

Severe trismus due to bilateral coronoid process hyperplasia in growth hormone therapy patient: a case report

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Bilateral coronoid process hyperplasia is a rare condition characterized by an enlarged mandibular coronoid process. The painless progressive reduction of a mouth opening is caused by coronoid process impingement on the posterior aspect of the zygomatic bone. Hyperplasia of the bilateral coronoid process leads to the restriction of a mandibular opening consequent to the impingement of the enlarged coronoid process on the temporal surface of the zygomatic bone or with the medial surface of the zygomatic arch. The process has been diagnosed as developmental hyperplasia. Otherwise, the development of the coronoid process may be associated with growth hormone. This paper describes a case of trismus caused by coronoid hyperplasia in an idiopathic short-stature patient who received growth hormone therapy by somatropin injections.

Key words: Coronoid process hyperplasia, Trismus

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I. Introduction

Bilateral Coronoid Process Hyperplasia is a rare condition characterized by an enlarged mandibular coronoid process. Continuously proliferating coronoid process causes trismus due to impingement on the temporal surface of the zygomatic bone or the medial surface of the zygomatic arch. According to Isberg et al.¹, coronoid process hyperplasia is the cause of approximately 5% of trismus patients. Many hypotheses suggest stimulating endocrine, increased activity of temporal muscle, injury, and heredity as the possible factors contributing to coronoid process hyperplasia, but the cause has yet to be identified clearly².

The most common clinical feature of coronoid process hyperplasia is painless and progressive trismus. Unilateral coronoid process hyperplasia is commonly combined with facial asymmetry and sometimes combined with mobile

uplift on the upper part of the zygomatic arch and mandibular displacement toward the affected side during mouth opening. Unilateral and bilateral coronoid process hyperplasia may occur in all age groups (average 25) but rarely in people aged 10 or under. Moreover, most patients were male, but female patients were rare.

As a treatment method, coronoidectomy can achieve meaningful enhancement of mouth opening, but there is a need to check for fibrosis caused by surgery or regrowth of coronoid process for a long time².

This case report deals with a patient diagnosed with idiopathic short stature (ISS) in 2008 by the Department of Pediatrics and administered growth hormone therapy and whose trismus—caused by bilateral coronoid process hyperplasia—was treated with coronoidectomy.

II. Case Report

On April 4, 2006, a 12-year-old male patient visited the Department of Pediatric Dentistry and Orthodontics, Pusan National University Hospital Korea due to the delayed eruption of permanent teeth and facial asymmetry. The patient weighed 34 kg, and he was 135 cm tall; he was relatively small compared with other children in his age, and many of his teeth had birth defects. The patient had received treatment

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at the Department of Pediatric Dentistry and Orthodontics without trismus; in August 2008 when he was 14, he was referred to Department of Oral and Maxillofacial Surgery by the Department of Orthodontics due to facial asymmetry and trismus. Clinical examinations revealed facial asymmetry, mouth opening level of 15 mm, and displacement to the left of the mandible during mouth opening; the panoramic photographs showed left coronoid process hyperplasia. The patient did not have any particular injury or infection or family history of similar symptoms, but he was diagnosed with ISS in January 2008 by the Department of Pediatrics in our hospital and administered growth hormone therapy using Eutropin (Recombinant Human Growth Hormone [somatropin], LG Eutropin Inj.; LG Life Sciences, Seoul, Korea) injection. Our Department planned to encourage active mouth opening movement, observe his status during the growth period, and perform surgery, but the patient did not visit again. Later, the patient visited again in July 2011 when he was 17 due to restriction of mouth opening. Clinical examinations revealed facial asymmetry, displacement to the left of the mandible during mouth opening, and maximum mouth opening level of 10 mm, decreasing compared to

2008.(Fig. 1) Moreover, he had a hard-end-feel on both sides when opening his mouth to the left and right. The panoramic radiographic inspection and dental cone-beam computed tomography (CT) (Pax-Zenith 3D; Vatech, Hwaseong, Korea) images showed hyperplasia on both sides of the mandibular coronoid process.(Fig. 2) In particular, the three dimensional (3D) images obtained by reorganizing the dental cone-beam CT images with 3D analysis program (Simplant; Materialise, Leuven, Belgium) revealed that the left coronoid process was close to the medial surface of the left zygomatic bone, and that the inner side of the left zygomatic bone was transformed.(Fig. 3) The patient's height increased from 143 cm in January 2008 when the growth hormone therapy was started to 153 cm when the patient visited us in August 2008 due to trismus. And in comparing panoramic radiographs, the panoramic images taken in August 2008 showed a more noticeable hyperplasia in the left coronoid process.(Fig. 4) The patient kept receiving growth hormone therapy until October 2009, and his height increased from 153 cm to 167 cm in July 2011. The maximum mouth opening level decreased from 15 mm in August 2008 to 10 mm in July 2011.(Table 1)

Therefore, based on the patient's medical history, clinical examinations, and radiographic inspection, we determined

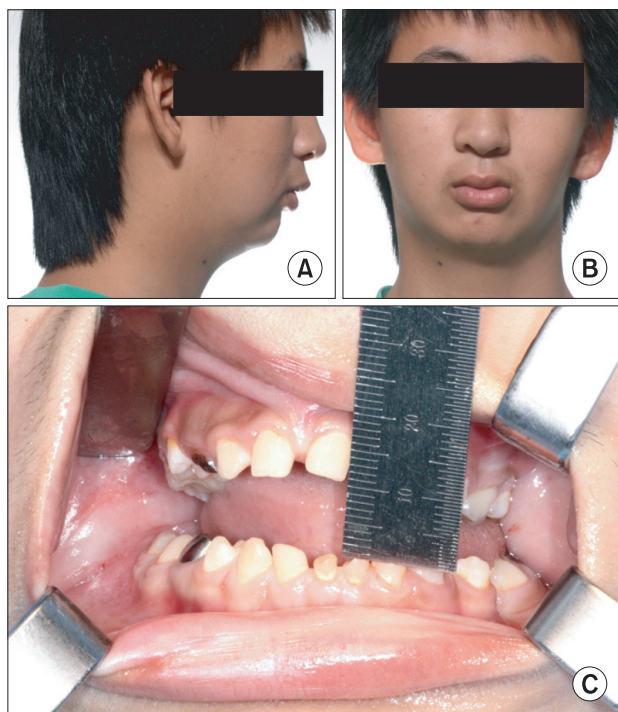


Fig. 1. Preoperative clinical photograph. A. Lateral photo view. B. Frontal photoview. C. Preoperative maximum mouth opening measurement was 10 mm.

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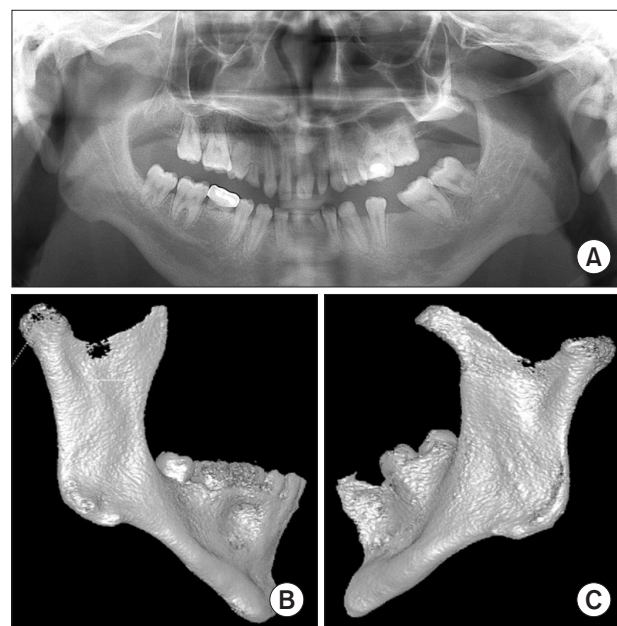


Fig. 2. Preoperative radiologic view show both hyperplastic coronoid process. A. Panoramic view. B. Dental conebeam computed tomography (CT) view (mandible right). C. Dental conebeam CT view (mandible left).

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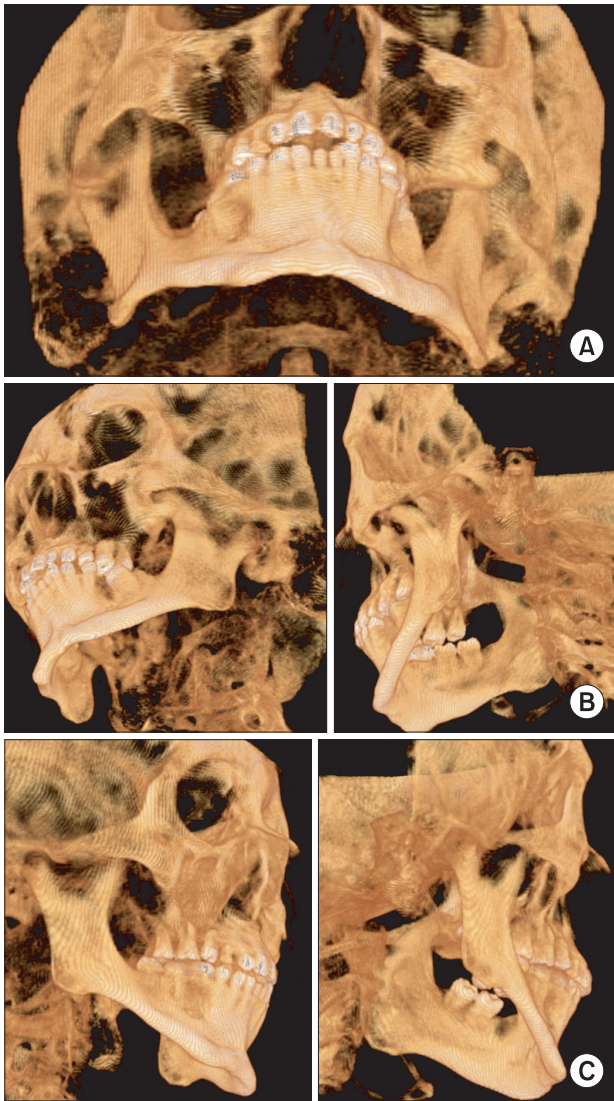


Fig. 3. Three dimensional implant image. A. The image show morphological abnormality of left zygomatic bone. B. Left coronoid process extend above the zygomatic bone and impinge on the posterior surface of the zygomatic bone. C. Right coronoid process have just hyperplastic morphology.

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that the growth hormone therapy caused hyperplasia in the bilateral mandibular coronoid process and diagnosed the transformation of the left coronoid process and impingement on the inner side of the zygomatic bone as the cause of trismus. As such, we planned to perform coronoidectomy on both sides of the mandible under general anesthesia in August 2011.

General anesthesia was performed with nasotracheal intubation using fiber optic scope because it was hard to execute nasotracheal intubation using head scope due to the insufficient mouth opening level. Considering the diagnostic process, left coronoidectomy was carried out first due to the hard-end-feeling and transformation of coronoid process. At first, left coronoidectomy was performed through the mouth; however, due to insufficient mouth opening level, the submandibular approach was taken. After the surgery, the maximum mouth opening level increased by 37 mm. Afterward, right coronoidectomy was done through the intraoral approach. After the surgery, the maximum mouth

Table 1. Trismus according to growth hormone therapy

Date (yy/mm/dd)	Growth hormone therapy	Height (cm)	Mouth opening
2006/4	×	135	W.N.L
2008/1/1	×	143	W.N.L
2008/1/21	Eutropin 21 vial	143	W.N.L
2008/2/18	Eutropin 42 vial	144	W.N.L
2008/4/14	Eutropin 75 vial	148	W.N.L
2008/7/2	Eutropin 80 vial	151	W.N.L
2008/7/30	Eutropin 21 vial	152	W.N.L
2008/8/26	Eutropin 42 vial	154	15 mm
2008/10/21	Eutropin 42 vial	156	-
2009/7/31	Eutropin 45 vial	160	-
2009/10/5	Eutropin 45 vial	161	-
2011/8/8	×	167	10 mm

(W.N.L: within normal length)

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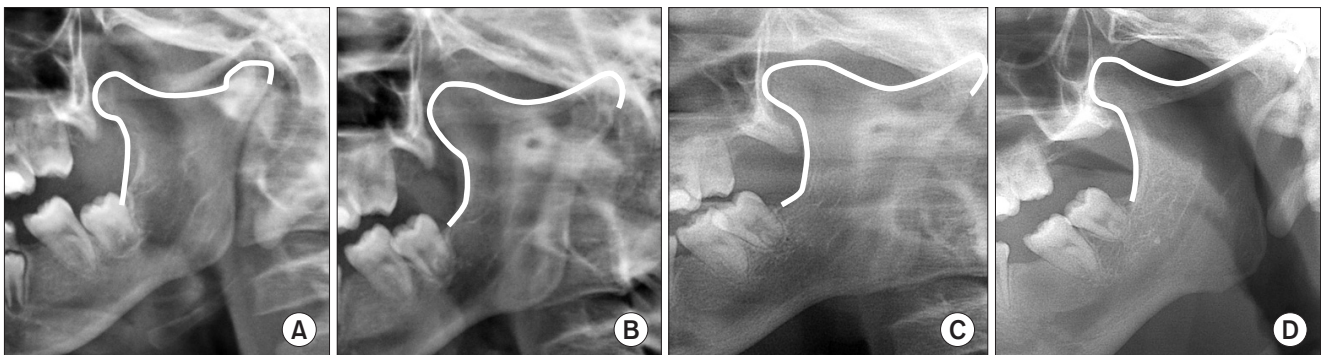


Fig. 4. Serial panoramic view. A. April 2006. B. August 2007. C. August 2008 after growth hormone therapy (January 2008). D. July 2011.

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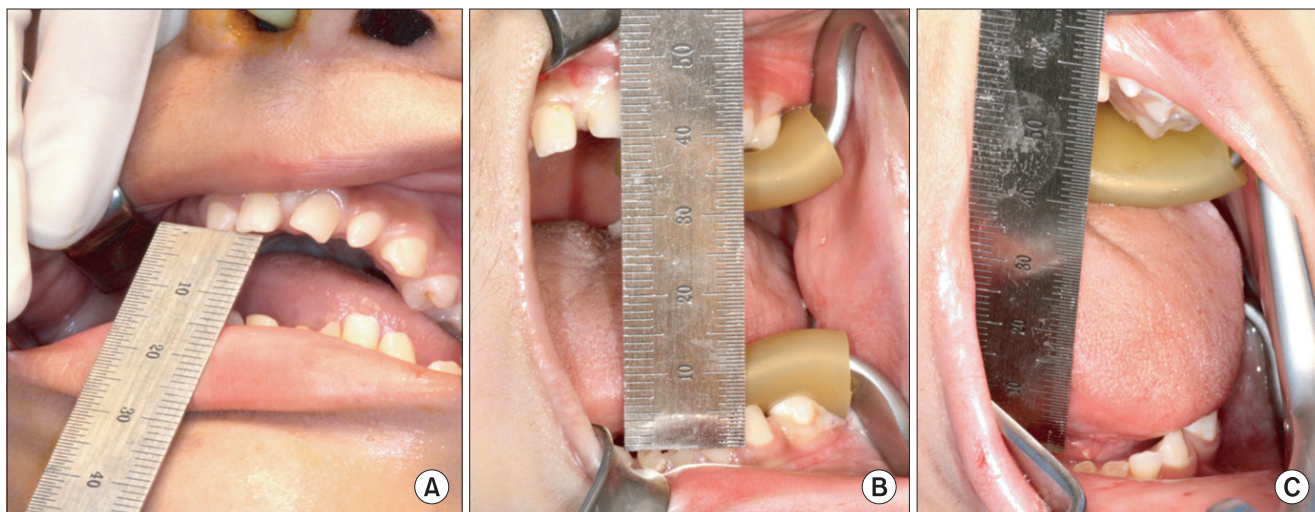


Fig. 5. A. Perioperative mouth opening was 10 mm. B. Mouth opening after left coronoidectomy was 40 mm. C. Mouth opening after both coronoidectomy was 53 mm.

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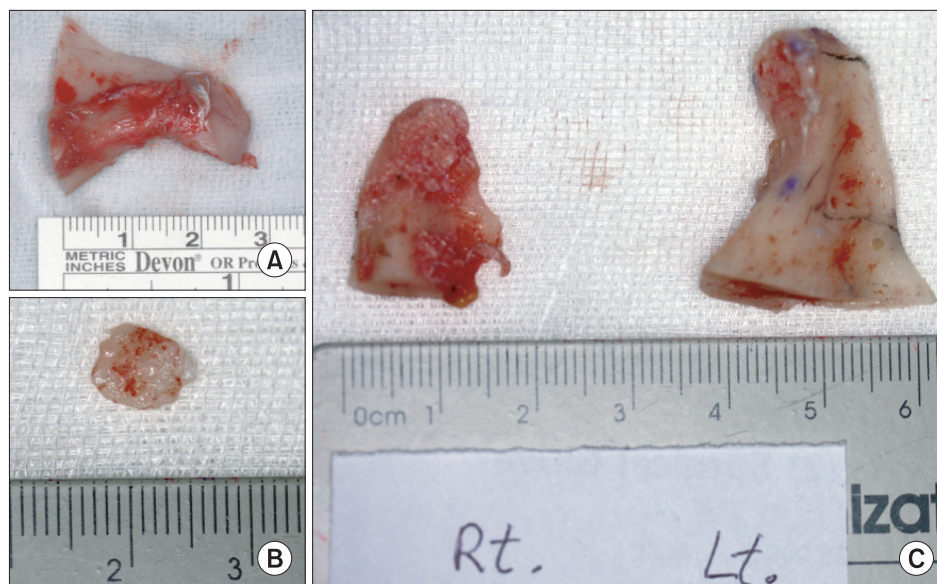


Fig. 6. A. Resected Lt. coronoid process. B. Lt. coronoid cartilage from coronoid process tip. C. Comparison resected Lt. coronoid process with resected Rt. coronoid process. (Lt.: left, Rt.: right)

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opening level increased by 53 mm.(Fig. 5) Extracted left coronoid process had hyperplasia horizontally; cartilage proliferating 2 mm thick was noted at the end. Extracted right coronoid process was not transformed, but it had overall hyperplasia.(Fig. 6)

One month after the surgery when the patient visited again, the maximum voluntary mouth opening level was 40 mm without edema, pain, and neural damage.(Fig. 7)

III. Discussion

Generally, trismus means all problems in mouth opening. The normal mouth opening level is 40-60 mm, based on



Fig. 7. A. One month mouth opening after surgery. B. Postoperative panoramic view.

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the distance between the maxillary and mandibular teeth³. A clinical method involves measuring finger breadth (FB, breadth of index finger's nail), generally 17-19 mm. The normal value is 2FB or 3FB⁴.

For the reason and classification of trismus, many researchers proposed various hypotheses⁵⁻⁷. Therefore, when a trismus patient visits, there is a need to perform diagnosis based on clinical opinions and diagnostic radiographic inspections considering possible reasons such as temporomandibular joint disorder, temporomandibular joint ankylosis, benign tumor, malignant tumor, dental infection, maxillofacial fracture, and other diseases.

When the patient in this case visited us, though the panoramic radiography inspection revealed hyperplasia of the left coronoid process and calcification of bilateral stylohyoid ligament, the clinical examinations did not find any feeling of irritation, throat discomfort, and odynophagia during mouth opening; only a hard feeling was noted. Therefore, we found that the calcified stylohyoid ligament does not influence mouth opening; with an additional dental cone-beam CT examination, the patient was diagnosed with trismus caused by the overgrowth of the left coronoid process and transformation of the left zygomatic bone.

When the patient visited us for the first time in August 2008 due to trismus, he was diagnosed with ISS; he had been administered growth hormone therapy since January 2008 using Eutropin (Eutropin, recombinant methionyl-hGH) injection (Somatropin). In particular, he had been treated by the Department of Orthodontics until August 2007, but there was no symptom of trismus. After the patient received growth hormone therapy, the deterioration of hyperplasia in the left coronoid process and generation of trismus were observed in the panoramic radiographic images.

Idiopathic short-stature patients have short stature (below -2.0 standard deviation compared to the average stature of children of the same age and sex⁸); though their blood growth hormone concentration is higher than the standard of growth hormone deficiency, their stature is similar to that of children with growth hormone deficiency⁹. Such is caused by the insufficient secretion of growth hormone, decrease of sensitivity to growth hormone, or combination of both factors.

Spiegel et al.¹⁰, Kjellberg et al.¹¹, and van Erum et al.¹² reported that, based on the cephalometric analysis of boys with short stature, not only height but the growth and development of face were influenced by growth delay; they had small, flat basis cranii and mandible located in the posterior area. On the other hand, Isaksoon et al.¹³ reported that the

growth hormone stimulates overall bones and muscles particularly the growth of cartilage through the insulin-like growth factor I (IGF-I). Pirinen et al.¹⁴ also stated that the growth of basis cranium and mandibular cartilage is affected by both excess and insufficiency of the growth hormone. Considering these research studies, insufficient vertical growth in the mandibular condyle and posterior skull base in charge of cartilage growth causes small mandible and short posterior facial height, eruption of dentition increase, lower facial height, and mandible rotated clockwise; hence the unique craniofacial shape of short-stature patients. The patient in this case also had the facial shape of short-stature patients.

A growth hormone agent (Somatropin) means a synthetic human growth hormone. Since 1958 when Raben¹⁵ injected a human growth hormone extracted from the pituitary gland of a cadaver into a patient with growth hormone deficiency for the first time and reported its effects, it has been used for 40 years. The growth hormone stimulates the growth of bones and muscles as well as the composition of IGF-I in liver. Therefore, the injection of growth hormone can resolve the problem of insufficient hormone as well as low sensitivity to growth hormone, thereby increasing the final height by stimulating growth during childhood and triggering the growth of spheno-occipital synchondrosis in basis cranii and mandibular condyle. Actually, Chung et al.¹⁶ reported that 2 years of growth hormone therapy for children with short stature led to the increase of posterior facial height and growth of the mandible.

Though there is no other case report on the relationship between the growth of the mandibular coronoid process and growth hormone as well as consequent complications such as trismus, this case report concluded that the coronoid process originating with cartilage may have hyperplasia of the mandibular coronoid process due to the growth hormone agent and contraction of temporal muscle. Nonetheless, more research on the evident reason is needed to identify the mechanism of coronoid process hyperplasia.

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