



CASE SERIES

Risk factors and surgical approaches in neglected subaxial cervical spine fractures-dislocations: Experiences with two cases and literature review

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Key Clinical Message

This case report describes our experience of surgical strategies of two patients with neglected subaxial cervical spine fracture-dislocation that came to our center with subsequent follow-ups. Subaxial cervical spine fracture-dislocation must be immediately diagnosed and treated. However, it can be neglected in some cases, especially in developing health care systems and patients with low socioeconomic status. We reported two neglected subaxial cervical fracture-dislocation with a mean age of 54 years old who presented with axial cervical pain, and decreased muscle forces. In one out of two, cervical closed traction was applied, then unsuccessful result led to circumferential decompression and fixation via anterior-posterior (AP) approach. Accordingly, we used AP approach without applying closed reduction in another patient successfully. Except one of our cases who died after 2 weeks of surgery due to aspiration pneumonia, other one found complete improvement at the end of 6-month follow-up. Our study emphasizes the importance of AP approach in patients with irreducible joint dislocations. The approach can minimize the surgical risks and increase the cost-benefit as compared to three or more staged approaches. Our approach is less intensive than some other AP approaches while is a safe and efficacious procedure since the posterior reduction is not performed before discectomy and decompression.

KEYWORDS

case report, cervical, dislocation, neglected, surgery

1 | INTRODUCTION

Subaxial cervical spine fracture-dislocation must be immediately diagnosed and treated.¹ However, it can be neglected in some cases, especially in developing health

care systems and patients with low socioeconomic status.² Moreover, living in the rural areas, inaccessibility of nearby specialized medical centers, unconsciousness and multiple traumatized patients account for the other risk factors predisposing to negligence of the spine.^{2,3} If the

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correct diagnosis occurs after 3 weeks of injury, it is considered as a delayed (neglected) subaxial cervical spine fracture-dislocation.⁴ These injuries often lead to neurological deficits, chronic pain, and cervical deformity.^{5,6} There is no clear approach for treatment of neglected subaxial fracture-dislocation in guidelines,^{7,8} but a few literatures have suggested algorithms to obtain maximum alignment and resolve neurological symptoms.^{2,7,9,10} Several surgical strategies such as single anterior,⁶ single posterior,¹¹ posterior–anterior,^{12–15} posterior–anterior–posterior,^{7,16} and anterior–posterior–anterior–posterior⁷ approaches have been reported but more studies are needed to conclude a definite protocol. We report our experience of surgical strategies of two patients with neglected subaxial cervical spine fracture-dislocation that came to our center with subsequent follow-ups.

2 | CLINICAL PRESENTATION

After the approval from Institutional review board and the research ethics committee, this study was conducted as a retrospective single-center case series including five consecutive cases who signed the consent form. The data that support the findings of this study are available from the corresponding author, upon reasonable request. This case series has been reported in line with the PROCESS Guideline.¹⁷

3 | CASE 1

A 36-year-old man presented in our clinic with complaints of disability in walking for the last 3 months. He had a history of falling from a height of 5 m, 3 years ago for which he underwent no operation and since then, he has had chronic neck pain. About 3 months ago, the neck pain diminished and gradually he lost the ability to walk. He also mentioned giving way of the knee and sudden dropping objects from his hands every so often. On neurological examination, he had American Spinal Injury Association (ASIA)-C, and Japanese Orthopedic Association (JOA) score of 12 accompanied by myelopathic presentation. The preoperative CT scan and MRI showed C5 fracture as well as kyphotic deformity of cervical spine (Figure 1). Regarding irreducible dislocation after applying the cervical traction, we planned a circumferential surgery (360-degree approach) for this patient. For the anterior–posterior (AP) approach, first, C4 and C5 corpectomy, adjacent discectomy, and insertion of adjusted cage were done via anterior approach. Three days later, the second surgery was conducted through a posterior approach for decompression and fusion. Few weeks

after the second surgery, the patient gradually regained the ability to walk with no neurological symptoms. At a follow-up of 6 months, there was acceptable bone fusion in surgical site and his ASIA-B, JOA score was 15.

4 | CASE 2

A 72-year-old man presented with complaints of quadriplegia. He had a history of a car accident 2 years ago which resulted in progressive neck pain, urine and stool incontinence, and inability to walk. On neurological examination, he had ASIA-A, and JOA score of 10. His cervical CT scan showed C5–C6 spondylolisthesis, and kyphotic deformity (Figure 2). After the irreducible spondylolisthesis was confirmed, we decided to perform anterior decompression and fusion by C5–C6 corpectomy and adjacent discectomy in the first session. After 72 h, posterior laminectomy and fusion were done. For the AP approach he obtained his preoperative condition but was intubated due to an aspiration pneumonia a week later which led to death after 2 weeks.

5 | DISCUSSION

Subaxial Cervical spine subluxation and dislocations comprise only 2%–3% of all the injuries due to blunt trauma, but the significantly high morbidity and mortality rates attached to it pose a challenge for the healthcare providers (HCPs).^{18,19} In approximately 4%–30% of the cases, the diagnosis is missed even after prompt utilization of radiological evidence.^{20–22} Various reasons have been reported for the same including improper interpretation of imaging, unsatisfactory radiographs, and ineffective examination due to altered sensorium.^{23,24} Most frequent cause identified was the misinterpretation of radiological imaging.²⁵ Consequently, patients present with symptoms that are way different as compared to acute trauma patients. Goni et al.¹⁴ hence recommended a radioscopy along with a three view cervical spine series in patients who were alert.

There are no clear-cut criteria that define “old” or “neglected” subaxial cervical spine injury, different studies have stated different criteria. For instance, any injury more than 2 weeks was considered “old” by Braakman et al.²⁶ 3 weeks was the cutoff in the study by Kiwerski et al.²⁷ while Bartels et al.⁷ kept it to as high as 8 weeks. However, most studies now have reported the cutoff to be 3 weeks in their studies and in our study as well, any injury post 3 weeks was considered “old” or “neglected”. Neglected cases present a surgical inconvenience for the HCPs since post 3 weeks injuries lead to contracture of surrounding

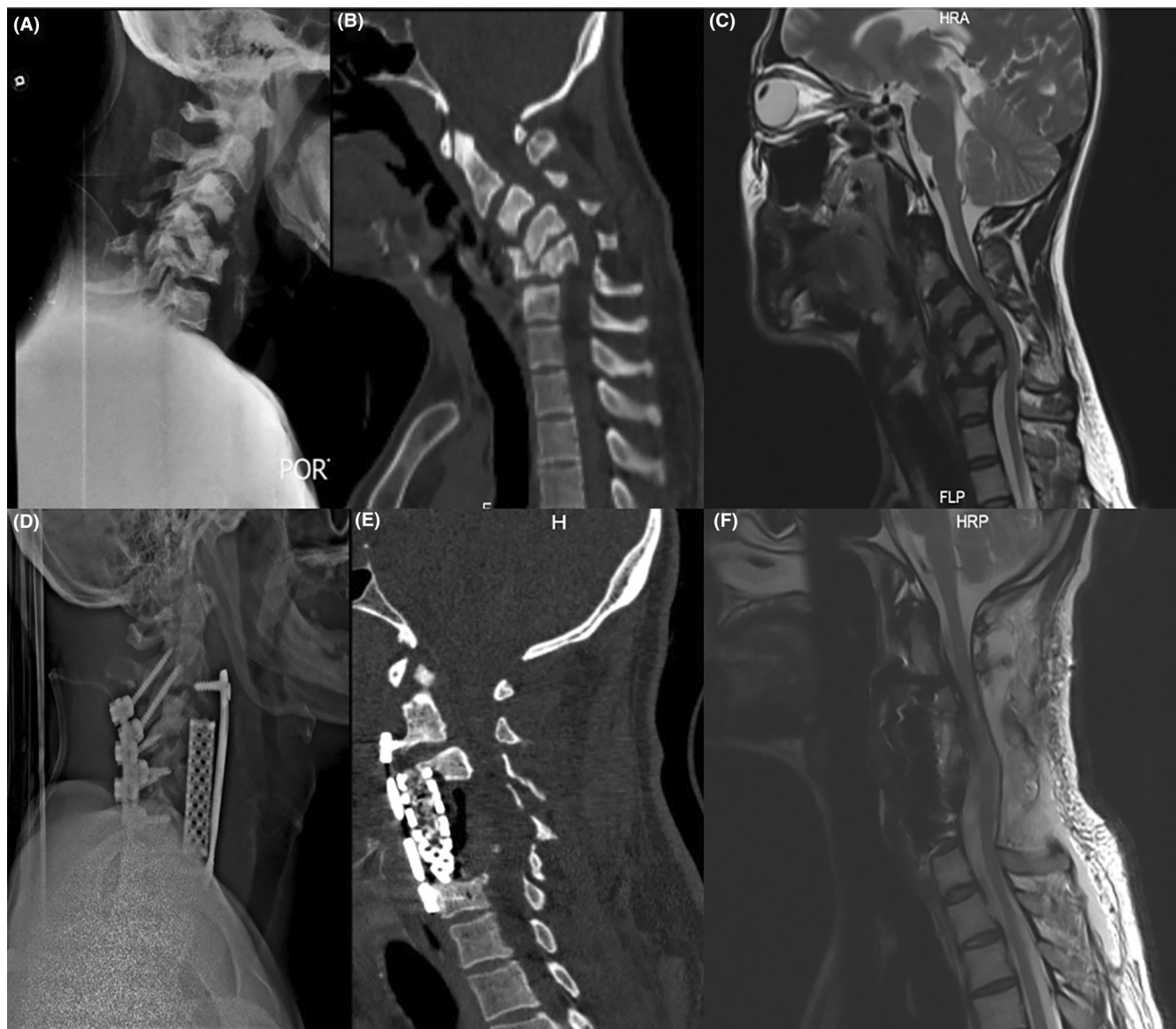


FIGURE 1 Preoperative (A) neck x-ray, (B) CT scan, and (C) MRI (T2-weighted sagittal view) show cervical kyphosis leading to severe spinal cord compression due to the previous trauma. Postoperative counterpart (D) x-ray, (E) CT scan, and (F) MRI (T2-weighted sagittal view) display spinal cord decompression and maximal safe correction via anteroposterior approach (AP approach).

structures thereby complicating the reduction procedure. Rather the success rate of closed reduction substantially fell to 20% in patients with dislocations diagnosed after 72 h of the injury from 64% in patients having diagnosed and treated for fresh dislocations.²⁸

We performed a review of literature to understand the presentation and management of neglected dislocation cases to aid in better neurosurgical decision making and patient care. Most common clinical presentations were neck pain with restricted neck motion, myelopathy, and radiculopathy. Other less common symptoms included loss of sensation, quadriplegia, and inability to move the limbs. In some cases, the patients might be asymptomatic for months and even years and by that time, the treatment becomes nearly unachievable. Hence, radiological

imaging holds considerable significance in the diagnosis and treatment of neglected dislocations. However, occasionally a simple radiograph may not show anything despite the patient complaining of pain. In such cases it is advisable to opt for more advanced radiological techniques like computed tomography (CT) or magnetic resonance imaging (MRI). Role of MRI in identifying the anterior disc herniation that could be highly dangerous during closed reduction is unclear. After closed reduction under general anesthesia, there have been reports of neurological deterioration,²⁹ while in patients who are awake and cooperative, it might be safe without a pre reduction MRI.³⁰ Therefore, in patients who are difficult to examine or uncooperative, it is suggested to have a pre reduction MRI. Bechet et al. suggested to carry out open reduction

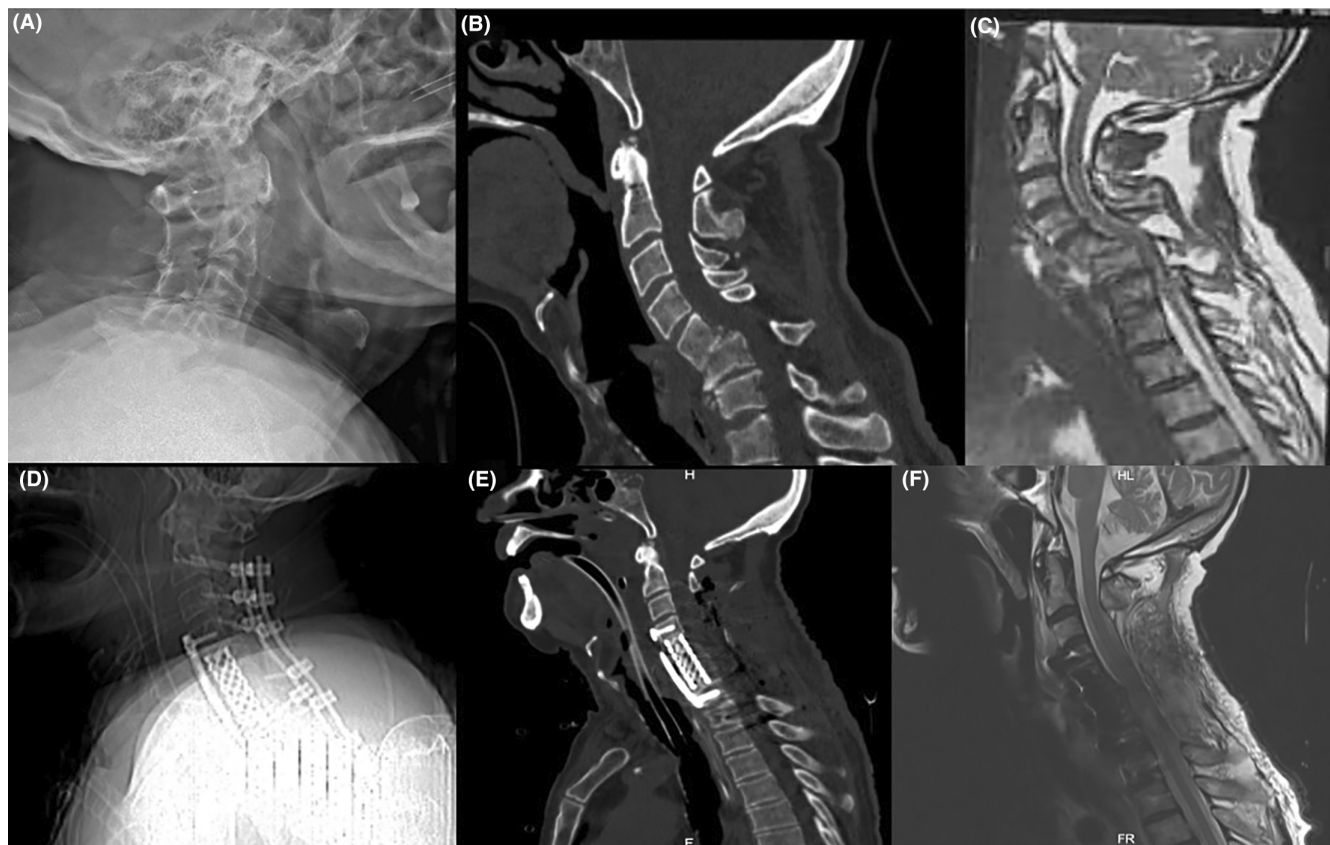


FIGURE 2 Preoperative (A) neck x-ray, (B) CT scan, and (C) MRI (T2-weighted sagittal view) show neglected C5-C6 fracture-dislocation result in cervical kyphosis. Postoperative counterpart (D) x-ray, (E) CT scan, and (F) MRI (T2-weighted sagittal view) reveal spinal cord decompression and maximal safe correction via anterior C5-C6 corpectomy and fixation, and then posterior laminectomy and fixation (AP approach).

(by anterior approach) in patients with radiological evidence of a disc fragment for removing the whole disc in order to avoid complications during the surgery.⁷

Although little, there is evidence of utilizing skull traction preoperatively for carrying out the reduction procedure. Basu et al.⁸ reported the successful traction of 10 out of 14 patients with unilateral dislocation while 1 in 5 patients with bilateral dislocation at mean duration of 2 days. Skull traction was successful in nearly 20% of the patients with cervical spine dislocations as observed by Kahn et al.³¹ Hassan et al.⁴ reported that even after a long traction of 1 week, only 2 out of 12 patients were successfully managed. In a study by Goni et al., only one out of six patients was satisfactorily aligned. There was no benefit of skull traction in patients with neglected injury of cervical spine for more than 3 weeks as reported by Goni et al.¹⁴ Indeed, skull traction could not realign the cervical spine in our case which is in line with Goni et al. study.

Owing to an extremely low incidence of neglected cases, there is no well-established treatment protocol and hence this still remains a neurosurgical conundrum. Several studies have been conducted to assess the efficacy of various techniques as a treatment modality for

neglected subaxial cervical dislocations. (Table 1) Though the literature is scarce, some conclusions can certainly be drawn from these studies and can be modified accordingly. There is no Level-I of evidence to evaluate these different approaches in neglected cervical dislocations, then surgeons must choose the existing approaches according to their experiences and their operating room setups. Accordingly, we choose the AP approach to treat our patients. In this approach, the least patient turning, and surgical procedure is done which will decrease the surgical time, surgical infection, surgical bleeding, and other probable postoperative complications such as myocardial infarction, esophageal tearing, vascular injuries, and CSF (cerebrospinal fluid) leakage that will be very crucial in elderly patients (our Case 2). The most important concern in this approach is that the surgeon may not be able to return the cervical curvature well.¹⁶ Although, we only had two cases treated via this approach and it needs more study, it is worthy to note that anterior cervical fixation did not make any restriction during posterior release and fixation in none of our cases.

The very first study to be reported was by Hassan et al. in 2002 wherein they managed 12 patients (Mean age:

TABLE 1 The results of the previous studies in the literature.

Author	Year	Patient's number	Mean age (years)	Mean period of presentation (days)	Main symptoms	U/B dislocation	CR	Surgical strategy	Outcome
Hassan et al.	2002	12	50	105	Neck pain Restricted CROM	2 U 10 B	Yes	2 <i>successful CR</i> Anterior fusion alone 10 <i>unsuccessful CR</i> PA	All improved
Bartels et al.	2002	3	71.33	90	Loss of sensibility Hyperpathia Neck pain Decreased CROM Paresis	B	No	2 cases: APAP 1 case: PAP	Full recovery in all
Payer et al.	2006	1	51	70	Neck pain	B	No	APA	Full recovery
Liu et al.	2008	9	45	84	Neck pain Restricted CROM	4 U 5 B	No	PA	Full recovery (8 cases) Mild pain (1 case)
Jain et al.	2010	4	48.2	120	Neck pain Restricted CROM Myelopathy	B	Yes	PA	Full recovery
Shimada et al.	2013	1	76	56	Neck pain, decreased CROM	B	No	PA	Improved
Srivastava et al.	2016	6	51	1260	Neck pain, Decreased CROM Myelopathy Radiculopathy	4 U 2 B	Yes	3 <i>successful CR</i> 1 case: Anterior 3 <i>unsuccessful CR</i> 2 cases: Posterior 1 case: Anterior	Full recovery Mild gait instability (1 case)
Prabhat et al.	2017	15	33	63	Neck pain decreased CROM	5 U 10 B	Yes	5 <i>successful CR</i> 4 cases: Anterior 1 case: AP 10 <i>unsuccessful CR</i> AP	All improved
Ding et al.	2017	17	45.2	96	Neck pain Restricted CROM	10 U 7 B	Yes	CR applied in only 5 cases: <i>Unsuccessful</i> AP	All improved
Marasini et al.	2020	3	48	100	Neck stiffness Quadripareisis	B	Yes	<i>Unsuccessful CR</i> APA	Improved (2 cases) Not improved (1 case)
Jain et al.	2021	1	26	120	Neck stiffness	B	Yes	PA	Full recovery
Bechet et al.	2022	1	70	7	Radiculopathy	B	Yes	Anterior	Improved
Present study	2022	2	54	410	Neck pain, Quadriplegia	B	Yes	CR applied in only 1 case: <i>Unsuccessful</i> AP	Improved (1 cases) Died of aspiration pneumonia (1 case)

Abbreviations: APAP, anterior-posterior-anterior-posterior; CR, closed reduction; CROM, cervical range of motion; U/B, unilateral/bilateral.

50 years) with neglected subaxial cervical dislocations, first on traction to achieve reduction which was achieved only in two patients with bilateral dislocation who subsequently underwent anterior fusion. While in the rest of the patients ($n=10$) reduction was unsuccessful and hence they were treated using the PA approach consisting of the posterior laminectomy (in four patients) or facetectomy (in six patients) followed by traction and anterior fusion and plating. At the last follow-up, all patients improved neurologically but evidence of exaggerated reflexes or some weakness or clumsiness of the small muscles of the hands was there in all patients.⁴

In the same year, Bartels and Donk⁷ described APAP approach for the first time: anterior complete discectomy (attempted reduction), posterior complete facetectomy (for removing the fibrous tissue), and anterior distraction of disc space and plate assisted fixation and finally posterior transpedicular fixation. The failure of attempted reduction in the first two cases was a lesson learnt and hence the authors performed PAP in the third patient: posterior release, anterior reduction and fusion followed by posterior fixation. This approach is beneficial since it avoids multiple turning of the patient which is particularly cumbersome and dangerous when the patient has an unstable spine, and no frames are available.

The APA approach was successfully used in a 51-year-old patient reported by Payer and Tessitore¹⁶ in 2006 without any postoperative complications or deficits. Anteriorly the fibrous tissue is removed followed by posterior reduction of the dislocation and fixation and finally (anterior) insertion of the cage with plating. The greatest advantage of this procedure is the negligible chance of concomitant since the removal of disc precedes the reduction of dislocation. Additionally, the number of times a patient is turned in APA approach (two times) is half as that of PAP (four times) right from intubation until the end of anesthesia. Marasini reported three patients who were treated with an APA approach after failed attempts of closed reduction: anterior decompression, posterior reduction by partial facetectomy, stabilization, and fusion by using lateral mass screws and bone graft, anterior cervical fusion by using a mesh cage, cancellous bone graft, and anterior cervical plating was performed. Two patients improved however, there was no neurological improvement in one patient at a follow-up of 6 months.²

Liu et al.³² provided supportive evidence for PA approach in nine patients with a mean age of 45 years. The sequence of surgery consisted of two stages: posterior procedure (soft tissue release, facetectomy, and interspinous wiring) followed by anterior stage (soft tissue release, discectomy, reduction, intervertebral grafting, and anterior plating). Full anatomical reduction was achieved

in all patients with a decreased frequency of turning the patient. Jain et al. utilized the similar technique in four patients and reported 100% neurological recovery. Jain et al. in 2020 presented a case of a 26-year-old patient with bilateral dislocation who was successfully treated with PA approach.¹⁰ Shimada et al. reported the case of bilateral dislocation which was managed successfully via PA approach as well: Posterior reduction and ACDF without preoperative reduction.¹²

Srivastava et al. performed closed reduction via skeletal traction in 50% ($n=3$) of the patients for 3 weeks beginning with 3 kg till 8–10 kg. These patients subsequently underwent ACDF following a successful reduction. Another 50% of the patients in whom traction was not possible or failed, were treated with posterior tissue release with facetectomy (in cases of failed open reduction). The recommended following clinical points:¹³

1. In cases of failed closed reduction an infeasible open reduction, posterior reduction, and release are accrued out along with excision of superior articular process of the lower vertebra
2. Following a successful posterior reduction, fixation is performed with the involvement of two levels above and below the level of lesion
3. A short-term fixation with the involvement of one level above and below the site of lesion can be carried out after ACDF in patients with preoperative neurological deficit or young age.

Series of 15 patients by Prabhat et al.³³ drew the importance of combined approach (ACDF plus posterior partial facetectomy and LMF) following an unsuccessful closed reduction via skull traction. In cases complicated by extrusion of disc, APA technique was suggested otherwise PAP approach was a safer option. Patients with successful closed reduction underwent ACDF alone which was less cumbersome. One disadvantage of combined techniques is a higher rate of dysphagia as compared to only anterior or posterior approaches.

Ding et al.³⁴ included 17 patients in their study of which 5 underwent closed reduction preoperatively. None of the reduction procedures were successful and ultimately all patients underwent surgery via AP approach: anterior discectomy for the removal of fusion mass and the anterior two third of the disc space was filled with morselized graft (from the iliac crest) without any implant. Posterior reduction and lateral mass fixation. We performed the AP approach as well however, there was no filling of disc space with the graft.

Bechet et al., reported an uncommon presentation of a symptomless case of complete cervical dislocation for 1 year who was successfully treated with anterior

discectomy and stabilization with a cage filled with bone allograft in the intersomatic space as well as anterior plating.³⁵ Ultimately, the current literature pointed out to this fact that the surgeon should select one of these wide range of approaches based on the patient characteristics. It means that the APA approach may be very helpful in one patient while it is not appropriate to another one. Therefore, surgeon's judgment will play an important role in treating these complex cases.

There are a couple of limitations to our study. The study size is very limited and hence, larger patient population trials are needed to draw a more meaningful conclusion. Also, we must design some powerful studies regarding this topic to shed light on this way and find the best way for treating these neglected patients.

6 | CONCLUSION

Our study emphasizes the importance of AP approach in patients with irreducible joint dislocations. The approach can minimize the surgical risks and increase the cost-benefit as compared to three or more staged approaches. Our approach is less intensive than some other AP approaches while is a safe and efficacious procedure since the posterior reduction is not performed before discectomy and decompression. In this approach, the surgeon can decompress the cervical cord as well as returning the normal cervical curvature as much as possible without significant restriction following anterior cervical fixation.

AUTHOR CONTRIBUTIONS

Ahmad Pour-Rashidi: Conceptualization; supervision; writing – review and editing. **Bhavya Pahwa:** Writing – original draft. **Mohammad Hossein Khanmirzaie:** Data curation; writing – original draft. **Mahshid Fallahpour:** Data curation; writing – review and editing. **Hamed Hanif:** Data curation; resources. **Mohammad Shirani:** Investigation; resources.

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CONFLICT OF INTEREST STATEMENT

Authors have no conflict of interest to declare.

DATA AVAILABILITY STATEMENT

Anonymized data not published within this article will be made available by request from any qualified investigator.

Investigators interested in working with the data should contact the corresponding author.

ETHICS STATEMENT

Approval was obtained from the ethics committee of Sina Hospital and the Tehran University of Medical Sciences ethics committee.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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