage. Pathological changes which occur are infiltration of the mucosa and submucosa by bacteria, ulceration, granulomas, fibroplasia, and tracheobronchial stenosis. Initially, mucosal and submucosal hyperemia occur due to infiltration of inflammatory cells, mainly lymphocytes. Tuberculous nodules are formed followed by caseous necrosis of the nodules and mucosal ulceration. Multiple superficial ulcers then become apparent and occur most commonly on posterior walls of the trachea and major bronchi. These ulcerations may spread to the tracheobronchial wall and become deep

ulcers, or become hyperplastic polyps which protrude into the tracheobronchial lumen resembling a tumor. At an advanced stage, fibrous hyperplasia and contractures develop, resulting in tracheobronchial stenosis, where the incidence is 68% in the first 4-6 months and increases as the disease progresses.[5]

Symptoms of tracheal fibro-stenosis depends on the degree of stenosis, which can occur inspiratory and expiratory stridor. Coughing becomes ineffective and complaints of shortness of breath appear due to increased work of the respiratory muscles. An increase in carbon dioxide levels can occur and trigger shortness of breath and restlessness.[12] Before the symptoms of stenosis arise, it is preceded by symptoms of tracheobronchial TB, which is symptoms of TB infection in general, including coughing, hemoptysis, chest pain, mild fever, weight loss, and loss of appetite. Obstruction symptoms, such as decreased breath sounds and stridor or wheezing, depend on the location of the lesion, may occur.[5]

Examination of acid-fast bacilli smears gave varied results, with positive results in 13.6-53% of patients with tracheobronchial TB. Sputum results can be negative because mucus is trapped in granulation tissue. Nuclear amplification tests such as Xpert MTB/Rif give better results and are recommended in suspected cases of tracheobronchial TB.[5,6] This finding is in line with this study because the patient was bacteriologically confirmed TB.

Chest X-ray and CT scans cannot rule out tracheobronchial TB. Lee, *et al*. reported that 10% of patients with tracheobronchial TB had normal chest X-ray but pulmonary parenchymal infiltrates might be found. Tracheobronchial stenosis can lead to atelectasis or post-obstructive pneumonia, which can be seen on a chest CT scan. On a chest CT scan, active TB will show an edematous and irregular airway, while the fibrotic stage will show a smooth airway, without airway thickening or edema.[13]

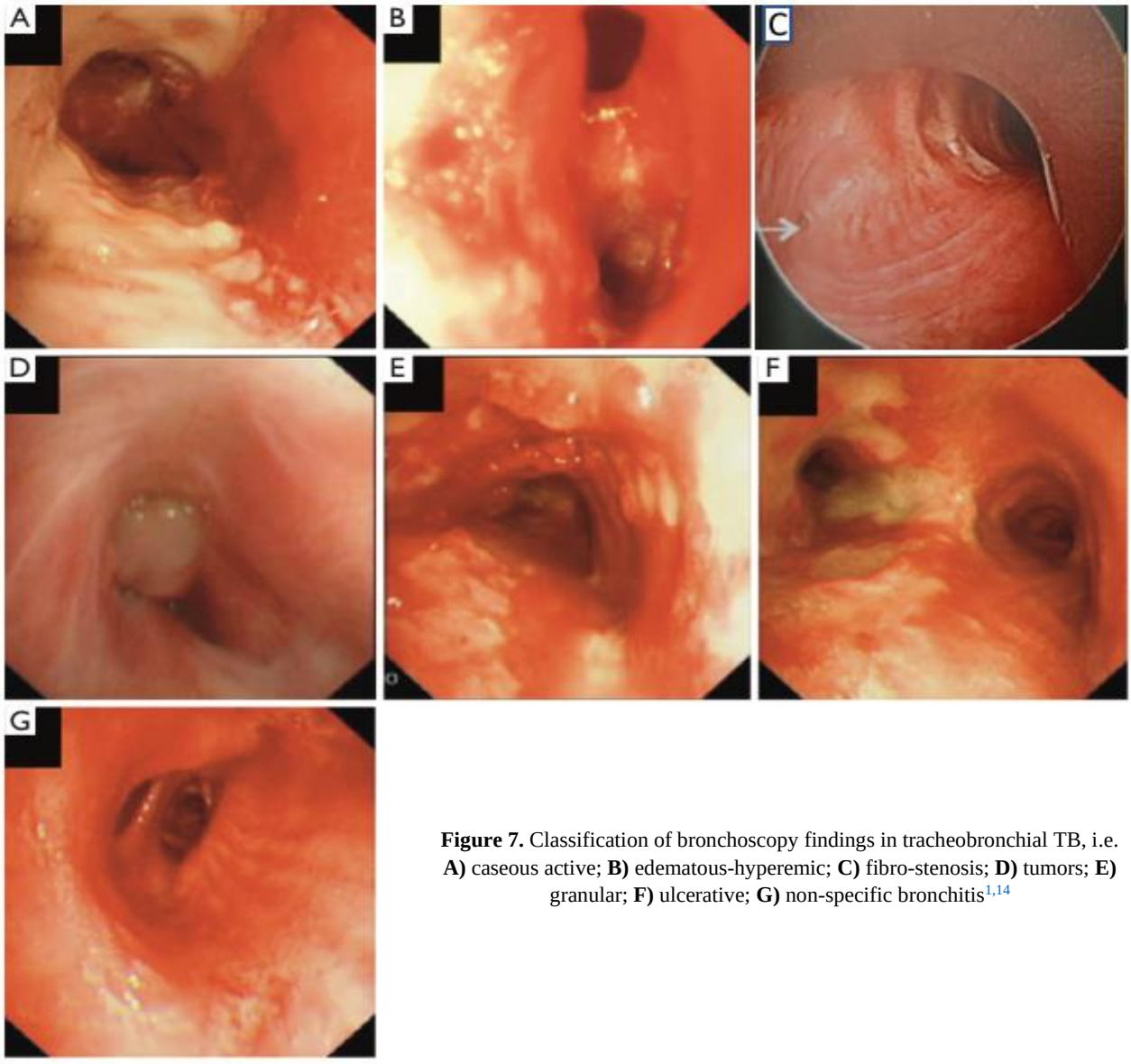

Figure 7. Classification of bronchoscopy findings in tracheobronchial TB, i.e. **A)** caseous active; **B)** edematous-hyperemic; **C)** fibro-stenosis; **D)** tumors; **E)** granular; **F)** ulcerative; **G)** non-specific bronchitis[1,14]

Bronchoscopy is the standard procedure for the evaluation of tracheobronchial abnormalities, which can be used to evaluate the function and anatomy of the trachea as well as to determine the location of the lesion. In addition, it can determine the degree of stenosis and its cause.[12] Chung, *et al*. divided tracheobronchial TB disorders based on bronchoscopy finding into seven type. Bronchoscopic features of endobronchial TB are (A) actively caseating (tracheobronchial mucosa is swollen, hyperemic and covered with a large amount of whitish cheese-like material); (B) edematous-hyperemic (tracheobronchial mucosa is severely swollen and hyperemic); (C) fibro-stenotic (tracheobronchial lumen narrows due to fibrous hyperplasia and contracture); (D) tumorous (hyperplastic focal tissue shapes, intraluminal mass like tumor); (E) granular (tracheobronchial mucosa appears severely inflammatory and is scattered by rice-like nodules); (F) ulcerative (tracheobronchial mucosa ulcerate); and (G) nonspecific bronchitic (tracheobronchial mucosa only is mildly swollen and/or hyperemia) (Figure 7).[1,14,15]

In this study, there was a fibro-stenosis type in the proximal trachea and a protrusion of the lumen in the distal trachea and right main bronchus which fit the tumor type description. Chung, *et al*. reported that the most common type was active caseous type (43%), while the fibro-stenosis type was found in 10.5% of cases.[14] This finding is also in line with the study of Su, *et al*. which found that tracheobronchial TB mainly involved the upper lobe (44.9%), followed by the main bronchus (32.8%), and trachea (31.3%). About 51.9% of tracheobronchial TB had two or more lesions. The most common complication was tracheobronchial stenosis (59.7%). Stenotic lesion mostly presented as mild-grade airway narrowing (65%), whereas 23.3% showed severe stenosis.[7] The image is a narrowing of the lumen due to fibrosis. Usually the lesions are not circular and look like crushed water drops. Active tuberculous tissue can be obtained from a biopsy of the inflamed mucosa at the edge of the lesion. These lesions tend to be progressive which lead to complete occlusion even after 2-3 months of therapy. The tumor type was found in 10.5% of cases, in the form of an endobronchial mass which could be covered by caseous tissue. The evolution of this type is difficult to predict. Some lesions will cause obstruction because of the growing mass, some will develop tumor lesions in other bronchi, and some will heal with fibrosis.[14]

The management of tracheal fibro-stenosis includes medical and interventional management. Optimal administration of anti-TB drugs is necessary in tracheal fibro-stenosis caused by tracheobronchial TB.[5] The use of corticosteroids in cases of tracheobronchial TB is still a matter of debate. Shim reported that corticosteroids are beneficial for tracheobronchial TB with edematous-hyperemic, caseous, and tumor types. As for the type of fibro-stenosis, corticosteroids do not provide much benefit.[16] Um, *et al*. stated that administration of corticosteroids equivalent to prednisolone 30 mg per day did not reduce the frequency of persistent airway stenosis.[17] Corticosteroids can also be given locally to the lesion. Rikimaru stated that administration of aerosol therapy with streptomycin 100 mg, dexamethasone 0.5 mg, and nafazoline 0.1 mg twice daily in conjunction with conventional oral therapy resulted in shorter healing of ulcerated lesions and less stenosis.[18] As for the patient in this study, the patient was still on anti-TB drug treatment and corticosteroid was not given.

Interventional managements include interventional bronchoscopy and surgery. Interventional bronchoscopy can use both rigid and flexible bronchoscopy. Rigid bronchoscopy provides a mechanical effect of dilatation of the stenotic lesion. In addition, balloon dilatation can be performed to widen the area of stenosis, then followed by stent placement. Ablation technique is used to maintain airway patency. This technique includes both hot and cold therapy. Mesh-like circumferential stenosis can be treated with electrocautery. Other modalities are argon plasma coagulation (APC) and laser with neodymium-ytrium-aluminum-garnet (Nd-YAG) or carbon dioxide (CO_2). Mitomycin C can be administered concurrently with cautery or laser with topical application to the area of stenosis. This is performed to reduce the inflammatory response and provide a better result. Cold therapy is performed with contact cryotherapy and spray. If interventional bronchoscopy fails or is not possible, the alternative option is surgery.[3] Due to the limited equipment for interventional bronchoscopy in the hospital, for the patient in this study, balloon dilatation could not be performed.

The type of surgery depends on the location of the stenosis, including anterior cricoid split, tracheal resection with end-to-end anastomosis, and slide trachea-plasty. Tracheal resection with re-anastomosis is considered the procedure of choice with a relatively high success rate (71-95%) and minimal morbidity. This procedure can be accomplished by a neck collar incision for high stenosis or median sternotomy for mid or lower tracheal stenosis. Multivariate analysis showed that the length of resection, age, diabetes, re-resection, laryngeal involvement, and the presence of a tracheostomy were prognostic factors for the development of complications.[3] In this study, a single layer tracheal fibro-stenosis was found in the 5th-6th tracheal rings, thus it was decided to perform fibrous tissue resection, release stenosis, and tracheostomy at the site of the stenosis.

CONCLUSION

Pin point trachea is a rare case. One of the causes is tracheobronchial TB. Its symptoms are usually shortness of breath and stridor. The most common complication which can occur is respiratory distress.

Pathologically, mucosal and submucosal hyperemia occur due to inflammatory cell infiltration. Tuberculous nodules are formed, followed by caseous necrosis and mucosal ulceration. Then, there are fibrous tissue hyperplasia and contractures, resulting in tracheal fibro-stenosis and pin point trachea.

Bronchoscopy is the main procedure which can be used for diagnosis and intervention. Managements for this case are optimal administration of anti-TB drugs and treatment for tracheal fibro-stenosis, which include interventional bronchoscopy with balloon dilatation, stent placement, laser photo-resection, argon plasma coagulation and cryotherapy, or surgery.

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Confict of Interest
The author stated there is no conflict of interest in this study.

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Authors' Contributions
Writing manuscript, collecting data of patient: NWC and VS. Reviewing and revising: KPY and IBNR. All authors contributed and have approved the final version.

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