Relief of Upper Airway Obstruction Using a Cervical Splint for Young Patients with Cerebral Palsy

Hyoju Son*/ Soohyeon Kim**/ Jaeho Lee***/ Chungmin Kang****/ Seunghye Kim*****

13-year old boy with spastic quadriplegia cerebral palsy visited dental clinic with chief complaints of mouth breathing and malocclusion. His mouth was constantly open at the resting position, with his mandible and tongue displaced downward. He breathed through his mouth, making a constant gurgling sound, a sign of upper airway obstruction. To enhance his mandible position, vertical chin cap was first considered, but it was not sufficient to reduce the gurgling sound or ease breathing. Then, cervical splint was considered, which effectively decreased the gurgling sound by repositioning his mandible to the anterior-superior position. Oxygen saturation was increased when the cervical splint was used. Cervical splint can effectively assist breathing in patients with cerebral palsy, but it should be carefully applied as long-term use can result in unexpected complications. Under instruction by a physician regarding proper usage, a cervical splint can be applied to assist breathing in patients with cerebral palsy.

Key words *: cerebral palsy, gurgling sound, upper airway obstruction, cervical splint,*

INTRODUCTION

Cerebral palsy (CP) involves non-progressive damage to developing embryos or children, causing permanent injuries in development of movement or position.[1] It generally stems from antenatal, laboring, and postnatal brain damage for various reasons; most cerebral palsy is known to be caused by damage during the antenatal period (75-80%).[2] The remainder of cases result from postnatal damage to the head and neck or postnatal infection of the central nervous system (10%).[2] Quadriplegia is generally caused by anoxia and is accompanied with sporadic convulsion; moreover, 75% of patients show intelligence decline.[3] Due to malfunction of the myelencephalon, drooling, swallowing difficulty, and pronunciation problems may simultaneously occur, and it is difficult for patients with severe quadriplegia to maintain their position and walk, even with a walking frame.[1]

Young patients with muscle hypotonia-related cerebral palsy or neurogenic disorder are at high risk to experience airway obstruction.[4] Due to maxillary hypoplasia, adenotonsillar hypertrophy, palatal hypotonia, glossoptosis, retrognathia, midface retrusion, transverse palatal collapse, and laryngeal dystonia, they also are at high risk for upper airway obstruction.[5-8] In addition, because of dysfunction in the brain stem, there is a change in not only alertness, but also cardiac and respiratory control.[9] Upper airway obstruction in cerebral palsy patients can trigger life-threatening complications. In addition, cerebral palsy patients have a high tendency for seizure, gastroesophageal reflux, and hypersalivation, which generally aggravates apnea.[4]

We report a case where application of a cervical splint successfully alleviated upper airway obstruction in a CP patient, helping the patient with breathing and increasing the level of oxygen saturation.

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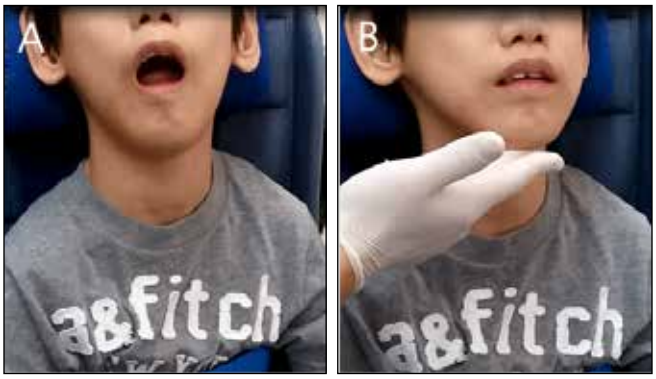
CASE REPORT

A 13-year-old patient with spastic quadriplegia CP visited the Pediatric Dentistry Department of Yonsei University Dental Hospital, Seoul, Republic of Korea, with the chief complaints of mouth breathing habit and malocclusion. In addition to quadriplegic CP, the patient had mental retardation and delayed development. He regularly visited the rehabilitation clinic for his lower limb orthoses. At resting position, his mandible was displaced downward. When his mandible was guided to centric occlusion (CO), open bite and severe labioversion of the upper anterior teeth were noticed with overjet and overbite of 14 mm and -2 mm, respectively. The molar key was class I on both sides. In the resting position, the mandible was displaced to the posterior-inferior position, as shown in Fig 1A, and he was breathing arduously with a loud gurgling sound. When we repositioned the mandible to an upward and forward position, the gurgling sound was significantly reduced (Fig 1B).

Figure 1. [A] Clinical photo of the patient at resting position showing muscle hypotonia and downward mandible position. Constant gurgling sound was present when the patient was breathing. [B] Clinical photo of the patient with mouth closed by hand manipulation. Gurgling sound was reduced in this position.



He was diagnosed with open bite with labioversion of the upper anterior teeth. Orthodontic treatment was not considered, but a vertical chin cap was considered to reposition his mandible to the upward and forward position, thereby widening his upper airway and help him breathe. When the vertical chin cap was applied, his oxygen saturation level was increased to above 96%, whereas it was 93-97% in the resting position. However, the patient's mouth was not fully closed and the gurgling sound during breathing remained even when the vertical chin cap was applied. After using the vertical chin cap for 3 months, his parents wanted to switch to an appliance that would be more effective.

At the next visit, we proposed a Philadelphia collar-type cervical splint (Fig 2A) for reposition of the mandible, improvement of breathing by airway expansion, head tilt, and chin lift. With the Philadelphia collar cervical splint, he breathed more comfortably in the resting position, and the gurgling sound was absent (Fig 2B). However, because of the stiffness of the equipment, he developed a skin rash, and his parents reported persistence of a slight gurgling sound. At his parents' suggestion, a cervical splint was customized with memory foam, the same material the patient was using for his pillow. The customized memory foam cervical splint enabled both effective breathing and patient comfort (Fig 3A). With this modified cervical splint, the gurgling sound disappeared and oxygen saturation was increased to 98%. In addition, the modified cervical splint, made with memory foam, was Velcro fastened in the front to allow adjustments to tightness and air-permeability (Fig 3B).

Figure 2. [A] Philadelphia collar cervical splint. [B] Clinical photo of the patient wearing cervical splint, which assisted mouth closing, thereby reducing gurgling sound.

Figure 3. Lateral view [A] and frontal view [B] of the patient wearing modified cervical splint

At the 2-month follow-up, the parents showed satisfaction with this modified cervical splint. The patient generally used the modified cervical splint for four to five hours per day, usually at school, and his parents reported that the patient breathed more comfortably with reduced gurgling sounds.

DISCUSSION

Cerebral palsy patients often experience upper airway obstruction due to various factors, such as displacement of the mandible and macroglossia, muscle malfunction, and enlarged adenoids and tonsils.[5-8] Upper airway obstruction in CP patients can trigger complications, from noisy breathing to complete obstruction, which may cause silent breathing sounds, apnea, and loss of consciousness in extreme cases.

Other features of airway obstruction include snoring, stridor (caused by obstruction at or above the laryngeal level), wheezing (caused by obstruction of the lower airway), and gurgling sounds (vomit, blood, or secretions in the airway). In unconscious patients, obstruction of the upper airway occurs at the pharynx.[10] The main reasons for this are the tongue is dropping to the posterior pharyngeal wall, decreased muscle tone, and reduced airway

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diameter.[10] To improve airway patency, methods of chin lift or jaw thrust maneuvers can be used.

The pharyngeal muscle is important for maintaining patency of the upper airway. Moreover, during regular breathing, the respiratory center stimulates the pharyngeal muscle to stiffly contract the pharynx before contraction of the diaphragm. If this process is hampered by disability, patients with CP could experience upper airway obstruction, and breathing may become noisy. These symptoms might be aggravated during sleep. It has been reported that young patients with severe CP show obstructive sleep apnea (OSA), even though its prevalence has not been fully studied.[11]

To relieve upper airway obstruction, a non-surgical method was introduced in the 1930s, involving repositioning of the mandible to the anterior position with a helmet and chinstrap. Another method involving a tongue retaining device and mandible advancement device was introduced in 1982 to reduce upper airway obstruction caused by OSA by repositioning the tongue and mandible.[12] These types of intra-oral equipment aim to change the structural features of the mandible, tongue, and oral pharynx and further reduce obstruction of the upper airway. Therefore, we think that these devices may be beneficial for young CP patients with upper airway obstruction who experience difficulty with respiration, even while awake.

Mandible advancement devices are applicable to most cases of OSA or snoring. However, if those patients have open mouth or anterior displacement of the mandible, these devices may be difficult to use. Alternatively, Shimoyama and colleagues reported that, in infants with severe CP who suffered from upper airway obstruction, respiration was improved by a tube-connected palatal appliance.[13] However, in 1995, Ogasawara et al. reported that intra-oral devices (like bite blocks) in patients with serious disability could cause hypoxemia.[14] Our patient's condition was not compatible with intra-oral devices, because he was young and could potentially swallow them.

To reposition the mandible and further help breathing, a helmet and a chin strap had been previously used. However, a chin cap causes a downward rotation of the mandible, lingual tipping of the lower anterior teeth, and an open bite if the patient has long lower facial height. Since this young patient had long facial height, we should have considered that this would be a problem. A vertical chin cap is also applicable for patients with an open bite, but a chin cap alone cannot maintain a closed position for the mouth. Therefore, we decided to use an extra-oral device called a cervical splint that can reposition the mandible to an anterior position.

In this case, a cervical splint was used to help keep the mouth closed and assist with breathing, although it is generally used for cervical spine support due to damage caused by trauma, surgery, bone fracture, or dislocation.[15] A cervical splint also decreases the pressure on nerves by supporting the cervical spine, helps in the healing of soft tissue by restriction of neck movement, and decreases the load of the head on the cervical spine. There are several types of cervical splints, such as the cervical collar, Philadelphia collar, four-poster, and halo vest. The Philadelphia collar can restrict movement even more than the cervical collar and is usually made with polyethylene and further supported with stiff plastic. This collar involves contacts from the mandible and occipital bone to the upper thorax, and this can limit the bend of the body. With a younger patient, as in our case, the Philadelphia collar can limit neck movement more than a hard collar; our upgraded device based on a memory foam pillow was more similar to a soft collar.

However, sustained use of a cervical splint can cause various side effects and should be used with caution. According to a study by Spitzer et al., after one week fixation, a structural change in muscle can be observed in animal models.[16] In another report[14], long-term usage of a cervical splint was reported to cause contraction of the neck muscles and soft tissue and further influence mental status Long-term usage of a hard cervical splint can cause numbness of the marginal mandibular nerve, full thickness ulcer of the skin, and airway obstruction during sleep due to compression. Moreover, according to a study by Dibsie and Krock, limitations on swallowing, coughing, breathing, vomiting, and aspiration can be caused by this device.[17,18]

Therefore, to prevent these side effects and complications, we recommend use of a cervical splint for short and sporadic periods under instruction of a rehabilitation physician. Despite the various risks of complications, limited use of a cervical splint can be useful to assist with breathing in patients with muscle hypotonia, especially in cases of upper airway infection, when breathing becomes very difficult. In such patients, consultation with an ear-nose-throat specialist is necessary to determine if there are any anatomical reasons (such as adenotonsillar hypertrophy, palatal hypotonia, or laryngeal dystonia) that might cause upper airway obstruction.

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