

Case Report

Development of Dysphagia and Trismus Developed after C1-2 Posterior Fusion in Extended Position

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Cervical misalignment after upper cervical fusion including the occipital bone may cause trismus or dysphagia, because the occipito-atlanto joint is associated with most of the flex and extended motion of the cervical spine. There are no reports of dysphagia and trismus after C1-2 fusion. The purpose of this paper is to demonstrate the potential risk of dysphagia and trismus even after upper cervical short fusion without the occipital bone. The patient was a 69-year-old man with myelopathy caused by os odontoideum and Klippel-Feil syndrome, who developed dysphagia and trismus immediately after C1-2 fusion and C3-6 laminoplasty. Radiographs and CT revealed that his neck posture was extended, but his symptoms still existed a week after surgery. The fixation angle was hyperextended 12 days after the first surgery. His symptoms disappeared immediately after revision surgery. The fixation in the neck-flexed position is thought to be the main cause of the patient's post-operative dysphagia and trismus. Dysphagia and trismus may occur even after short upper cervical fusion without the occipital bone or cervical fusion in the neck-extended position. The pre-operative cervical alignment and range of motion of each segment should be thoroughly evaluated.

Key words: dysphagia, trismus, os odontoid, Klippel-Feil syndrome, atlantoaxial posterior fusion

There are several reports that a posterior occipito-cervical long fusion may develop in the upper airway and cause pharyngeal trouble, such as respiratory failure, dysphagia, or trismus [1-6]. The main cause of these symptoms is suspected to be the physical obstruction of the upper airway and pharyngeal space when the patient's neck position is flexed. The pharyngeal space never enlarges because of the loss of range of motion after fusion. Especially

in the case of spinal fusion from occipital bone to thoracic spine, surgeons need to pay great attention to the patient's neck position. On the other hand, the risk of pharyngeal troubles seems to be low in the case of posterior cervical short fusion. Yoshida reported a case of upper airway obstruction after a posterior short fusion from the occipital bone to the second cervical vertebra [7]. However, there are no reports on pharyngeal troubles after a posterior fusion that did not involve an occipital bone. We report the case of a 69-year-old man with myelopathy caused by os odontoideum and Klippel-Feil syndrome, who developed dysphagia and trismus immediately after C1-2 fusion.

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Case Study

A 69-year-old man presented with a two-year history of neck pain and a two-month history of lancinating pain to his body and extremities in the neck-extended position. The patient had no subjective muscle weakness and no dysfunction of the bladder or bowel function. His tendon reflexes in the upper and lower extremities were increased. There were no problems on the ADL until the lancination pain occurred. Plane and stress radiographs showed severe instability between C1 and C2 because the dens was separated from the C2 vertebral body. The C4 and C5 vertebrae were fused; this is typical of Klippel-Feil syndrome (Fig. 1A-C). CT after myelography showed os odontoideum, and the atrophy of the spinal cord at the C1 level indicated severe instability at this level (Fig. 1D, E). MRI showed a signal change in his spinal cord at the C1 level and narrowing of the spinal canal at the C3 and C4 level as well as at the C5 and C6 level (Fig. 1F). His diagnosis was myelopathy due

to os odontoideum and spinal canal stenosis due to the adjacent segment disease with congenital fused vertebrae.

Atlantoaxial posterior fusion was performed. Lateral mass screws were inserted into the atlas, and pedicle screws were inserted into the axis. These screws were connected as the os odontoideum was reduced and extended as much as possible. Laminoplasty from C3 to C6 was performed. The operation time was 150 min, and the estimated blood loss was 250ml. Immediately after extubation, he complained of pharyngeal discomfort and difficulty in opening his mouth, but there were no remarkable changes in his vital signs. In light of the fact that he had just undergone intubation and cervical spinal surgery, we considered his complaints minor. As time passed, however, his pharyngeal discomfort improved but his trismus and dysphagia did not resolve. He was able to swallow only liquid meals. A dentist said that there was something physically interfering with his ability to open his mouth. Radiographs and CT were performed 1 week

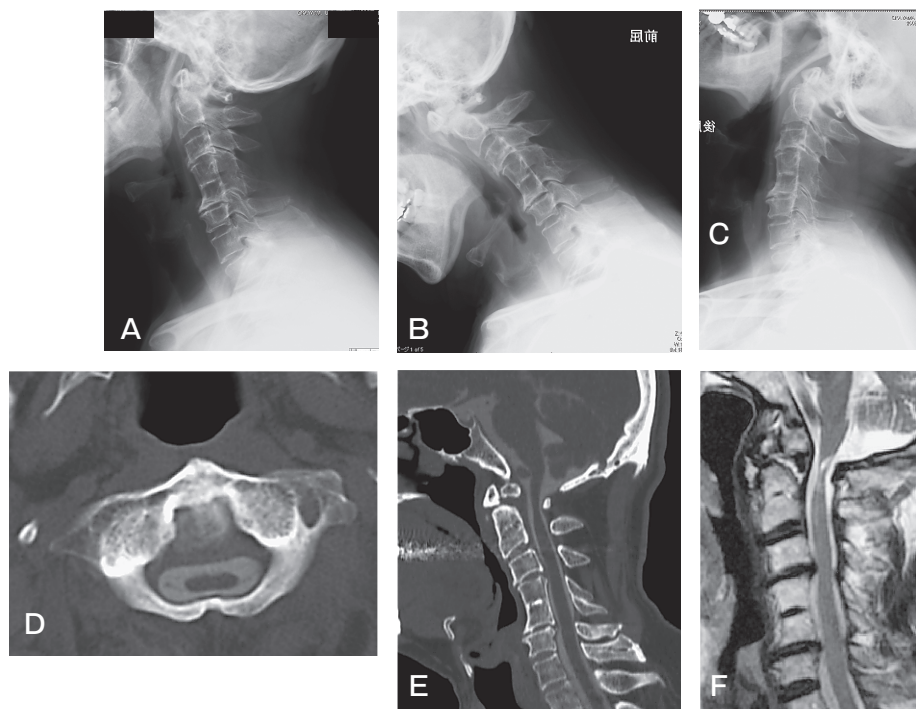


Fig. 1 Preoperative images. Lateral radiograph shows the os odontoideum and C4 and C5 fused vertebrae (A). Dynamic radiographs show severe instability at the C1 level (B, C). CT myelography shows the os odontoideum, and the atrophy of the spinal cord at the C1 level indicated severe instability at this level (E, F). MRI shows a signal change in his spinal cord at the C1 level and spinal canal stenosis at the C3 and C4 level as well as at the C5 and C6 levels (F).

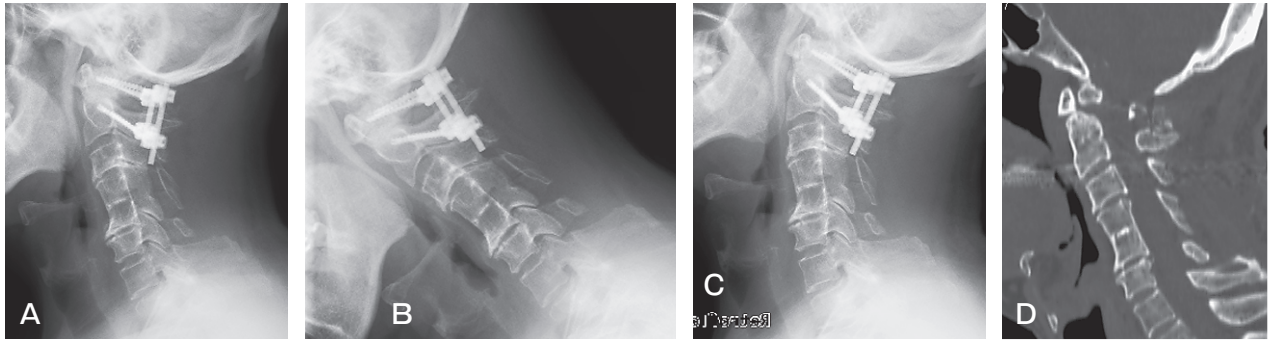


Fig. 2 Postoperative images. The radiograph in the neutral position shows that the O-C2 angle was 27° and within the normal range. Postoperative dynamic radiographs show that his cervical spine’s range of motion is not small, but that of his occipito-cervical junction is almost zero. The heads of the C1 lateral mass screws touch the occipital bone and interfere with extending motion. There is no space between his mandibular bone and vertebrae (A-C). The postoperative CT shows C1 and C2 were fixed in the position so that the os odontoid was reduced acceptably (D).

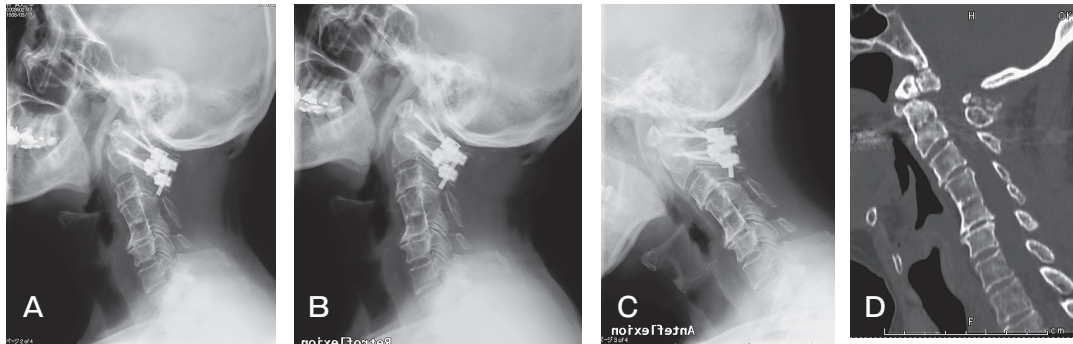


Fig. 4 Post-revision images. His neck is fixed in the hyperextended position. The range of motion of his occipito-cervical joint is not zero (A-C). Post-revision CT shows that the os odontoideum is displaced posteriorly (D).

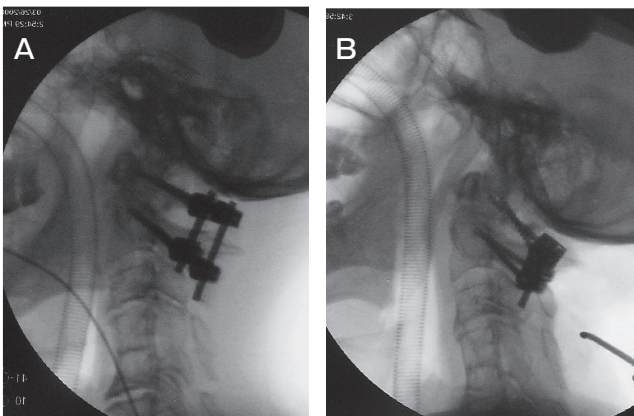


Fig. 3 Images during revision surgery. Pre-revision image (A) and post-revision image (B). His neck is fixed in the hyperextended position. The post-revision O-C2 angle is 43°, larger than the pre-operative O-C2. There is space between his mandibular bone and cervical vertebrae after the revision surgery.

after the operation (Fig. 2A-D). The postoperative CT showed C1 and C2 were fixed in position, so that os odontoid was reduced acceptably. The radiograph in the neutral position showed O-C2 and C1-C2 angles of 27° and 43°, respectively. This meant that his fixation posture was in the extended position. The O-C2 angle in the extended position was 27° and was the same in the neutral position. The screw heads inserted into C1 came in contact with the occipital bone and interfered with extended motion. Furthermore, there was no space between his mandible and cervical vertebral body. We suspected these were the causes of his complaints. His trismus and dysphagia remained, and we performed an operation to arrange his fixation angle 12 days after the first operation (Fig. 3A, B). The fixation angle of C1-C2 was changed to the hyper-extended position (Fig. 4 A-C). Post-revision CT shows that the os odonto-

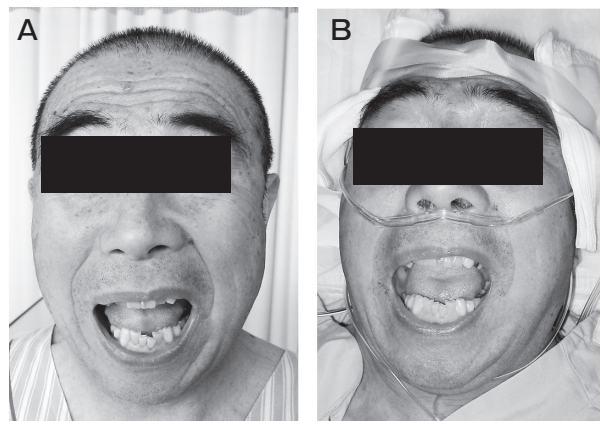


Fig. 5 Post-operative and post-revision pictures. These pictures were taken with his mouth open as widely as possible after the first operation (A) and just after the revision (B). These pictures show he was able to open his mouth wider after the revision.

deum is displaced posteriorly (Fig. 4D). His trismus and dysphagia were resolved immediately after the revision surgery (Fig. 5A, B). Postrevision O-C2 and C1-C2 angles were 43° and 59° , respectively, both of which were larger than the respective preoperative angles.

Discussion

Postoperative dysphagia in patients undergoing cervical spine surgery has been well documented [1-6]. Smith-Hammond *et al.* reported almost half of the anterior and more than 20% of posterior cervical spinal surgery patients demonstrated dysphagia on postoperative videofluoroscopic swallow evaluation [8]. They described the risk factors as anterior surgery, age, use of instrumentation, duration of surgery, spinal levels involved, and the number of spinal levels fused. A posterior occipito-cervico-thoracic fusion in a flexed position may cause dysphagia or dyspnea [9].

There are a few reports of postoperative dysphagia after occipito-cervical fusion, and Yoshida reported that even a short fusion like occipitoaxial fusion may cause respiratory failure [7]. The major causes of the pharyngeal troubles after posterior cervical fusion are suspected to be obstruction of the pharyngeal space induced from the flexed position and the loss of range of motion. The occipito-C1-C2 complex is responsible

for 40% of all cervical flexion-extension and for 60% of all cervical rotation [10]. Panjabi *et al.* reported that the mean extension ranges of motion of O-C1 and C1-C2 in a fresh cadaver experiment were 21.0° and 10.9° , respectively [11]. The occipito-C1 joint is primarily responsible for extension. Fusion without the occipito-C1 joint preserves the extended motion of the neck and is unlikely to cause an obstruction of the pharyngeal space. Actually, there are no reports of postoperative dysphagia after upper cervical fusion without the occipitoaxial joint.

The fixation angle was thought to be the most important factor in preventing postoperative pharyngeal troubles. Matsunaga reported that the O-C2 angle was important [12]. Takami reported that the angle between the McGregor line and the posterior longitudinal line of the C2 vertebra (ACVJ: the angle of the craniovertebral junction) was the most reliable [13]. These angles were measured in our patient (Table 1). The pre-operative O-C2 angle and ACVJ were 37° and 119° , respectively, whereas the post-operative O-C2 angle and ACVJ were 27° and 106° . The post-revision O-C2 angle and ACVJ were 43° and 122° , respectively. The normal O-C2 angle and the normal ACVJ in males over 60 years of age are about 15° [12] and about 100° , respectively [13]. Our patient's postoperative O-C2 angle and ACVJ were closer to the normal controls than to the preoperative angles. In addition, these angles were larger than the normal controls, indicating his neck was fixed in an extended position. Nevertheless, trismus and dysphagia occurred after the operation. It is true that the postoperative O-C2 angle was 27° , indicating his post-operative neck position was extended.

This patient had abnormal profiles of these preoperative angles. His O-C2 angle was 10° lower after the first operation. Was this rather small decrease a critical factor? The cervical laminoplasty and impingement of the C1 lateral mass screw with the occipital bone were examined.

His C4 and C5 were fused congenitally, and cervical laminoplasty was done. Baba reported that the cervical range of motion (ROM) was decreased after cervical laminoplasty [14]. Did the ROM decrease in the subaxial cervical spine cause our patient's symptoms? Actually, his preoperative and postoperative C2-C7 ROM were 30° and 27° , respectively. ROM decreased by only 10% in this case. The post-revision

Table 1 Measurement of the O–C2 angle, ACVJ, O–C1 angle, and segmental ROM

		Pre-operation	Post-operation	Post-revision
O–C1 (°)	Neutral	–19	–16	–16
	Flexion	–23	–16	–20
	Extension	–12	–13	–14
C1–C2 (°)	Neutral	56	43	59
	Flexion	43	43	62
	Extension	65	40	63
O–C2 (°)	Neutral	37	27	43
	Flexion	20	27	42
	Extension	53	27	49
ACVJ (°)	Neutral	119	106	122
	Flexion	97	107	122
	Extension	133	108	125
ROM (°)	O–C1	11	3	6
	C2–C7	30	27	27

O–C1 angle: the angle between the McGregor line and the line from the upper end of the anterior arch to the upper end of the posterior arch; C1–C2 angle: the angle between the line from the upper end of the anterior arch to the upper end of the posterior arch and the inferior surface of the axis; O–C2 angle: the angle between the McGregor line and the inferior surface of the axis; ACVJ: the angle between the McGregor line and the posterior longitudinal line of the C2 vertebra; plus means lordosis and minus means kyphosis.

ROM was 27°. His symptoms decreased after the revision despite the lack of change in ROM after it. The decrease in the subaxial ROM after laminoplasty had no significant effect on his complaints.

The screw heads were in contact with the occipital bone, possibly interrupting the extended motion. The O–C1 angle and O–C1 ROM were measured. The O–C1 angle was measured between the McGregor line and the line from the upper end of the anterior arch to the upper end of the posterior arch, because the center of the posterior arch was not visible after implantation. The pre-operative and post-operative ROM in O–C1 were 11° and 3°, respectively. The pre-operative and post-operative O–C1 angles in the extended position were –12° and –13°, respectively. The O–C1 ROM was decreased but the O–C1 angle in the extended position showed little change. On the other hand, the pre-operative and post-operative O–C1 angles in flexed position were –23° and –16°, respectively. He developed a limitation in flexion motion in the O–C1 segment. The O–C1 angle in the extended position was –14° and increased only a little after the revision. As a result, dysphagia and trismus

arose after the first operation and diminished after the revision. The implant bulge probably played no major role in his symptoms. However, an implant bulge may cause a limitation of O–C1 extension. The posterior arch of the atlas was hypoplastic, so the space between the occipital bone and the C2 spinous process was small. C1 lateral mass screws were inserted by Tan's method, and the screw heads were higher than that in Goel's method. Tan's method has many advantages, such as less bleeding or less invasion to the C2 nerve root. Impingement between the screw head and the occipital bone might be one of the pitfalls of this procedure. The literature contains no reports of impingement between various implants, such as wires or screws, and the occipital bone. It is important to know the potential risk of implant impingement to the occipital bone.

This patient complained of trismus as well as dysphagia. There are many reports on trismus or limitations of the mouth after radiation therapy [15], surgery in the neck region [16], or after tetanus [17], but no reports after surgery of the cervical spine. In fact, his symptoms arose immediately after the first

operation and were relieved immediately after the revision. The relatively flexed position in the C1–C2 segment was assumed to be the cause of these symptoms. The postoperative C1–C2 angle was 43°. The average C1–C2 angle in 201 persons with various cervical alignments was reported to be around 9° [18]. This seemed to mean the fixation angle was in a hyper-extended position. Surprisingly, however, this C1–C2 angle was equal to the preoperative C1–C2 angle in the flexed position. After the revision, the C1–C2 angle was around 60°, **larger than the preoperative angle in the neutral position.**

Some reports describe the normal profiles of the occipito-cervical joint, but there are no reports of a benchmark position for pharyngeal troubles. This was our patient's greatest pitfall with abnormal cervical alignments. The angles in the neutral position are probably the safest. A radiograph of the neutral position is taken when the patient relaxes. It is unlikely that pharyngeal and neurological troubles would occur in this relaxed position. A fluoroscope is often used in surgeries such as cervical fusion. The O–C2 angle or ACVJ is easily observed during surgery. The normal ranges of these factors are well known, but they may not always correspond to the best alignment for each patient. Measuring the O–C2 angle or ACVJ in the pre-operative neutral position before surgery and comparing that with the intra-operative angles may be the simplest strategy for avoiding pharyngeal problems.

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