Congenital Right Intermediate Bronchial Stenosis With Carina Trifurcation: Successful Management With Slide Tracheobronchial Plasty

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Congenital bronchial stenosis is rarely described and is difficult to manage. Here we report two cases of right intermediate bronchial stenosis (stenotic orifice with complete cartilage rings). Both cases were associated with ventricular septal defects, and a “trifurcation” pattern was found in both carinas. Both patients underwent surgical repair of the ventricular septal defects but could not be separated from ventilator support despite successful cardiac operations. Slide tracheobronchial plasty was applied to the right intermediate bronchus and lower trachea. After correction of the bronchial stenosis, both patients could be extubated and live without supplementary ventilation support.

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air leakage before the patient was weaned from cardio-
pulmonary bypass. The bypass time was 67 minutes. She
was gradually weaned from the ventilator and suc-
cessfully extubated 17 days after the operation. She was
discharged 3 weeks later. A CT scan performed 2 months
later disclosed well-reconstructed trachea and bronchi
(Fig 2C). She has remained alive and well for 1 year
without any supplementary oxygen or mechanical
ventilation.

Patient 2
A 3-month old male infant was referred to our hospital for
respiratory distress and heart murmur. His body weight
was 3293 g (<3rd percentile). He had substernal retraction,
which persisted despite diuretic medication, and required
positive pressure ventilation support throughout the day.
Echocardiography revealed a perimembranous VSD
about 5 mm in size. Bronchoscopy and CT also revealed a
trifurcation pattern in the carina and a stenotic right in-
termediate bronchus from the ventral side of the carina,
quite similar to the condition in patient 1. As before,
we tried to repair the VSD as the first operation to see
whether respiratory conditions could be improved.
However, after the operation, carbon dioxide still accu-
mulated every time we decreased ventilator support. We
decided to repair the right intermediate bronchial ste-
nosis 10 days after the VSD repair. The surgical approach
was similar to that in patient 1, and slide tracheobronchial
plasty was performed smoothly. The patient was extu-
bated 6 days after operation and was easily discharged
2 weeks later. He has remained alive without medication
or respiratory support for 6 months after operation.

Comment
The association of tracheobronchial stenosis has been
reported as one of cause of failure to extubate after car-
diac operations in infants [4]. Congenital tracheal stenosis
is frequently associated with congenital heart defects, but
isolated bronchial stenosis has been much less frequently
reported. A recent report showed it was experienced by
only 2 of 27 patients at a medical center in 9 years [4].
Abnormal arborization of the trachea has also been
found in patients with congenital tracheal stenosis, and a

![Image](image_url)
trifurcation pattern was recently reported in the Great Ormond Street classification of congenital tracheal stenosis [5]. Our 2 patients were unique in the associated with VSD, isolated intermediate bronchial stenosis, and the trifurcation pattern of the carina. We did not find any similar combinations in the literature review.

The management of bronchial stenosis is more difficult than that of tracheal stenosis because of the small luminal diameter and the proximity of the lung parenchyma to the bronchial lesion [5]. The traditional approach to bronchial plasty included classic thoracotomy or thoracoscopy [6]. Bronchoplasty under the support of extracorporeal membrane oxygenation has also been reported [1, 7].

We performed the two procedures through a median sternotomy approach with cardiopulmonary bypass because we were already familiar with slide tracheoplasty and the correction of associated bronchial stenosis through this approach [8]. To achieve a good slide tracheobronchial plasty, the intermediate bronchus was mobilized by separating the surrounding hilar vessels. The right lobar arteries and the right upper pulmonary vein were dissected and gently retracted for exposure. Given that the lesion involved the carina, we performed the slide procedure across the carina up to the lower trachea.

The timing of operation in isolated bronchial stenosis has not been well studied. Some reports show that patients with right main bronchial stenosis can survive for several months [6]. Initially we tried to do a cardiac operation first, hoping that the symptoms would lessen and the patient could grow up to a bigger size. However, these two cases showed that the intermediate bronchial stenosis itself could be severe enough to result in dependence on a ventilator. The concomitant correction of the bronchial stenosis and the VSD might be a reasonable choice next time.

In conclusion, a unique pattern of congenital right intermediate bronchial stenosis with a trifurcation pattern in the carina could be a reason of prolonged intubation after surgical repair of VSD. Slide tracheobronchial plasty could achieve good results in infants.

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References
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