Uncommon Cause of Recurrent Pneumonias Enormous Tracheal Diverticulum with Fistula Case Report - Diagnostic Part

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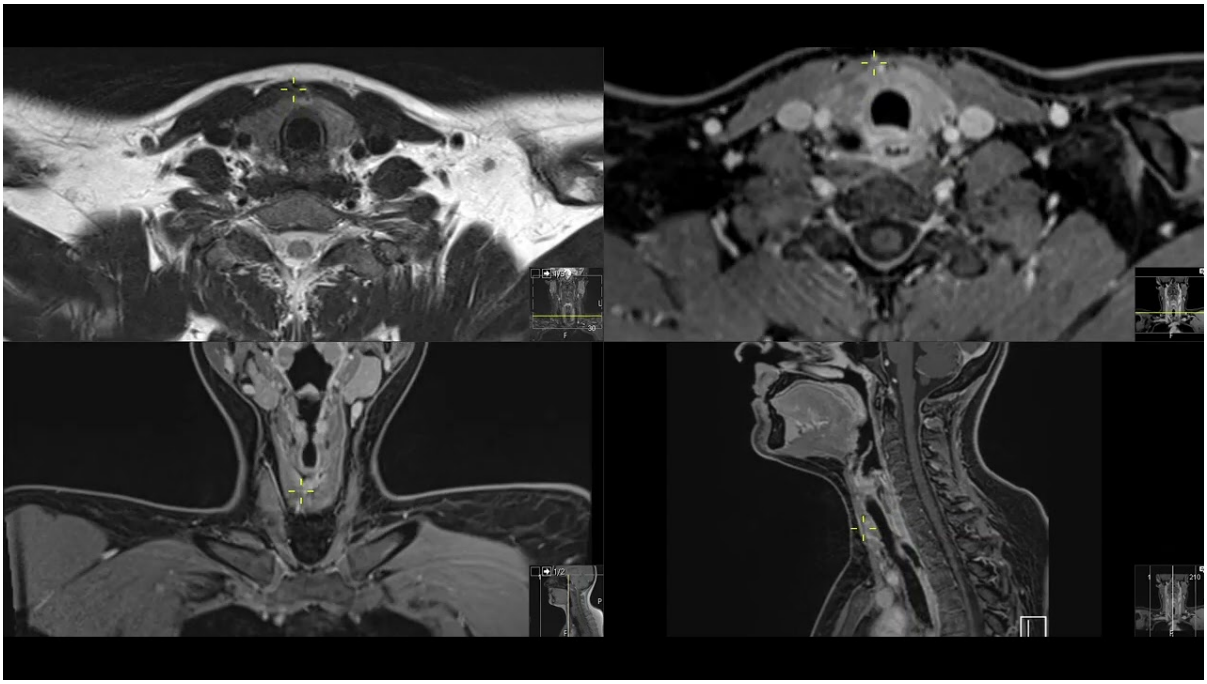
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received: 02.09.2023 - **published:** 30.09.2023

Swiss Journal of Radiology and Nuclear Medicine - www.sjoranm.com - SJORANM GmbH - CH-3072 Ostermundigen bei Bern - Switzerland



Abstract

A patient who presented with sudden shortness of breath, stridor, and recurrent laryngeal nerve paralysis was diagnosed with a tracheal diverticulum. The diagnosis was established through CT scans and confirmed via endoscopy. Distinctive CT findings revealed an air-filled, tubular structure located behind and slightly to the right lateral of the trachea, which communicated with it. Endoscopy demonstrated a narrow-mouthed diverticulum originating from the posterior right wall of the trachea, approximately 8 cm below the level of the true vocal cords. Successful medical treatment was administered, and the patient experienced an uncomplicated recovery.

Keywords: recurrent pneumonia, dysphagia, tracheal diverticulum, tracheal pseudo-diverticulum

Introduction

The diagnosis of a tracheal diverticulum dates back to its first description by Mr. Carl Freiherr von Rokitanski in 1838, which at that time was only possible during an autopsy. Today, it is the imaging techniques, primarily Multi-detector Computed Tomography (MDCT) followed by Magnetic Resonance Imaging (MRI) and projection

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radiography, that can detect these forms of the condition in vivo.

A distinction is made between a genuine, meaning congenital, tracheocele and the acquired tracheocele. Most often, they are incidental findings in thoracic diagnostics using CT. Their size varies between 0.7 - 1.9 cm. They are predominantly clinically asymptomatic. Only when there is an infection of tracheal mucus within the diverticulum, abscess formation is not uncommon. Very rarely, there is also a direct fistula to the trachea. This can lead to the spread of pathogenic bacteria via the fistula into the trachea and then into the bronchi and alveoli, resulting in pneumonia.

Therapeutic options include:

-Medication-based antibiotic therapy
-Surgery in individual cases or via endoscopy
-Radiologically minimally invasive abscess drainage

These treatment approaches aim to prevent further bacterial passage from the trachea in cases of laryngitis, tracheitis, or existing pneumonia.

Case Report:

We report on the case of a 36-year-old patient with recurrent pneumonias in the presence of an unusually large tracheal diverticulum in combination with a fistula to the trachea and consequent abscess formation.

Medical History:

The 36-year-old patient with recurrent infections in the years 2021 and 2022 presented herself in July 2023 at the outpatient clinic of the University Hospital of Inselspital in Bern, Department of Ear, Nose, and Throat Diseases, due to difficulty swallowing. The referral was made due to increased susceptibility to infections in the recent past, with symptoms such as fever, dysphagia, sore throat, yellowish sputum, and a sensation of a foreign body in the larynx. In recent weeks, there have even been difficulties swallowing larger tablets. There is no reported dysphonia or dystonia.

Clinical Findings:

Following adenotonsillectomy in childhood, there are no symptoms explaining the clinical presentation, with a normal inspection of the oro- and hypopharynx. Laboratory tests do not provide a definitive diagnosis. However, there is a homozygous alpha-1-antitrypsin deficiency. Specific antibodies against pneumococci are completely absent.

Radiological Findings:

An esophagus barium swallow test with regular and timely contrast passage (Fig.1) does not provide an explanation for the patient's complaints. The contrast-enhanced computed tomography (CT) indicates a paratracheal abscess in the upper mediastinum (Fig.2) as well as a tracheal diverticulum with a clear fistula extending to the trachea (Fig.3).

The MRI examination of the neck region and upper thorax confirms the findings visible in the CT (Fig.4 and 5).

Discussion:

Tracheal diverticula are protrusions of the layers of the tracheal wall into the surrounding tissues. They are found adjacent to the trachea, primarily on the right side, depending on the imaging modality used, such as DMCT, MRI, and less commonly with tracheoscopy, bronchoscopy, and in autopsies, as incidental findings (1). The frequency varies depending on the examination method, ranging from 0.75 to approximately 8% (2,3,4,5), with a localization of 97-98% on the right paratracheal



Fig.1 fluoroscopic biphasic esophagram

tracheal diverticula, involving a breach in the tracheal wall, can develop, for example, in the context of evolving COPD, after repeated violent coughing attacks, or very rarely after blunt trauma (1, 2, 3).

Tracheal diverticula are mostly clinically inconspicuous and are often detectable as incidental findings on CT scans of the neck/thorax (1). In isolated cases, infections have been reported due to the accumulation of mucus in the diverticulum, irritation of the vagus nerve, recurrent laryngeal nerve paralysis (6,7), or complaints in the upper respiratory tract and difficulty

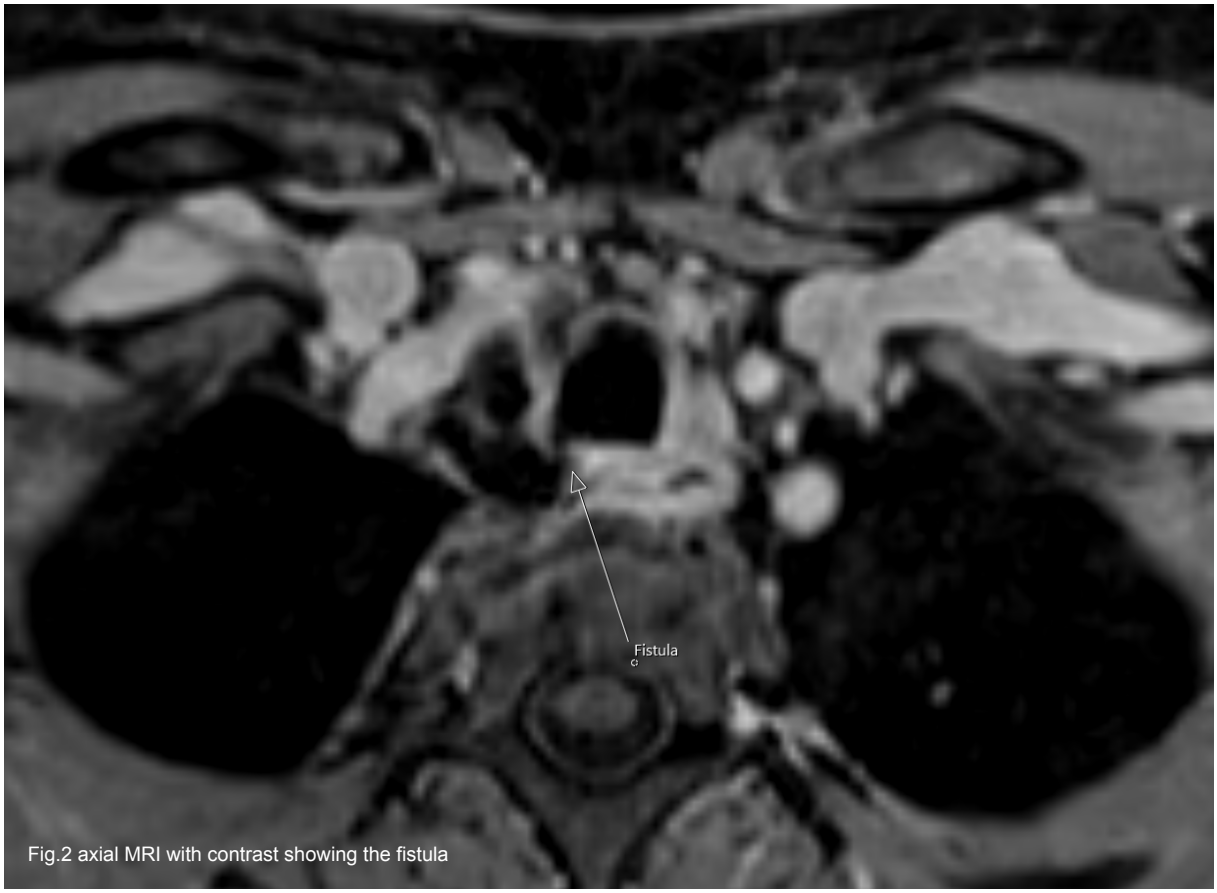


Fig.2 axial MRI with contrast showing the fistula

side (6) and a much less common 2nd location at the level of the carina tracheae. A distinction is made between congenital tracheal diverticula, also known as tracheoceles, and acquired tracheal diverticula (1). In the English-speaking scientific literature, they are more accurately described as Paratracheal Air Cysts (PTCA) (1). Acquired

swallowing (1,2,3,8,9). In rare cases, right-sided paratracheal air can be misinterpreted as an incipient tension pneumothorax. Tracheal diverticula with evidence of a fistula to the trachea, as seen in our patient on MDCT and MRI, have been very rarely described so far. The dimensions of the diverticulum in our patient, measuring 19 x 24 x 33



Fig.5 coronal MRI with contrast showing the diverticulum (see arrowhead)

mm, are unusually large, as most CT-detected cases are described as significantly smaller (1) (Fig.2). Yuan Zang et al. demonstrate in their study comparing the walls of non-infected diverticula in 25 examined patients by MRI, with diameters ranging from 3.0 to 15.0 mm (average: 8.5 mm), and by CT, with diameters ranging from 2.8 mm to 14.7 mm (average 7.8 mm) (12). This slight size difference in the examination of the same patient population is attributed to the different imaging capabilities of the two imaging modalities. The determined sizes clearly show that the size of our described diverticulum, compounded by the presence of a fistula to the trachea, is exceptional. This rare fistula formation could well be considered a possible cause for the pneumonia reported in the patient's recent medical history.

The space-occupying effect of the large diverticulum with moderate displacement of the esophagus to the left likely explains the increasing swallowing difficulties reported by the patient recently.

Specific Medical Conditions:

Some diseases are associated with not just a single tracheal diverticulum. Tracheal diverticula are more frequently observed in cystic fibrosis (10) and Mounier-Kuhn syndrome, an idiopathic tracheobronchomegaly (11,13). This syndrome occurs with a prevalence of 0.4-1.6% according to Krustins (14) in individuals aged between 30 - 40 years. It is eight times more common in men compared to women and presents clinically with recurrent infections. Additionally, these patients often have multiple tracheal diverticula (15,16).

In comparison, the combination of an unusually large tracheal diverticulum with an existing fistula to the trachea, infection of the diverticular contents primarily diagnosed as a paratracheal abscess causing swallowing disorders due to its size, and as a possible cause of recurrent pneumonias in the patient's medical history, as found in our patient, has not been described to date in this manner according to our last PubMed research from July 20, 2023.

Further Outlook

We will discuss various possible treatment approaches in the second part of this case report and provide updates on the clinical course of our patient. As soon as possible we will report on the further course and the decision of the patient regarding which therapy approach she ultimately chose. That article will be the continuation of this Case Report as a second part with the new subtitle *Therapeutic Part.*

Conflict of interest:

The authors declare that there were no conflicts of interest within the meaning of the recommendations of the International Committee of Medical Journal Editors when the article was written.

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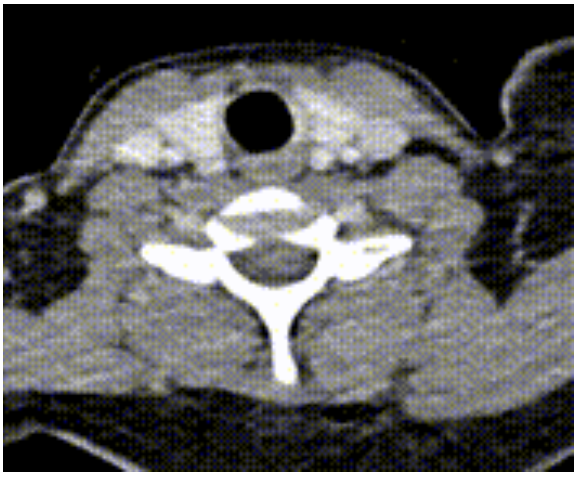


Fig.4 axial CT in soft and airways window

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