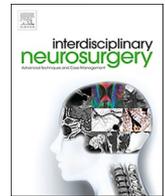


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Technical notes & surgical techniques

C1 laminoplasty and posterior atlantoaxial fusion for large retro-odontoid pseudotumor with Instability: A technical note

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ABSTRACT

Background: Retro-odontoid pseudotumor (ROPT) is a non-neoplastic pathology of the craniovertebral junction that is usually associated with atlantoaxial instability. The mass compresses the spinal cord, causing cervical myelopathy and potentially resulting in severe disability. Posterior atlantoaxial fixation without laminectomy is a suitable surgical option when the symptoms are mild and the static compression is moderate. In the setting of patients with severe symptoms and large ROPTs, posterior decompression becomes necessary. However, achieving solid posterior atlantoaxial fusion is difficult without a bony surface, namely the C1 posterior arch. Here, we describe a novel technique of C1 laminoplasty to achieve C1 decompression, and posterior atlantoaxial fusion with a modified Goel technique.

Methods: An 83-year-old man was referred to our orthopedic department with quadriplegia due to atlantoaxial instability. His daily life had been affected by severe neck pain, clumsiness of bilateral hands, and gait disturbance for more than 2 years. Preoperative cervical radiographs revealed atlantoaxial instability, and magnetic resonance imaging showed a large pseudotumor compressing to the spinal cord.

Results: The patient underwent C1 laminoplasty to decompress the spinal cord and retain the posterior arch as a bony surface for grafting and posterior atlantoaxial fusion. The procedure was well tolerated. The patient's quadriplegia was improved and his Japanese Orthopedic Association score improved from 9/17 to 15/17 and his visual analog scale score for neck pain improved from 75 mm to 28 mm at the 1-year follow-up.

Conclusion: C1 laminoplasty and posterior atlantoaxial fusion appears useful when C1 decompression and atlantoaxial fusion become necessary.

1. Introduction

Retro-odontoid pseudotumor (ROPT) is usually a reactive fibrocartilaginous proliferation or pannus formation of the odontoid process and neighboring anatomical structures of the craniovertebral junction (CVJ). ROPT is generally associated with atlantoaxial instability (AAI), diffuse idiopathic skeletal hyperostosis, rheumatoid arthritis, psoriatic arthritis, hemodialysis, and other pathological conditions that cause mass-like changes [1–9]. When ROPT becomes large and causes cervical myelopathy, surgical intervention is mandatory [2–4,8,9]. Several reports have shown that posterior atlantoaxial fusion

(PAAF) without decompression can reduce ROPT and its symptoms, so C1 laminectomy as a means of decompression is unnecessary [2–4,10,11]. However, simple C1 laminectomy might be a suitable option in the elderly and for patients without obvious AAI, as the intervention is minimally invasive and carries a low rate of complications [12,13]. For patients with large ROPTs and/or severe myelopathy, C1 laminectomy with posterior occipitocervical fusion is recommended because of the superior surgical outcomes [11,14,15]. The time required for regression of ROPT is shorter when compared with fusion alone [15], but the risks of dyspnea and dysphagia are higher [16]. Considering these issues, we describe a novel technique of “C1 laminoplasty and

Abbreviations: AAI, Atlantoaxial instability; CT, Computed tomography; CVJ, Craniovertebral junction; JOA, Japanese orthopedic association; LM, Lateral mass; MRI, Magnetic resonance imaging; PAAF, Posterior atlantoaxial fusion; ROPT, Retro-odontoid pseudotumor; VAS, Visual analogue scale.

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PAAF” for the treatment of patients with large ROPTs and severe myelopathy.

2. Case presentation

Due approval from the institutional ethics committee was obtained for this study. Necessary consents were obtained from the patient.

2.1. Patient history

A previously healthy 83-year-old man with no history of systemic disease was referred to our orthopedic department for quadriparesis due to atlantoaxial instability. His daily life had been affected by severe neck pain, clumsiness of bilateral hands, gait disturbance for more than 2 years. He had shown gradual deterioration over the last 4 months and had become disabled. Treatment with a soft collar and rehabilitation were recommended at another medical facility, but the conservative approach proved unsuccessful and the neurological condition of the patient deteriorated to quadriparesis. Preoperative Japanese orthopedic association (JOA) score of the patient was 9/17 and visual analogue scale (VAS) for neck pain was 75 mm.

2.2. Physical examination

The patient was unable to walk or stand unaided at the time of admission. He showed clumsiness in both hands, and numbness in both hands and feet. The patient also described severe neck pain and showed a limited range of neck motion. He reported urinary and bowel disturbances. Hyperreflexia of the legs and bilateral upgoing toes were observed on examination.

2.3. Preoperative imaging

Radiography on the initial visit demonstrated AAI with atlantodental distance of 5 mm in the flexed position (Fig. 1A). Preoperative computed tomography (CT) showed slight erosion of the dens, and spontaneous fusions of the C2–3 and C4–5 vertebrae. Preoperative magnetic resonance imaging (MRI) revealed severe stenosis of the spinal canal at the C1–2 level, and compression of the spinal cord by ROPT (Fig. 1B–E).

2.4. Treatment decision

Based on the clinical condition of the patient, with progressive neurological deterioration, severe quadriparesis and upper motor

neuron findings, as well as the results of radiological studies showing spinal canal stenosis, cord compression, and AAI due to ROPT, surgery was indicated.

2.5. Surgery and postoperative images

C1 laminoplasty and PAAF were performed (Fig. 2A, B). Post-operative CT revealed successful decompression and correct screw positioning (Fig. 2C–F). The surgery took 2 h 50 min, and estimated blood loss was 150 ml. The patient tolerated the procedure well, and moderate pain relief was noted immediately postoperatively. He was able to walk with a cane at the time of discharge from hospital.

2.6. Follow-up results and images

Four months after surgery, the patient was able to walk without a cane, and sensation in both hands and feet had improved. He was capable of most personal activities at the 1-year follow-up. Final follow-up CT demonstrated solid bony fusion of C1–2 and decompression of the spinal canal (Fig. 3A, B). In terms of clinical outcomes at the 1-year follow-up, JOA score had improved from 9/17 to 15/17, and VAS for neck pain had improved from 75 mm to 28 mm (63%).

3. Operative procedure and technical implications

3.1. Patient positioning

The patient is placed in a prone position on an adjustable hinged carbon operating table (OSI Axis Jackson table; Mizuho, Union City, CA, USA) to perform CT using an O-arm (Medtronic Sofamor Danek, Minneapolis, MN, USA). The surgical procedure is performed under neuro-monitoring. To easily access C1, the neck is positioned in a slightly flexed fashion, and the head is fixed with a special carbon three-pin fixation device. Checking for vascular anomalies and a high-riding vertebral artery on preoperative images is crucial.

3.2. C1 laminoplasty

A midline posterior cervical skin incision of approximately 7 cm is made then the cervical paraspinal muscles are bilaterally dissected, exposing the atlas and axis. The reference frame is attached to the spinous process of C2. The O-arm is then positioned, and 3-dimensional reconstructed CT images are obtained and transmitted to the StealthStation™ surgical navigation system (Spine 7^R; Medtronic Sofamor

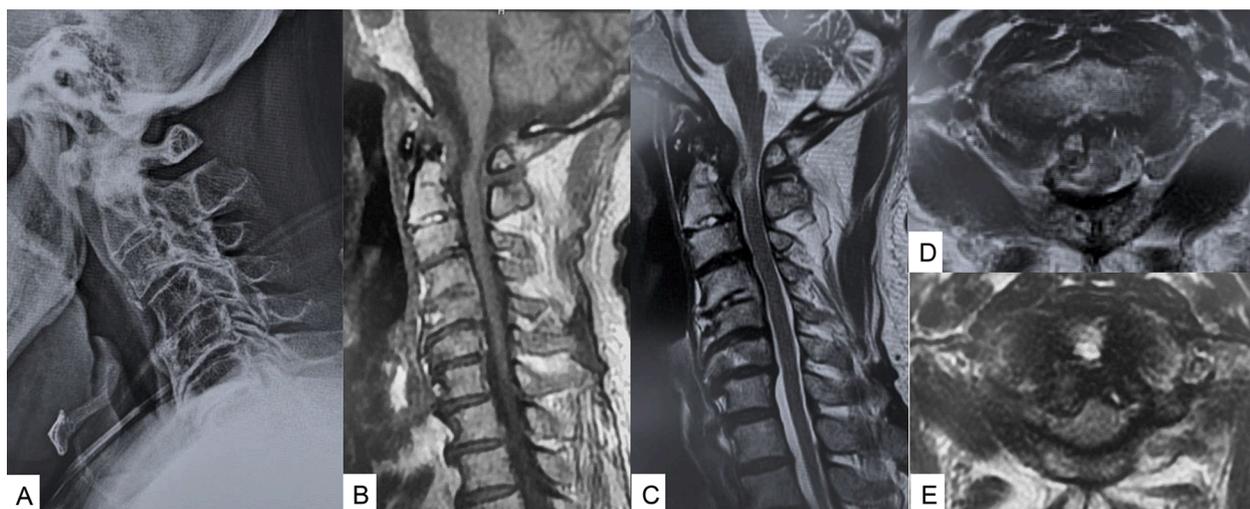


Fig. 1. Preoperative radiograph and MRI A) Lateral radiogram in flexion. B) Mid sagittal T1 weighted image. C) Mid sagittal T2 weighted image. D) Axial T1 weighted image at C1 E) Axial T2 weighted image at C2.

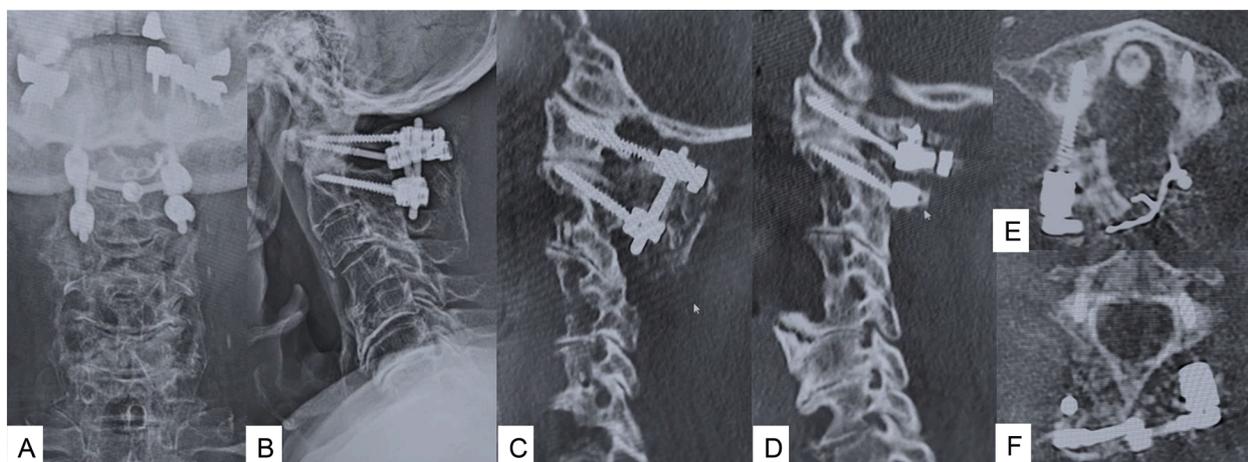


Fig. 2. Postoperative images: A) Anteroposterior radiogram. B) Lateral radiogram. C) Right parasagittal 3D reconstructed image. D) Left parasagittal 3D reconstructed image. E) Axial image at C1 level. F) Axial image at C2 level.

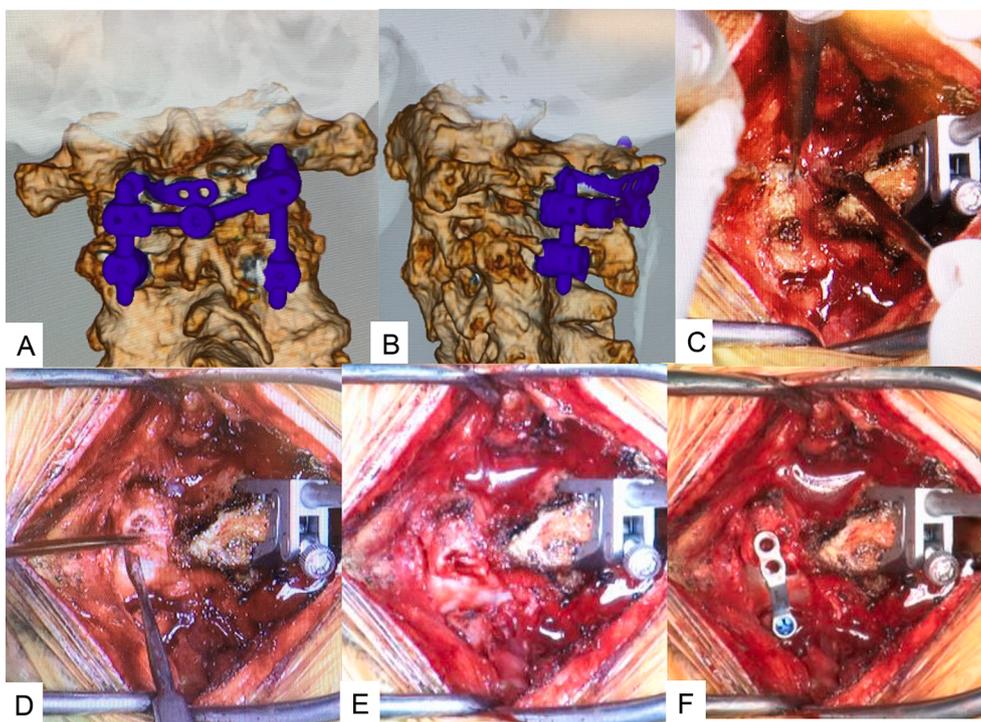


Fig. 3. CT scan at 12 months' follow-up and C1 laminoplasty. A) 3D CT posterior aspect. B) 3D CT lateral aspect. C) Bilateral gutter for laminoplasty. D) Separating the adhesion with a Penfield dissector. E) Left side posterior arch is open. F) Laminoplasty with a plate.

Danek). After verifying every navigated spinal instrument, the accuracy of navigation is checked. The best cutting point for C1 laminoplasty is marked using a navigated pinpoint probe. Typically, the cutting points should be approximately 15–18 mm from the center of the posterior tubercle of C1. Using a navigated high-speed burr, bilateral gutters are created for the laminoplasty to avoid injuring the C2 nerve roots and vertebral arteries (Fig. 3C). According to the method described by Hirabayashi et al. [17], C1 laminoplasty was performed using a laminar plate (Figs. 3D–F, 4). Again for this step, attention should be paid during screwing to avoid injury to the vertebral artery, which is located just cranial to the gutter.

3.3. Posterior atlantoaxial fusion (modified Goel technique)

After C1 laminoplasty and plating, the entry point for the C1 lateral

mass (LM) is marked with a probe. Using the navigated high-speed burr (Electric Stealth-Midas; Medtronic Sofamor Danek), an entry is made in C1 LM, and a C1 LM screw is inserted using the Lees notching technique [18] (Fig. 5A). The entry point for the C2 pedicle screw is then marked, and the navigated high-speed burr is used to make the hole. The surgeon should avoid any pressure on C2 because of the spinal cord compression and AAI. A navigated pedicle probe and navigated tap are used, and C2 pedicle screws are inserted (Fig. 5B). Intraoperative O-arm CT is again performed to ensure correct placement of the screws. Rods are then inserted and attached, and the subluxation is reduced (Fig. 5C). When tightening the screws to the rods, care must be taken to avoid creating a gap between C1 and C2. Finally, a transverse crosslink connector is fixed between the C1 screws, and PAAF is performed using autogenous iliac bone graft (Fig. 5D). Immobilization of the patient with a soft neck collar is recommended for one week after the operation.

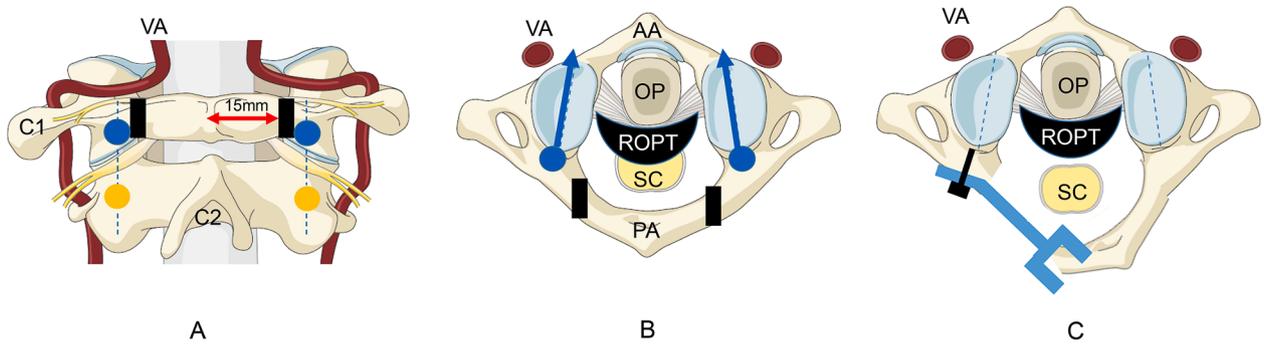


Fig. 4. Schema of C1 laminoplasty VA; vertebral artery, AA; anterior arch, PA; posterior arch, OP; odontoid process, ROPT, Retro-odontoid pseudotumor, SC; spinal cord A) Posterior schema of C1 and C2; C1 gutters (black line) are made 15 mm from the center. B) Axial C1 schema before C1 laminoplasty; C1 gutter (black line) C) Enlarged laminae is maintained by a plate (blue) and a screw (black).

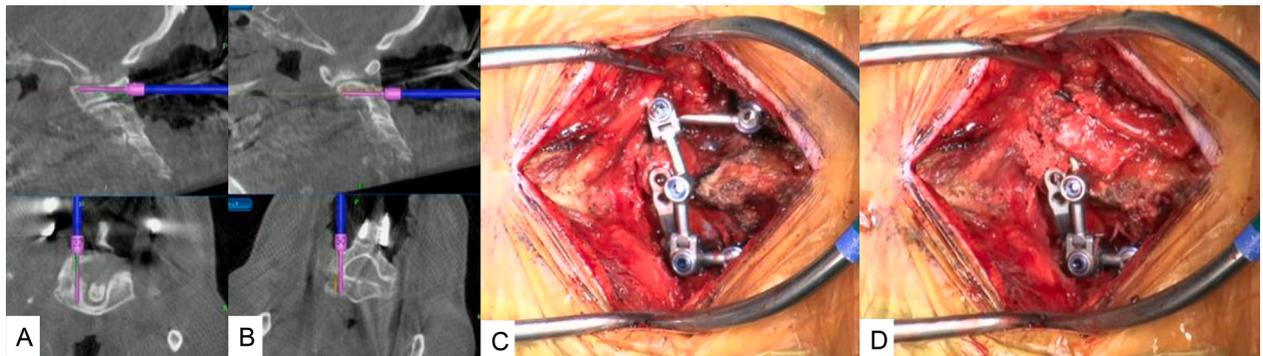


Fig. 5. Screwing, reduction, and bone graft A) C1 lateral mass screw B) C2 pedicle screw C) Reduction. D) Bone graft.

4. Discussion

In this report, we have presented a novel surgical technique for the treatment of ROPT. Our new technique involved C1 laminoplasty with a plate for spinal cord decompression, and additional PAAF using a

modified Goel technique for AAI. Moreover, autologous bone graft was applied for bony fusion. All procedures were performed under O-arm CT and spinal navigation without any radiation exposure to operating room personnel.

The etiologies of the ROPT are infectious process such as

Three methods of C1 LMS placement

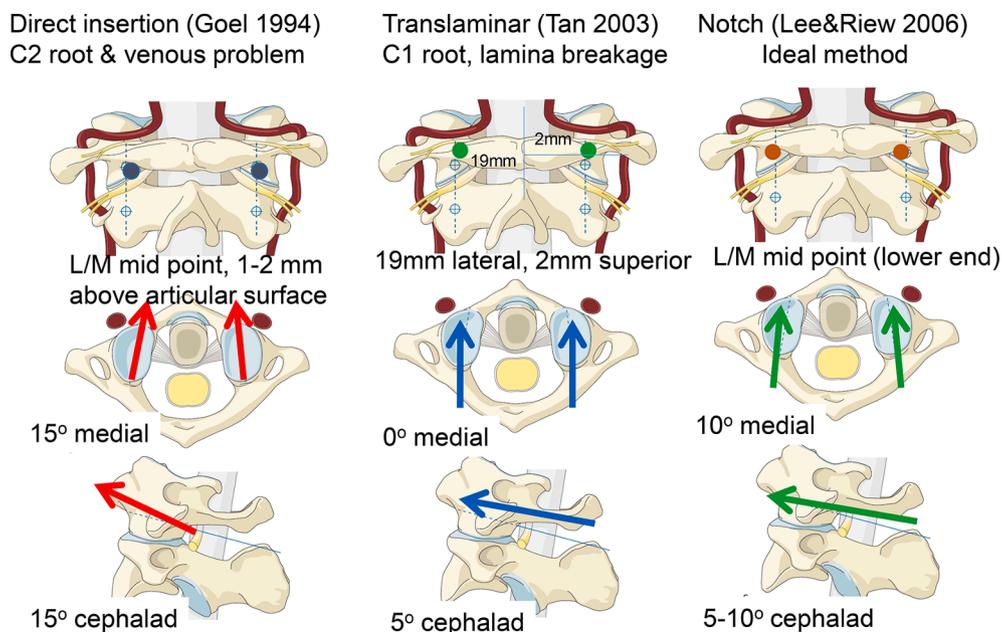


Fig. 6. Three methods of C1 lateral mass screw placement.

mycoplasma, inflammatory granulation tissue of rheumatoid arthritis [19], or gout [20], reactive tissue of mechanical instability due to Os odontoideum, AAI [21] or without instability [22]. The *retro*-odontoid pathology lies in the anterior part of the spine, surgical treatments were initially defined using direct routes by transoral, transsphenoidal, transpharyngeal, or anterolateral approaches to excise the lesion and decompress the neural tissues [2,3,14,23–26]. Over time, the pathophysiology of *retro*-odontoid tissue has been better understood, and more recently, without directly addressing the pathology, posterior surgical interventions to the CVJ (with or without decompression, with or without fixation) have been accepted as providing susceptible, reliable, and effective treatment in the majority of cases [2–4,8,9,27,28].

Understanding the CVJ anatomy, biomechanical state, and systemic condition of the individual as well as the neurological status drive the spine surgeon to make the right decisions while choosing surgical interventions for ROPT. Various classifications and treatment algorithms have been reported [2–4,8,29,30]. Briefly, in a patient with a prominent AAI, if the anatomy of the C0–C1–C2 joints is normal with no signs of instability at the C2–C3 level, C1–C2 posterior fixation using a Magerl [31] or Goel-Harms [32,33] technique is sufficient to achieve solid fixation [2,3,27,28]. In our new method, we utilized Notch technique for C1 lateral mass screw to avoid vertebral artery injury (Fig. 6) [18]. On the other hand, occipitocervical fixation becomes a valid method in settings of a severely affected atlanto-occipital joint or congenital malformations of the occipital bone and/or atlas [2,3,28,34]. The best surgical strategy in the absence of any signs of overt AAI remains under debate [8,12,28]. In such cases, a simple posterior neural decompression by performing C1 laminectomy or C1 laminoplasty is considered sufficient, providing less-invasive intervention with minimal surgery-related risks [12,13,35]. In contrast, relieving the mechanical stress on the atlantoaxial joint and preventing progression while inducing regression of the ROPT by atlantoaxial fixation is beneficial and recommended [1,2,4,8,28]. Of note, an additional decompression procedure (C1 posterior arch resection) is necessary in cases with severe cord compression, and is also commonly performed for early neural decompression while awaiting ROPT reduction [1,2,28]. In such a situation, occipitocervical (OC2) fixation is preferable because posterior atlantoaxial fusion become difficult without C1 posterior arch. From such descriptions to date, evaluation of the abovementioned characteristics is critical when tailoring a treatment plan.

Here, the clinical condition of this patient with progressive neurological deterioration, severe quadriparesis and upper motor neuron findings without any known history of systemic disease, as well as the results of radiological studies (i.e., spinal canal stenosis, cord compression, and AAI in accordance with ROPT) provided the indications for surgery. We performed C1 laminoplasty using a laminar plate and PAAF under O-arm CT and with spinal navigation. The unique parts of this technical report were: 1) the use of a laminar plate for C1 laminoplasty; 2) the implantation of transverse crosslink connectors between C1 lateral mass screws in a laminoplasty case combined with PAAF; and 3) the application of O-arm CT and spinal navigation to make C1 gutter and C1 lateral mass screws and C2 pedicle screws. Suetsuna et al. were the first to report C1 laminoplasty for the treatment of ROPT with successful clinical outcomes [35]. Technically, they performed midline splitting plus bilateral lateral gutters, and inserted a hydroxyapatite spacer fixed with a suture [35]. Because of the risk of artificial bone fracture, and the inconvenient anatomical characteristics of the C1 posterior arch (i.e., small and fragile, rarely allowing tight contact with artificial bone and stabilization), Lee et al. introduced a new technique for PAAF combined with C1 double-door laminoplasty augmented with an allograft spacer and titanium miniplate [36]. Later, in attempting to achieve safer and better decompression, Tarukado et al. performed C1 laminoplasty without fusion in some patients with compressive myelopathy without obvious instability at the C1 level [33]. They used a laminoplasty technique similar to that described by Suetsuna et al. [35], and identified C1 laminoplasty as a viable alternative procedure to laminectomy

[37]. Here, we chose laminoplasty for decompression as a method providing adequate decompression while ensuring an appropriate bone-grafting site and maintaining a normal anatomical shape without the disadvantages of laminectomy (i.e., the risks of C1 anterior arch fracture or postoperative spinal cord compression due to dynamic paraspinous muscle impingement or scar tissue formation) [35–37]. As a commonly used laminoplasty technique for the subaxial cervical spine, a modified Hirabayashi method with a laminar plate was performed at C1, possibly representing the first description of this in the English medical literature. This was combined with PAAF. Moreover, as a method yielding early bony fusion [38], an on-the-screwhead transverse crosslink connector was applied between C1 lateral mass screws in this case. Of note, we performed all these procedures under O-arm CT and spinal navigation, minimizing the hazards of radiation exposure [39].

Despite the beneficial features, this technique shows several potential disadvantages. As a rare pathology localized in an anatomically complex region, the method absolutely requires experienced, up-to-date surgeons for treatment success [36]. Drilled gutters must be situated correctly to avoid injury to the C2 nerve roots and vertebral arteries. Moreover, when drilling the closed side, the surgeon should avoid creating a true fracture. From the perspective of spinal navigation, the reference frame must be positioned safely. If moved, the risk of errors and complications is increased [40], so surgeons should be aware of and checking the navigational accuracy in every step; if any uncertainty arises, another O-arm CT should be performed.

5. Conclusion

If the ROPT is large, atlantoaxial subluxation is overt, and the symptoms are severe, posterior decompression with posterior atlantoaxial fixation appears to offer the ideal surgical treatment. To achieve this goal, we applied the novel technique of spinal cord decompression by means of C1 laminoplasty with additional PAAF, under the guidance of both O-arm CT and spinal navigation. This technique helps the spine surgeons achieve solid atlantoaxial fusion and good clinical outcomes for the patient while also minimizing radiation exposure to the operating room staff.

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CRedit authorship contribution statement

Masato Tanaka: Writing – original draft. **Selim Ayhan:** Writing – review & editing. **Taro Yamauchi:** Data curation. **Shinya Arataki:** Data curation. **Yoshihiro Fujiwara:** Data curation. **Akihiro Kanemaru:** Data curation. **Shin Masuda:** Data curation. **Kenta Torigoe:** Data curation. **Yasuyuki Shiozaki:** Writing – review & editing.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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