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Case report

Therapeutic management for patients with cleft lip and palate complicated by sleep apnea syndrome: A case report

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ABSTRACT

We treated a patient with cleft lip and palate by palatoplasty, and he subsequently developed sleep apnea. We then performed tonsillectomy, to improve the sleep apnea, and were able to do this successfully without impairing velopharyngeal function. This successful result was achieved by collaboration between the oral and maxillofacial surgeons who initially performed the palatoplasty and the otolaryngologists who treated the patient for sleep apnea. The decision to remove the palatine tonsils was based on careful observation of the respiratory and sleeping conditions of the patient during sleep, together with polysomnography (PSG). The tonsillectomy was done using a two-step procedure. We obtained a good treatment outcome in a patient with cleft lip and palate who had sleep apnea, based on collaboration between otolaryngologists and oral and maxillofacial surgeons.

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1. Introduction

During palatoplasty, the soft palate is pushed backwards to obtain good velopharyngeal function and accurate articulation [1–3], resulting in narrowing of the upper respiratory tract. In addition, after palatoplasty, the palatine and pharyngeal tonsils undergo physiologic enlargement in early childhood, resulting in more serious stricture of the tract. Both of these factors are causes of sleep apnea. If the bilateral palatine tonsils are extirpated to improve sleep apnea developing after palatoplasty, velopharyngeal insufficiency may result.

We treated a patient with cleft lip and palate by palatoplasty, and he subsequently developed sleep apnea. We then performed tonsillectomy, to improve the sleep apnea, and were able to do this successfully without impairing velopharyngeal function. This successful result was achieved by collaboration between the oral and maxillofacial surgeons who initially performed the palatoplasty and the otolaryngologists who treated the patient for sleep apnea.

The decision to remove the palatine tonsils was based on careful observation of the respiratory and sleeping conditions of the patient during sleep, together with polysomnography (PSG). The tonsillectomy was done using a two-step procedure. Here, we describe the process leading to this good outcome, with reference to the literature.

2. Case report

The patient was a boy born by cesarean section at 38 weeks and 5 days gestation in a local obstetric clinic. He was the second child of a 33-year-old father and 30-year-old mother. Intrauterine growth retardation was identified, and he weighed 1965 g at birth. Because cleft lip and palate were identified at birth, he was referred to the pediatric department of a local hospital for whole-body examination. Staining of G-banded chromosomes performed there revealed a eupyrene 46, XY karyotype. At 28 days after birth, when he weighed 2436 g, the patient was referred to our center for treatment of the cleft lip and palate. Although cheiloplasty was initially scheduled to be performed when the child was 11 months old, the operation was postponed due to the sudden development of thrombocytopenia, diagnosed by the pediatric department of the local hospital. Cheiloplasty was rescheduled for when the boy was 12 months old, but again postponed due to chickenpox and thrombocytopenia. The child underwent cheiloplasty at 16 months of age, after the platelet count had normalized. Palatoplasty was

[☆] AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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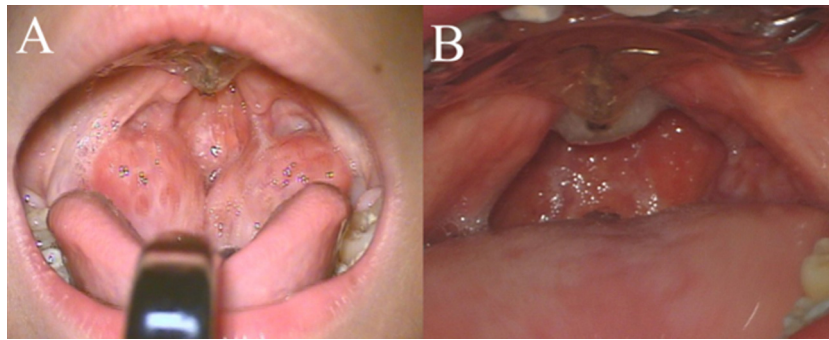


Fig. 1. Photographs of the patient's oral cavity. (A) The oral cavity before palatine tonsillectomy. The palatine tonsils are hypertrophied to the point that the tonsils on the left and right sides are nearly touching, like "kissing tonsils" central apnea. (B) The oral cavity after palatine tonsillectomy.

performed when he was 30 months old. Velopharyngeal insufficiency (VPI) occurred after palatoplasty. The VPI was corrected with a palatal lift prosthesis with attached bulb.

Recheiloplasty was performed when he was 8 years old, to reshape facial structures for esthetic reasons. An otolaryngologist at the local clinic had diagnosed the child with sleep apnea when he was 5 years old, but it had become progressively worse since then. Examination of the child at our center revealed that he sometimes fell into a state of somnolism. We therefore referred him to an otolaryngologist who is the chief director of the Good Sleep Center at Nagoya City University for closer examination (co-author Nakayama). Although the adenoid tonsils were not seriously hypertrophied, the palatine tonsils were hypertrophied to the extent that the tonsils on the left and right sides were nearly touching, like "kissing tonsils" (Fig. 1A).

The Good Sleep Center performed PSG to clarify the sleeping conditions. PSG uses the following investigations to clarify biophysiological changes: electroencephalography (EEG);

electro-oculography; electrocardiography; chin electromyography, leg electromyography, nasal and oral airflow measurements, and respiratory plethysmography for movements of the chest wall and upper abdominal wall; and pulse oximetry for oxygen saturation. Polysomnographic technicians certified by the Japanese Society of Sleep Research performed the above studies overnight in a soundproof room at the Good Sleep Center while monitoring the patient with an infrared camera. Rechtschaffen and Kales scores [4] were used to evaluate the sleep stages and distinguish non-rapid eye movement (NREM) sleep from rapid eye movement (REM) sleep using EEG. NREM sleep comprises four stages. Stages 3 and 4 of NREM sleep are considered present when delta waves are apparent. Short-duration (2–15 s) and long-duration (>15 s) periods of arousal when the body was moving were recorded. The number of apneic events, total sleep time (TST), sleep efficiency (ratio of the number of minutes of sleep to the number of minutes in bed), apnea hypopnea index (AHI), and apnea index (AI) were also recorded. Obstructive apnea was defined as a cessation

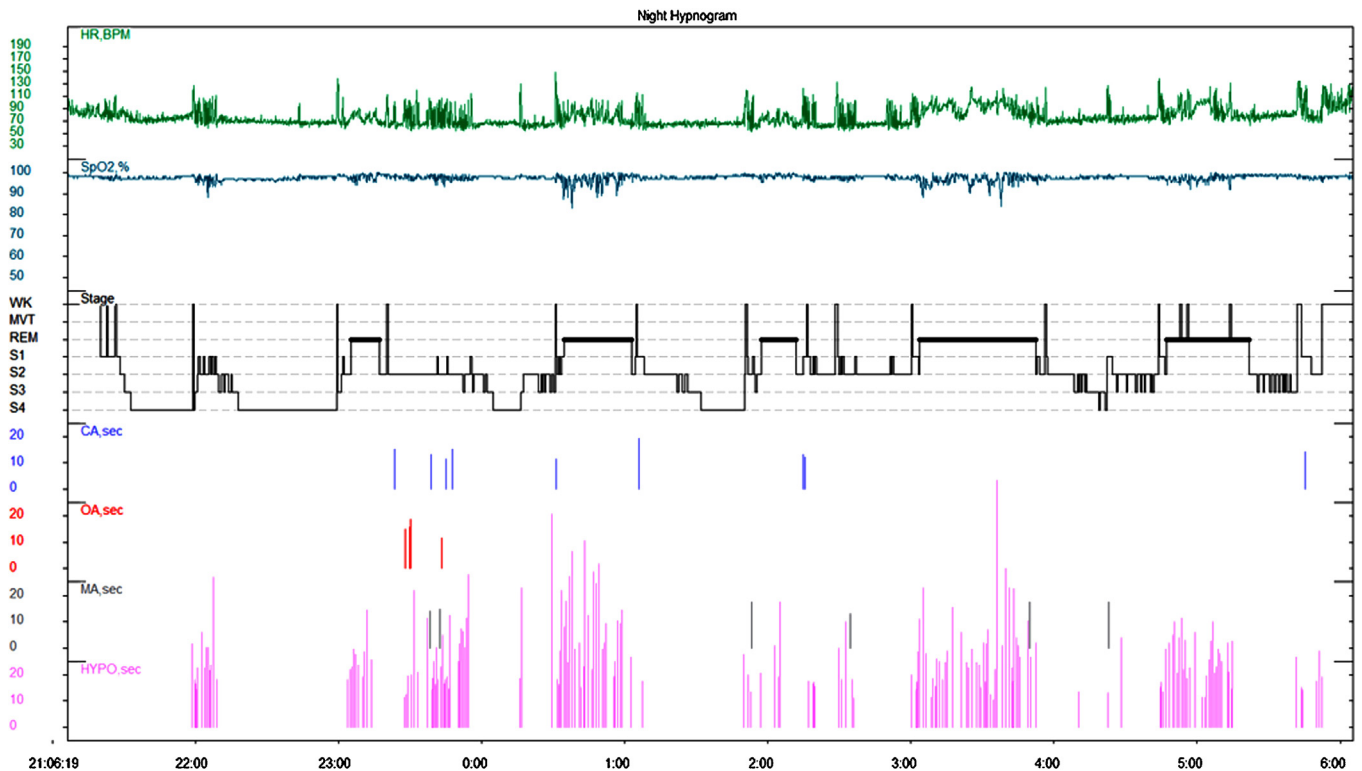


Fig. 2. Night hypnogram and respiratory findings before palatine tonsillectomy. HR BPM, heart rate beats per minute; SpO₂, arterial oxygen saturation; WK, wakefulness; MVT, movement; REM, rapid eye movement; S, stage; CA, central apnea; OA, obstructive apnea; MA, mixed apnea; HYPO, hypopnea; Sec, second.

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