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Severe tracheal stenosis with tracheopathia osteoplastica-like changes due to traumatic blunt injury

Kazutaka Kakinuma[1], Kei Morikawa[1], Yasuhiro Miyamoto[2], Hisashi Saji[3], Masamichi Mineshita[1] & Teruomi Miyazawa[1]

[1]Department of Internal Medicine, Division of Respiratory and Infectious Diseases, St. Marianna University School of Medicine, Kawasaki, Japan
[2]Department of Otorhinolaryngology, St. Marianna University School of Medicine, Kawasaki, Japan
[3]Department of Chest Surgery, St. Marianna University School of Medicine, Kawasaki, Japan

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Correspondence
Teruomi Miyazawa, Department of Internal Medicine, Division of Respiratory and Infectious Diseases, St. Marianna University School of Medicine, 2–16-1 Sugao, Miyamae-ku, Kawasaki 216–8511, Japan.
E-mail: miyazawat@marianna-u.ac.jp

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Abstract

A 17-year-old man was injured in a motorcycle accident, leading to a complex cerebral contusion and hepatic injury. Approximately one month after being discharged from the hospital, the patient experienced gradually progressive dyspnea on exertion. Chest computed tomography revealed severe upper tracheal stenosis; thus, emergency tracheotomy and subsequent tracheoplasty were performed. Microscopically, the deformation of tracheal cartilage and extensive interstitial ossification/fibro-elastic changes were observed. To our knowledge, this is the first report documenting the ossification of the trachea that rapidly progressed after injury, which was confirmed by surgical resection of the upper trachea.

Introduction

Approximately 60% of tracheal blunt injuries occur from traffic accidents [1]. We report a case of severe upper tracheal stenosis resulting from a traffic accident. This condition was missed on the first admission, and identified after the patient experienced progressive dyspnea. Pathological evaluation revealed an unusual, rapid-growing ossification of tracheal cartilages. Here we report this rare condition with some indications for the early detection, treatment, and follow up.

Case Report

A 17-year-old man, without notable past or familial history, was injured in a traffic accident while driving a motorcycle. The patient's injuries comprised a complex cerebral contusion and hepatic trauma with severe intra-abdominal bleeding. The patient was immediately intubated and hepatic artery embolization was performed. The general condition gradually improved during the first week after admission. However, approximately one month after being discharged from the hospital, he experienced dyspnea on exertion, which gradually worsened. The patient complained of difficulty sleeping in supine position and instead felt better in prone position. Upper tracheal stenosis was diagnosed by chest X-ray (Fig. 1A) and chest computed tomography (CT) at a local hospital, and he was immediately referred to our institution.

Vital signs were considered normal on admission, but his breathing was retractive and stridor was noted. Plain and three-dimensional chest CT on the admission revealed a 27-mm long segment stenosis of the trachea of which the shape was similar to a bottleneck (Fig. 1B, C). At the time of admission at our institute, the airway was successfully secured with fiber-optic assisted intubation and emergency tracheotomy (Fig. 1D). Subsequently, tracheal resection and primary reconstruction were performed. Tubular resection of the first to fourth tracheal cartilages and end-to-end anastomosis without tracheal release were performed.

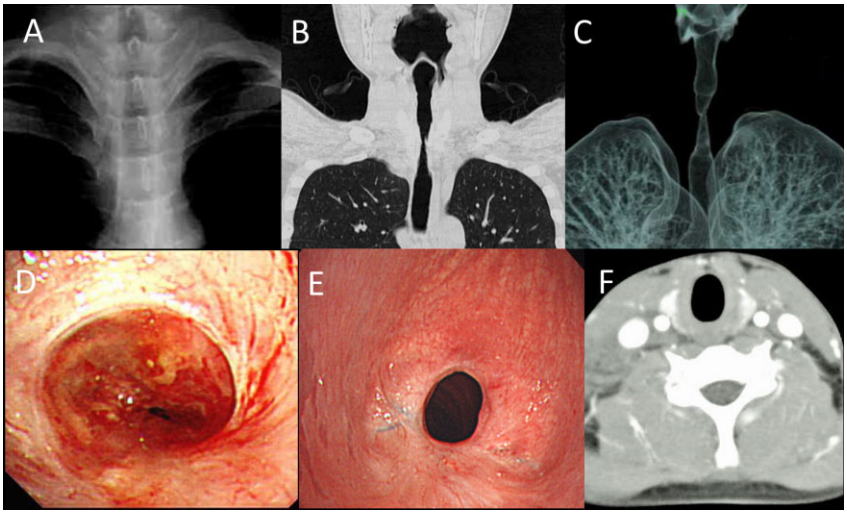

Figure 1. (A) Chest radiograph 90 days after injury showed upper tracheal stenosis. (B) Chest computed tomography (CT) on admission demonstrated a long segmental stenosis below the vocal cords. (C) The stenosis was visualized with a three-dimensional CT on admission showing a bottle neck pattern with a length of 27 mm. (D) Bronchoscopic findings of emergency tracheotomy showed a pinhole stenosis with adhesion and deformation of tracheal cartilage. (E) Airway patency was confirmed by bronchoscopy 6 months after surgery. (F) The initial CT taken at the previous hospital showed tracheal swelling at the upper area of the stenosis.

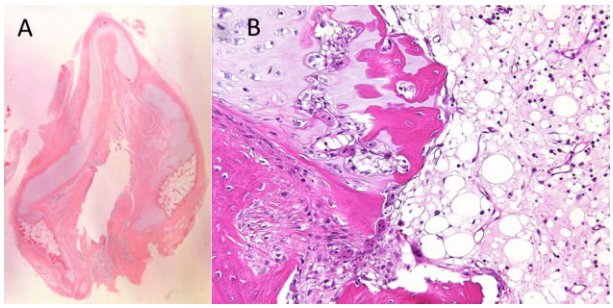

Figure 2. (A) Pathologic findings confirmed the deformation of tracheal cartilage (hematoxylin and eosin stain [H&E], loupe magnification). (B) Histopathological findings of the trachea demonstrated a deformation of the tracheal cartilage and an extensive interstitial ossification/fibro-elastic changes (H&E, original magnification ×200).

Fracture of the second and third tracheal rings was found at surgery.

After confirming the airway patency by postoperative bronchoscopy on the following day, extubation was successfully performed. In pathological findings, a deformation of the tracheal cartilage and an extensive interstitial ossification/fibro-elastic change of the trachea were detected (Fig. 2A, B). Although the trachea still remains stenotic (Fig. 1E), the patient is currently free of symptoms.

Discussion

Generally, pathological tracheal ossification is observed in rare disorders including tracheobronchopathia osteochondroplastica (TO) and relapsing polychondritis (RP). TO, for which patients are usually over 50 years old and etiology still remains unclear, is a slowly progressive ossification of tracheal cartilages [2]. Although typical clinical features of RP are recurrent pain and swelling of the external ears and nose, CT of RP patients also shows frequent subglottic stenosis and thickening of the airway walls [3]. Uncommonly, cartilages involved by RP exhibit a chronic ossification/fibrosis as well as inflammation. Ossification of the thyroid and arytenoid cartilage in an RP patient has been reported [3]. The acute clinical presentation of the present case, which results in the extensive and rapid tracheal ossification in a span of 3 months, is different from those diseases mentioned earlier.

We also suspected post-intubation tracheal stenosis because of its clinical course. Post-intubation tracheal stenosis usually involves the site where intubation tube cuff contacts tracheal mucosa. Hence, the stenosis is usually limited to a short length of trachea. In this case, the stenosis was observed at the long segment of subglottic area (the cuff was at the lower trachea) and the lesion extended to the depth of tracheal cartilage as well as tracheal mucosa.

Currently, there has been no report describing a case with severe airway stenosis and dyspnea caused by tracheal cartilage ossification with hyperplastic granulomatous formation after a traumatic blunt injury. Although clinicopathological and molecular genetic studies on larger number of cases would be necessary to clarify the pathogenesis of the present case, we hypothesized that the increased volume of granular tissue that contains many undifferentiated mesenchymal cells could cause bone tissue

formation during the reparation after the destruction or fracture of tracheal cartilage.

The initial CT taken at the local hospital clearly showed tracheal swelling at the upper area of the stenosis (Fig. 1F), which might have been an initial sign for traumatic blunt injury of upper trachea. However, neither tracheal cartilage deformation nor obvious airway stenosis was observed at this time. Therefore, it was difficult to predict that this injury would develop into a severe stenosis. For early detection of airway stenosis in the present case, it might be useful to check patient's clinical manifestation carefully. Changes in the degree of stenosis are often induced by respiration, cough, or body position [4]. Especially respiratory conditions may vary depending on how the head is positioned in cartilage fractures. In this case, the patient slept in the prone position. This action suggested a severe stenosis in airway, and chest CT revealed a bottle neck-type stenosis in the upper trachea.

Indications for the treatment in tracheal stenosis are based on the degree of cartilage damage of airway [5]. As the patient had difficulty breathing on admission, a team of chest surgeons and otolaryngologists performed emergency tracheotomy and subsequent surgical tracheoplasty. In this case, end-to-end tracheal anastomosis was appropriate as four tracheal cartilages wholly became deformed. As tracheal resection extended to four cartilaginous rings, the lower trachea was lifted and the stenosis remained at the anastomosis site. However, the patient is currently free of symptoms and we confirmed airway patency has not worsened by bronchoscopy after surgery. Careful observation will be necessary for several years.

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Disclosure Statements

No conflict of interest declared.

Appropriate written informed consent was obtained for publication of this case report and accompanying images.

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