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Case Report

Tracheal Stenosis Caused by Unnoticed Foreign Bodies

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We describe an extremely rare case of tracheal stenosis caused by unnoticed microscopic fiber-like foreign bodies. A 66-year-old woman complained of dyspnea with inspiratory stridor. Magnifying electroendoscopy and computed tomography revealed stenosis involving the entire circumference of the tracheal lumen. Tracheotomy and biopsy were performed. Histologically, the lesion showed chronic inflammation with a deposition of fiber-like foreign bodies. The patient had no history of trauma or inhalation injury, but had undergone intratracheal intubation on 4 occasions. The lesion was incised using semiconductor laser photoresection, and the postoperative course was good. To the best of our knowledge, this represents the first report in the English literature of tracheal stenosis caused by unnoticed foreign bodies. The origin of these fiber-like foreign bodies remains unclear but might be related to chronic inflammation resulting from intratracheal intubations.

Key words: tracheal stenosis, fibrous foreign body, intubation, tracheotomy, laser

Tracheal stenosis is usually divided into 2 groups: congenital and acquired. Acquired stenosis is caused by trauma, infectious diseases, tumor, collagen disease, or idiopathic disease [1]. The most common cause of tracheal stenosis is trauma from endotracheal intubation [2, 3]. Subglottic stenosis has been attributed to reparative fibrosis following particularly severe acute intubation injury, and another factor may be delayed healing of the subglottic mucosa, possibly exacerbated by full-thickness necrosis of the cricoid cartilage [4]. Tracheal foreign bodies, which usually arise from inhalation injury, may also cause tracheal stenosis [5]. A few cases of

tracheal stenosis caused by iatrogenic foreign bodies such as surgical gauze have also been reported [6]. We describe a rare case of tracheal stenosis presumably caused by infiltration of fibrous foreign bodies during intratracheal intubations performed more than 30 years earlier.

Case Report

A 66-year-old Japanese woman presented with a 1-month history of inspiratory stridor. Despite treatment with anti-asthmatic medications at another clinic, her dyspnea worsened. The treatments for "bronchial asthma" included administration of prednisolone,

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tulobuterol patch, and inhaled corticosteroid. On presentation to our department, findings for the larynx were unremarkable, but concentric narrowing of the tracheal lumen causing >75% obstruction and leaving a tiny patent trachea was observed under magnifying electroendoscopy (Fig. 1). Computed tomography revealed involvement of the entire circumference of the trachea at the level of the cricoid cartilage and extending 1.0cm caudally (Fig. 2). The distal bronchial tree was normal. Emergent tracheostomy was performed under local anesthesia. The patient's medical history was unremarkable except that she had had 4 Caesarean sections under general anesthesia, with the last occurring when she was 36 years old. The patient was a full-time housewife and had no relevant occupational history. The patient also had no relevant history of habitual or environmental factors that would have exposed her to fibers (e.g., smoking, bonfires, pets, or exposure to fiberglass and/or cellulose insulation in the home). Laboratory testing revealed a white blood cell count of 7,200/ μ L

(normal, 3,100–9,200/ μ L) and a serum C-reactive protein level of 0.09mg/dL (normal, 0–0.3mg/dL). The differential white blood cell counts were: neutrophils, 68.5% (normal, 40–67%); basophils, 0.1% (normal, 0–1%); eosinophils, 0.1% (normal, 1–5%); lymphocytes, 24.3% (normal, 24–48%); and monocytes, 6.8% (normal, 3–9%). Testing for proteinase 3 anti-neutrophilic cytoplasmic antibody, myeloperoxidase anti-neutrophilic cytoplasmic antibody, and whole-body interferon-gamma enzyme-linked immunosorbent assay all yielded negative results. Serum level of squamous cell carcinoma-related antigen was 1.5ng/ml (normal, 0–1.5ng/ml). Biopsy of the stenotic area was performed under general anesthesia using the laryngoscope. Histological examination showed granulation tissue (Fig. 3A). Some fragments of fiber-like foreign bodies were embedded in the granulation tissue (Fig. 3B). Polarized light highlighted these foreign bodies with birefringence, which were scattered across the whole specimen (Fig. 3C, D). This foreign body deposition was suspected to

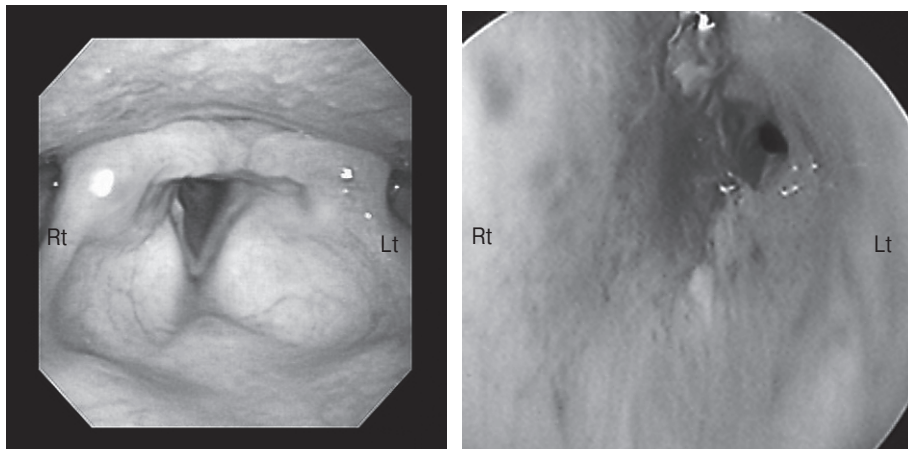


Fig. 1 Magnifying electroendoscopy reveals good mobility of the vocal cords and concentric narrowing of the tracheal lumen by >75%.

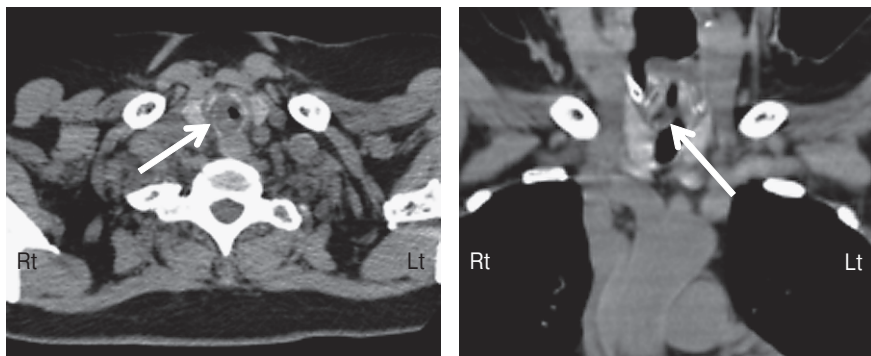


Fig. 2 Computed tomography reveals stenosis of the entire circumference of the trachea over a length of 1.0cm. Left, axial; right, coronal.

have caused an inflammatory reaction. The endotracheal stenotic rings were incised by semiconductor laser photoresection using flexible bronchoscopy under general anesthesia. Administration of prednisolone (PSL) was started soon after the operation at 60 mg/day, then tapered by 5-10 mg/day every 2-3 days. The postoperative course was good, and the tracheal cannula was removed 1 year postoperatively. No relapses have been observed in the 2 years after the patient was discharged from the hospital (Fig. 4).

Discussion

Tracheal stenosis is usually encountered as an

acquired inflammatory lesion, typically due to prolonged intubation and tracheostomy using a high-pressure cuffed endotracheal tube or tracheostomy tube. In the present study, the patient had a history of four Caesarean sections under general anesthesia. Although the duration of each of those intubations might not have been long, some studies have mentioned that even 24 h of intubation is sufficient to result in this complication; moreover, in some cases with this complication, the patients were intubated so long in the past that they did not remember it [7]. In addition, scar and granuloma formation producing stenosis may worsen every time a new event is added [5]. However, each of the intubations in the present

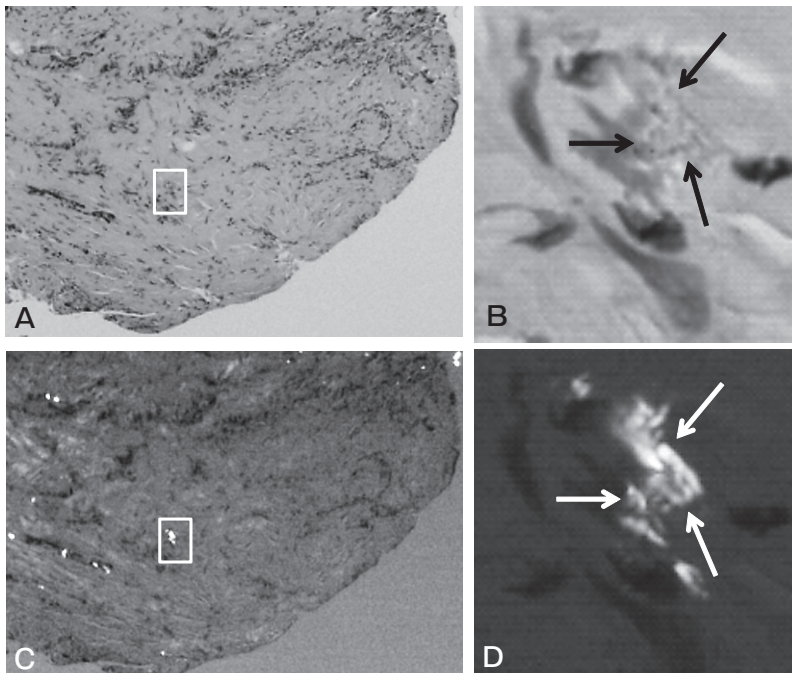


Fig. 3 A, Histological study shows granulation tissue; B, Some fragments of fiber-like foreign bodies are embedded in the granulation (arrows); C, D, Polarized light highlights these foreign bodies with birefringence (arrows).

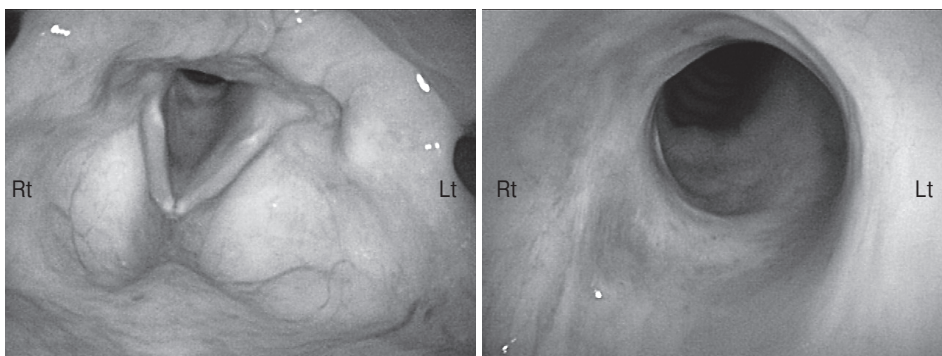


Fig. 4 Magnifying electroendoscopy 2 years after discharge from the hospital reveals no relapse has occurred.

case might have lasted much less than 24h, and the time lag from the last intubation to the onset of this complication was 30 years. We therefore concluded that the entry of foreign bodies into the tracheal lumen had occurred at the time of endotracheal intubation, causing tracheal stenosis. We speculated that the stenosis might have progressed little by little for a long time, and at last asthma-like symptoms appeared. Most cases with tracheal stenosis caused by a foreign body are caused by inhalation injury [2, 5]. To the best of our knowledge, the entry of foreign bodies into the tracheal lumen has not been reported in cases with subglottic stenosis caused by endotracheal intubation [8–10]. Endotracheal sutures during thoracic surgery can form foreign bodies causing tracheal stenosis, and symptoms may arise 2–25 years postoperatively [11]. The origin of the fiber-like foreign bodies in the present study remains obscure, but some microscopic fibrous tissue might have been attached to the intubation tube.

Various options for treating tracheal stenosis have been reported, including balloon dilatation, laser resection, argon plasma coagulation, stent placement, and resection of the stenotic lesion with end-to-end anastomosis of the trachea [12]. Myer *et al.* reported that an operation is required for stenosis involving >71% obstruction of the lumen [13]. Zias *et al.* reported that the general approach to manage stenosis should be nonsurgical, reserving the surgical option for recurrent and refractory cases [14]. Use of steroids has been reported as a predisposing factor for the development of tracheal stenosis, because of its effects on wound healing [12], and systemic use of corticosteroids or antibiotics is probably not effective [14]. On the other hand, Braidy *et al.* suggested that topical and systemic corticosteroids might be useful in the management of early post-intubation tracheal stenosis [15]. In the present case, we performed radial incision by semiconductor laser photoresection, and administered corticosteroids soon after the operation. The tracheal stenosis healed, and no relapses have been observed in the 2 years after the procedure.

We have described a rare case of tracheal stenosis caused by fiber-like foreign bodies against a back-

ground of 4 intratracheal intubations many decades earlier. The origin of the fiber-like foreign bodies was unclear, but entry might have occurred during the intubations, causing chronic inflammation. To the best of our knowledge, this represents the first report in the English literature of tracheal stenosis caused by fibrous foreign bodies.

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