

Congenital Web-like Tracheal Stenosis Cured Surgically in Adult

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Received for publication, June 28, 1985.

A 60-year-old, Japanese woman, with congenital web-like tracheal stenosis surgically treated. Successful relief from tracheal stenosis was obtained.

Detection in adulthood and chance of the treatment for congenital tracheal stenosis is very rare. As far as this case is concerned, delay in detection and treatment is discussed of congenital tracheal stenosis.

INTRODUCTION

It is well known that tracheal stenosis arises from various causes. Sustained tracheal stenosis in congenital origin cured in adulthood is very rare in frequency.

Tracheal stenosis is ominous for life saving. In advances of surgery, successfully treated cases are increasing in number. We now report an experience, with surgical treatment for tracheal stenosis in congenital origin.

CASE REPORT

A 60-year old Japanese woman was admitted on July 25, 1984 for the treatment of varial obstruction. On her preoperative pulmonary function test, obstructive pulmonary

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dysfunction was pointed out and the flow volume curve (Fig. 1) at exspirum revealed a reduction in peak flow, demonstrating a presence of the upper air way stenosis. A result of arterial blood gas analysis showed somewhat high PaCO₂ value as compared with the normals.

Chest xp disclosed a shadow of the thickened pleura in the bilateral apex, an increase in bronchovascular shadow on the right upper field, an enlargement of the hilar shadow and a calcified shadow of the aortic arch.

Tomogram (Fig. 2) also revealed a circumscribed stenosis of the trachea, 1cm long,

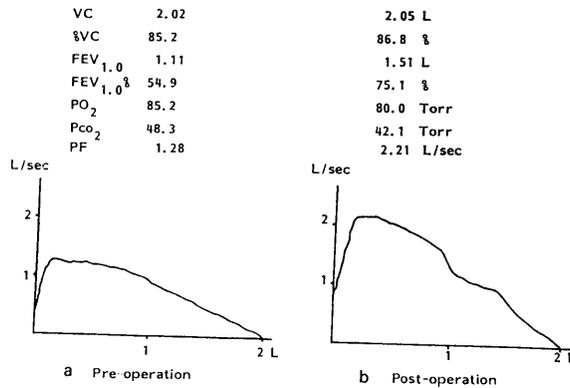


Fig. 1 Flow-volume curve prior to surgery.

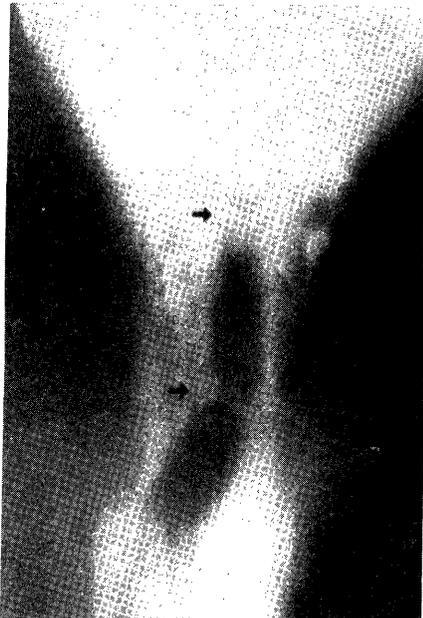


Fig. 2 Tomogram, showing a tracheal stenosis in the cervical portion before surgery.

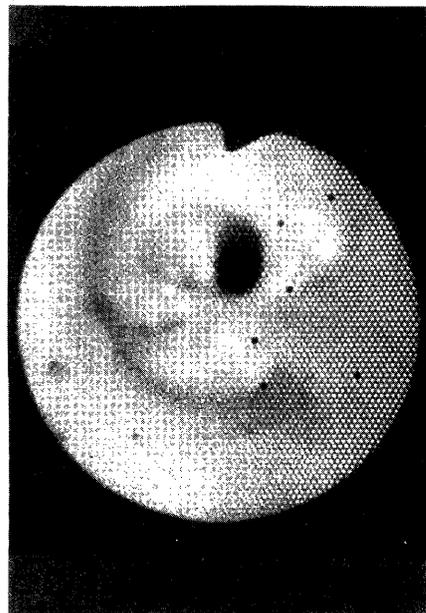


Fig. 3 Preoperatively bronchoscopic finding, demonstrating a circular stenosis of the trachea.

4cm below the subglottis. On the fiberbronchoscopic examination using a Olympus BF-4B2, a circular stenosis of scar formation was defined as being round in shape, smooth in surface as shown in Fig. 3. It is impossible to introduce the fiberscopic tube distally across a stenotic portion. Bronchogram (Fig. 4) showed a stenosis of 1cm long which was of a protruding lesion on the right wall and of a concaving one on the left on the AP view and a funnel shape stenosis of 1.5cm long on the anterior wall and 1cm long on the posterior wall on the lateral view. On September 1, 1983, the cervical trachea was surgically exposed under local anesthesia to remain the airway open. After the trachea with a 3cm stenosis was removed just below the cricoid cartilage, a new tracheal tube was introduced into the distal trachea and rapid induction of anesthesia was achieved. Both cut edges of the trachea were seen intact, not remaining stenotic lesion at all.

The tension-free anastomosis was made possible at a line of the subglottis without tracheal mobilization, using a tracheal tube previously introduced from the operative field at first, followed by use of orotracheal tube after the posterior wall was carefully sutured. The membranous portion of the distal trachea was lessened by suturing to make it fit to the size of the subglottis space. Tracheal anastomosis was completed using the interrupted sutures of 3-0 Dexon without trouble.

Postoperative course was uneventful. Postoperative flowvolume curve was more improved rather than the control as shown on the left in Fig. 1. On the bronchoscopy 1 month postoperatively no stenosis in the anastomotic site was revealed. Surgical specimen (Fig. 5) showed a stenosis of 17mm in diameter, a maximum of 5mm. The cartilage tissues were not affected. Histologic findings (Fig. 6) indicated that stenotic portion was covered with squamous epithelium, which was containing cell infiltration and edema ac-

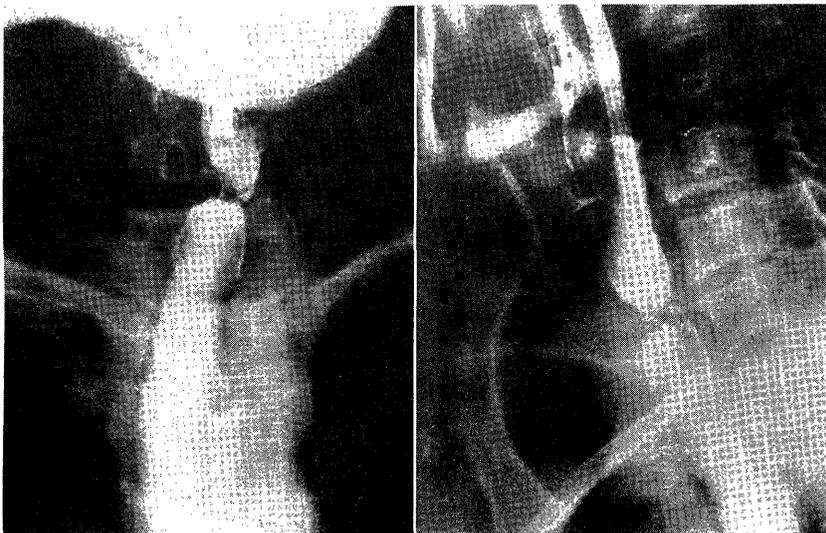


Fig. 4 Preoperative bronchogram, showing a tracheal stenosis on the AP and lateral views.

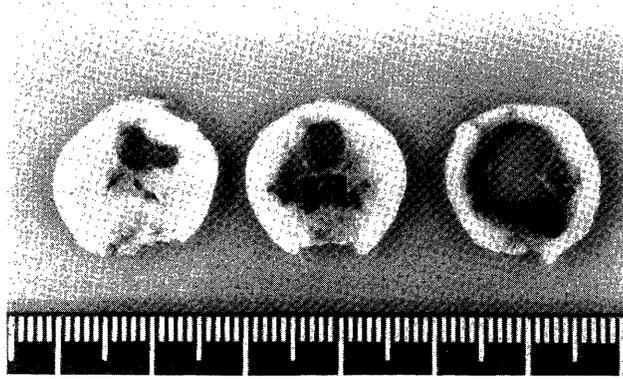


Fig. 5 Histologic finding of a stenotic trachea resected, showing overgrowth of fibroconnective tissues.



Fig. 6

companying the bronchial glands and mainly comprised overgrowth of fibroconnective tissues. It is definitively suggestive of congenital origin.

DISCUSSION

The causes of tracheal stenosis were considered to be inflammatory, traumatic neoplastic and iatrogenic. In the case reported herein, traumatic and iatrogenic origins out of etiologic causes of tracheal stenosis were excluded from her past history, and also it was possible to rule out a neoplastic origin from its histologic features. Plausible explanation for etiology of tracheal stenosis is of inflammatory origin.

Etiologic consideration with respect to inflammatory origin of tracheal stenosis is

generally divided into two, specific and non-specific.

Specific inflammation such as syphilis, tuberculosa, diphtheria, and typhoid are not accepted in this case from her past history. Diffuse stenosis of the trachea is usually so prominent in non-specific inflammatory origin that a localized tracheal stenosis in this case is not compatible with its origin. We are reasonably considered congenital in origin. It, however, is questionable that the symptoms of tracheal stenosis are mild in childhood and not pronounced with age. Of interest is that its onset is a delay. We draw a conclusion with respect to etiology that congenital web stenosis has become progressed by repeated inflammation with age. It is reported that many of cases with tracheal stenosis in congenital origin have an associated anomaly in the cardiovascular respiratory systems, and extremity.

HOLINGER reported a case who has web like tracheal stenosis of unknown etiology. Congenital tracheal stenosis is not common even in the world literatures. Furthermore detection in youth is very rare. There are few cases who are detected and treated at age 60 in our seeking literatures in which only 5 cases over 16 years old, not reaching 30 years old are detected.

The stenotic types were divided by WOLMAN, Pallachy and Cantrell. Fibrous stenosis is common in congenital origin. According to HOLINGER' classification, fibrous stenosis of tracheal segment is strongly coincident in the pathogenesis of its origin. We consider that main cause is segmentally developmental disturbance of the trachea in the course of recanalization in fetus as cited by Mc. MILLAN. This case is interesting with respect to delay in appearance of symptom and also in surgical treatment.

Addendum: This case has been reported by OKA M *et al.* in Japanese.

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