A Case of Incidental Tracheal Diverticulum with Unusual Form in Patients Presenting with Respiratory Distress

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Received: 18 May 2021
Accepted: 04 June 2021
Published: 11 June 2021

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Citation:

Keywords:
Tracheal Diverticulum; Pulmonary; Computed Tomography

1. Abstract
Tracheal Diverticulum (TD) is a very rare disease. In most cases, it is asymptomatic. A 79-year-old woman visited outpatient clinic due to recent worsening of difficulty in breathing. On computed tomography of the lung, the H-shaped trachea-connected diverticulum with a height of 4.4 cm in the left-posterior wall of the trachea was noted. This unusual form of TD was considered to be more likely congenital than acquired form. So, we report this case with literature review to share with physician.

2. Introduction
Tracheal Diverticulum (TD) is a very rare disease that appears asymptomatic in most cases [1]. Symptoms are often nonspecific. So, TD is often found by chance in imaging test. In order to diagnose TD, chest Computed Tomography (CT) with three-dimensional reconstructive images can be used to confirm the location, origin, size and the shape of TD [1]. We report a case of TD with unusual form accidentally found on chest CT performed in a patient who presented to the outpatient clinic with chronic respiratory distress for a year.

3. Case
A 79-year-old woman who had been treated for tuberculous pleurisy 9 years ago, visited outpatient clinic and was admitted due to recent worsening of difficulty in breathing for a year. The vital signs of the patient were stable. She had alert mental status, but showed chronic ill looking appearance. The laboratory finding was nonspecific. The breathing sound was changed during inhalation, which made physician suspect extra-thoracic variable stenosis. The heart sound was normal. Abnormal shadowing was suspected on the side of the trachea on Chest X-ray (Figure 1A). Cardiac markers and transthoracic echocardiography performed to distinguish respiratory distress caused by heart disease, showed normal findings. In pulmonary function tests, variable extra-thoracic obstruction was suspected (Figure 1B).

The diverticulum of the posterior wall of the trachea was suspected on the cross-sectional image of CT (Figure 2A). The three-dimensional reconstruction was performed in order to assess the size and location of TD. There was the H-shaped trachea-connected diverticulum with a height of 4.4 cm in the left-posterior wall of the trachea (Figure 2C). It was difficult to distinguish between TD and tracheo-esophageal fistula by chest CT. Esophagogram showed there was neither tracheo-esophageal fistula nor other deformity combined with TD.

The patient did not complain signs and symptoms of infection such as fever, cough, and sputum. The clinical manifestations were improved after nebulizer treatment with bronchodilator. Furthermore, surgical treatment was not proper option considering the patient's age. The patient showed no further problems at follow-up. If infection occurs repeatedly in the future, surgical resection should be considered.
Figure 1: (A) On Chest X-ray, abnormal air density was not definite near the main bronchus of the left lateral tracheal wall. (B) The type of variable extra-thoracic obstruction was suspected in the flow-volume curve of pulmonary function test.

Figure 2: (A) Diverticulum adjacent to the trachea of the posterior tracheal wall on Chest CT, axial image. (B) Diverticulum is communicated with the left lateral tracheal wall on Chest CT, coronal image. (C) H-shaped diverticulum measuring 4.4cm (yellow circled, white arrowed) connected to the trachea on three-dimensional reconstruction image. (D) There was no evidence of tracheo-esophageal fistula on esophagography; (E) enlarged three-dimensional reconstruction image of the diverticulum (white arrowed). (F) The diverticulum is similar to the nose band of the glasses.
4. Discussion

On the basis of the symptom and anatomical structure on CT scan, the cause of TD was considered to be more likely congenital than acquired form. However, the fistula that could be seen in congenital type was not found in this patient.

TD are caused by congenital or acquired factors [1] [2]. Kurt et al. [1] reported that TD is more common in men (64%) than in women (36%). The congenital TD was known to occur 4-5cm below the vocal cord or several cm above the tracheal carina and generally occurs on the right side of the trachea. The neck of congenital TD connected with the trachea is narrow and may be accompanied by other congenital deformity, such as tracheo-esophageal fistula [1]. Wong et al. [2] showed in meta-analysis that patients with other congenital anomalies were 15 times more likely to have a TD (odds ratio=14.89; 95% confidence interval 7.09-31.22). But this lady did not have other congenital anomaly in our case. Her TD with narrow neck was considered to be main reason why she had no recurrent infection when she was young in the light of her history.

In acquired form of TD, the neck is wide and associated with increased tracheal lumen pressure due to bronchial-pulmonary disease with chronic cough, such as chronic obstructive pulmonary disease. Although this lady presented respiratory symptoms for some time recently, CT imaging showed that the junction was not wide enough to make diverticulum and the unusual shape of the diverticulum did not look like a sac. That is why it is unreasonable to regard it as a lesion secondary to chronic respiratory problem.

Sharma et al. [3] reported that small air collection in the para-tracheal region in the iatrogenic case. In our case, the air density was not definite in Chest X-ray image. For diagnosing a tracheal diverticulum, chest CT was known as proper option to evaluate the location, origin, size of diverticulum and communication between the diverticulum and trachea [4]. It is known that TD can cause complications like pneumothorax or abscess during assisted ventilation if undetected [3, 5]. In our case, ventilation was not needed because she improved after conservative treatment.

Most diverticula are asymptomatic. Treatment is not necessary in asymptomatic patients [6, 7]. The medical treatment such as antibiotics and mucolytic agents and/or surgical resection should be considered in case of TD with clinical symptoms [8]. Before surgery, patient’s age, performance, and the response to conservative treatment should be checked [8]. In this case, surgical treatment was not an option considering she was old and her symptoms improved in response to conservative treatment. The patient was followed without further problem.

5. Conclusion

In this case, the H-shaped probable congenital TD was identified on CT, which was performed because airway stenosis was suspected based on coarse breathing sound during inhalation. The conservative treatment improved her symptoms. Therefore, surgical treatment was not required. Hopefully, this unusual image case will help clinicians discover the TD in CT scan.

References