

## Bilateral Vocal Fold Abduction Dysfunction: A Report of Two Neonatal Cases

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Stridor is caused by oscillation of the narrowed upper airway. The most common cause of neonatal stridor is laryngomalacia, followed by vocal fold abduction dysfunction. Herein, we present two neonatal cases of idiopathic dysfunction of vocal fold abduction. A neonate was admitted to the neonatal intensive care unit (NICU) on day 4 of life for inspiratory stridor, intermittent subcostal retraction, and cyanosis. A second neonate was admitted to the NICU on day 7 of life for inspiratory stridor and cyanosis when crying. Neither patient had dysmorphic features or unusual cardiac ultrasonography findings. The diagnosis was confirmed by laryngo-bronchoscopy. Conservative treatment with biphasic positive airway pressure was effective in both cases and symptoms resolved within a few months. Resolution of vocal fold abduction dysfunction was confirmed by repeat endoscopy. Clinical manifestations of vocal fold abduction dysfunction vary widely. Although most cases resolve spontaneously, prolonged tube feeding, or even tracheostomy, is needed in some severe cases. Diagnosis of vocal fold abduction dysfunction requires a laryngo-bronchoscopy study; thus, there may be a large number of undiagnosed patients. Vocal fold abduction dysfunction should be considered in the differential diagnosis for neonatal inspiratory stridor. (*J Nippon Med Sch* 2024; 91: 249–251)

**Key words:** neonate, stridor, vocal fold abduction dysfunction

### Introduction

Stridor is caused by oscillation of the narrowed upper airway. The most common cause of neonatal stridor is laryngomalacia, followed by vocal fold immobility. We present two neonatal cases of idiopathic dysfunction of vocal fold abduction. Both cases were diagnosed by laryngo-bronchoscopy, and symptoms resolved after biphasic positive airway pressure (BiPAP) therapy. Written informed consent for publication was obtained from the patient's parents.

### Case 1

The first patient was born at 39 weeks and 6 days of gestation (birth weight 3,060 grams, −0.06 standard deviation) and was delivered via vacuum delivery because of the non-reassuring fetal status. His APGAR scores were 8 and 9 at 1 and 5 min, respectively. He was admitted to

the neonatal intensive care unit (NICU) on day 4 of life for intermittent subcostal retraction, inspiratory stridor, and cyanosis. These symptoms were present even at rest; thus, laryngo-bronchoscopy was performed. There was no evidence of dysmorphic features, and cardiac ultrasonography revealed no signs of congenital heart defects, including vascular rings. There was no hoarseness, hypercapnia, desaturation, or feeding difficulties. An endoscopic study on day 7 revealed oropharyngeal narrowing, glossoptosis, and mild tracheomalacia. Conservative treatment with continuous positive airway pressure (CPAP; positive end-expiratory pressure [PEEP]: 5 cm H<sub>2</sub>O) was initiated. A follow-up laryngoscopic study conducted 4 weeks later showed minor improvement and additional findings, including intermittent bilateral vocal fold abduction dysfunction when crying (**Fig. 1**). Hence, the patient was switched to BiPAP at 8 cm H<sub>2</sub>O to pre-

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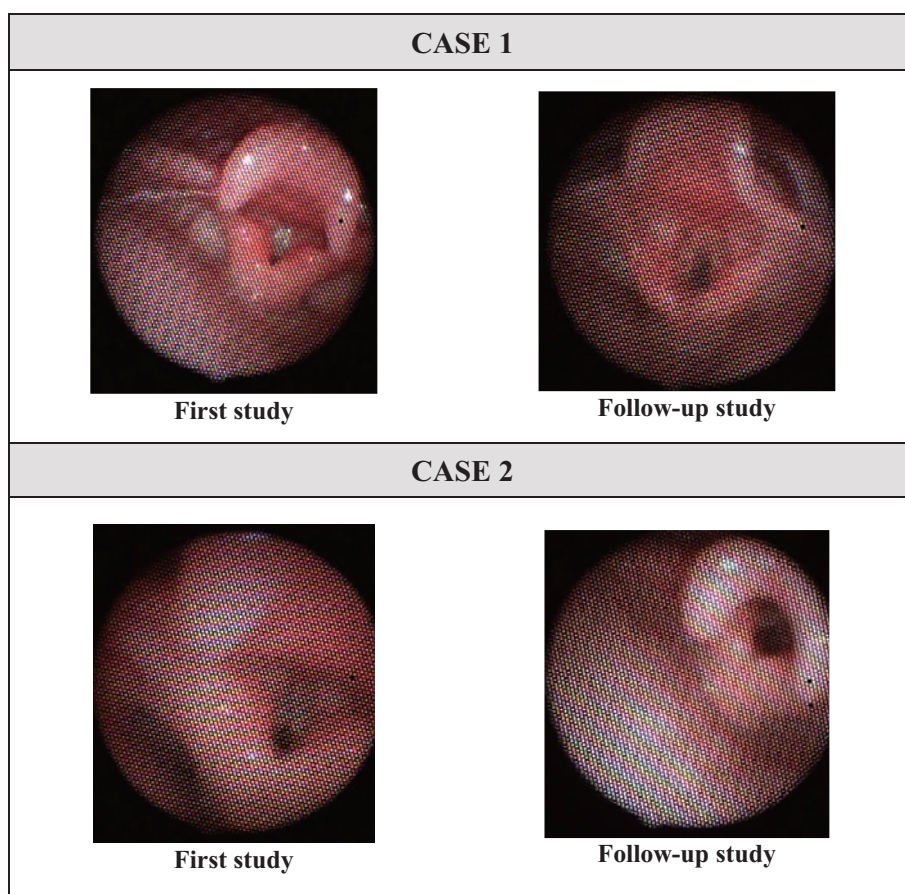


Fig. 1 Images from the endoscopic study

All four images show vocal fold position during inspiration. The vocal fold is normally abducted during inspiration. However, the vocal fold is not fully abducted during inspiration in a patient with vocal fold abduction dysfunction, thus resulting in inspiratory stridor. Both cases showed marked improvement in a follow-up study.

vent progression of tracheomalacia. Clarithromycin was started as prophylaxis for respiratory infections. Repeat laryngo-bronchoscopy on day 60 revealed marked improvement of bilateral vocal fold abduction dysfunction (Fig. 1). After that, the patient used BiPAP only at night and was discharged on day 75.

#### Case 2

The second patient was born at 38 weeks and 5 days of gestation (birth weight, 2,790 grams,  $-0.37$  standard deviation) and was delivered by elective cesarean section because the mother had undergone cesarean section during a previous pregnancy. His APGAR scores were 9 and 10 at 1 and 5 min, respectively. He was admitted to the NICU on day 7 for inspiratory stridor and cyanosis on crying. He had no dysmorphic features, and no signs of congenital heart defects, including vascular rings, on cardiac ultrasonography. There was no evidence of hoarseness, hypercapnia, desaturation, or feeding difficulties. Because the overall clinical course resembled that of case

1, we suspected vocal fold abduction dysfunction. Therefore, respiratory support with CPAP (PEEP: 5 cm H<sub>2</sub>O) was initiated and then switched to BiPAP (PEEP: 8 cm H<sub>2</sub>O) on day 7. An endoscopic workup on day 26 confirmed the diagnosis, and tracheomalacia was not observed (Fig. 1). A follow-up study on day 85 showed resolution of bilateral vocal fold abduction dysfunction, and BiPAP treatment was discontinued (Fig. 1). The patient was started on clarithromycin for prophylaxis of respiratory infection. There was no recurrence of symptoms and the patient was discharged on day 93.

#### Discussion

The most common cause of neonatal stridor is laryngomalacia, followed by vocal fold abduction dysfunction<sup>1</sup>. However, vocal fold abduction dysfunction is not well-documented. Inspiratory stridor is often accompanied by cyanosis, desaturation, CO<sub>2</sub> retention, and feeding difficulties that require feeding tubes. A definitive diagnosis is confirmed by laryngo-bronchoscopy. There are no es-

established criteria for selecting candidates for airway evaluation, but we conducted an endoscopic workup because the first patient (case 1) was symptomatic even at rest. An endoscopic study was also conducted in the second patient (case 2) because the clinical course of case 2 closely resembled that of case 1, which suggested vocal fold abduction dysfunction. Another key point is that symptoms started to appear during the first week of life, suggesting that both patients had potentially severe dysfunction. Clinical manifestations of vocal fold abduction dysfunction vary widely. Inspiratory stridor alone is not harmful but can become a concern when accompanied by symptoms such as inspiratory stridor at rest, cyanosis, hypercapnia, and poor feeding. Although most cases resolve spontaneously, prolonged tube feeding or even tracheostomy might be needed in severe cases<sup>2</sup>. For this reason, early intervention is essential. CPAP therapy is often initiated to prevent progression of comorbidities such as tracheomalacia<sup>3</sup>. Conservative treatment with BiPAP was effective in our patients and symptoms resolved within a few months. In particular, the rapid marked improvement in the second patient is likely attributable to the fact that we suspected vocal fold abduction dysfunction soon after admission because of our experience with the first patient. Hence, CPAP therapy was started earlier, even before the endoscopic study. The laryngeal abductor and adductor muscles must work in coordination to open and close vocal folds. Normally, the vocal fold is abducted during inspiration. However, the vocal fold is not fully abducted during inspiration in persons with vocal fold abduction dysfunction, thus resulting in inspiratory stridor (**Fig. 1**). The proposed mechanism of vocal fold abduction dysfunction is impaired synkinesis of the laryngeal muscle<sup>4</sup>. We classified laryngeal muscle desynchrony into four types based on endoscopic observation<sup>4</sup>, and the present cases are classified as type III, in which the vocal folds are medialized. Some children with a history of obstetric trauma or thoracic surgery may develop

acquired vocal fold abduction dysfunction. Other cases are classified as congenital, which includes central nervous system disorders such as meningomyelocele and Arnold Chiari malformation<sup>1</sup>. Most are idiopathic<sup>5</sup>. However, there were no apparent comorbidities in our patients.

Because diagnosis of vocal fold abduction dysfunction requires a laryngo-bronchoscopy study, there may be a large number of undiagnosed patients. Therefore, to ensure optimal outcomes from early introduction of positive airway pressure devices, vocal fold abduction dysfunction should be considered in the differential diagnosis of neonatal inspiratory stridor.

**Conflict of Interest:** The authors declare no conflict of interest.

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