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Case report

Neglected cervical spondyloptosis of the $5^{TH} - 6^{TH}$ cervical spine following cervical manipulation: A case report

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ABSTRACT

A 16-year-old boy was diagnosed with spondyloptosis of the cervical spine at the C5–6 level with a neurologic deficit following cervical manipulation. He could not move his upper and lower extremities, but the sensory and autonomic function was spared. The pre-operative American Spinal Cord Injury Association (ASIA) Score was B with SF-36 being 25%, and Karnofsky's score was 40%. The patient was disabled and required special care and assistance. We performed anterior decompression, cervical corpectomy at the level of C6 and lower part of C5, deformity correction, cage insertion, bone grafting, and stabilization with an anterior cervical plate. The patient's objective functional score had increased after six months follow up and assessed objectively with the ASIA Impairment Scale (AIS) E or Excellent, SF-36 score 94%, and Karnofsky score was 90%. The patient could carry on regular activity with minor signs or symptoms of the disease. This case report highlights severe complications following cervical manipulation, a summary of the clinical presentation, surgical treatment choices, and a review of the relevant literature. In addition, the sequential improvement of the patient's functional outcome after surgical correction will be discussed.

1. Introduction

Spinal manipulation is widely used to treat different conditions and symptoms to relieve neck or back pain and increase range of motion. In Indonesia, traditional healers, including traditional spinal manipulation and massage, are still regarded as the standard option for spinal problems. There are controversies concerning the benefit of such treatment. Some regard it as a beneficial and cost-effective therapy compared to other conservative modalities. However, many disagree [1,2]. One systematic review concluded that spinal manipulative therapy has no statistically or clinically significant advantage in treating spinal pain [2]. Another factor to consider is the possibility of severe complications following spinal manipulation therapy. The sudden thrust mechanism applied when manipulating the spine generally cannot be resisted by the patients [3]. Complications after spinal manipulation of the cervical spine varied from mild to severe. Reported mild complications including transient pain, increased musculoskeletal pain or discomfort, stiffness, headache, dizziness, tingling, or numbness in the upper limb. Other complications include spinal cord injury, nerve root lesion, disc herniation, ruptured discs, cervical fracture, epidural hematoma [13], diaphragmatic paralysis resulting from C3–5 injury, quadriplegia, spondylolisthesis, and even death [3,4,14].

Spondyloptosis is a term to denote grade V spondylolisthesis - a vertebra having slipped so far with respect to the vertebra below that the two endplates are no longer congruent. It is usually seen in lower lumbar spine but can be seen elsewhere rarely [5]. Spondyloptosis is most commonly caursed by trauma and is defined as the dislocation of the spinal column in which the spondyloptotic vertebral body is either anteriorly or posteriorly displaced (>100%) on the adjacent vertebral

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Fig. 1. Clinical presentation of the patient at the initial examination.

Table 1Pre-operative ASIA SCORE.



body. Only a few occurrences of cervical spondyloptosis have been recorded in the literature compared to lumbar spondyloptosis. The cervical cord injury in most patients is complete and irreversible. In most cases of cervical spondyloptosis, regardless of whether there is a neurologic deficit or not, reduction and stabilization of the fracture-dislocation is the management of choice [6,7].

This case report highlights one stage-anterior only surgical management of cervical spondyloptosis following cervical manipulation with sequential follow-up after the procedure, showing significant clinical improvement. This case report has been reported in line with the SCARE Criteria [8].

2. Case report

2.1. Clinical presentation

This 16-year-old boy was consulted by a neurologist to our outpatient clinic with chief complaints of weakness on both upper and lower

Table 2

ASIA Score improvement within six months.



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Fig. 2. Pre-operative plain radiograph of cervical spine demonstrating 5th cervical vertebra was displaced anteriorly toward 6th cervical vertebra.

extremities for four months. Five months before, he had a history of falling while walking on a slippery floor with the back of his head hitting the ground first. After the incident, the patient could walk and perform daily activity normally, but he had frequent neck pain. One month later, because the pain did not subside, he went to a traditional massage and

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performed cervical manipulation. Two days following the cervical manipulation, the patient noticed weakness in his upper and lower extremities and had difficulty standing or walking. His parents initially brought him to a nearby hospital and got oral medication. After two months of therapy, the weakness was not improved, so the patient was



Fig. 3. Pre-operative cervical CT Scan.

referred to our hospital. The neurologist performed an x-ray and MRI examination and then referred to the Orthopaedic spine clinic for further management.

On examination, the patient was wheelchair-bound without any bruises or wound in the cervical region (Fig. 1). There was no deformity on the inspection, with loss of cervical lordotic seen from the lateral aspect. The atrophic appearance of the cervical musculature was also noticed. The patient's cervical spine was not freely mobile. There was a step off at the level of 5th – 6th cervical spine. We also found large café-

au-lait spots on his skin extending from the neck until the proximal part of the posterior thoracic region. From the neurological examination, all modalities for sensations were preserved, but we found that the patient had decreased motoric function from the upper and lower extremity. Muscle strength of both upper and lower extremity were only 1 with positive pathologic reflex and clonus. The patient pre-operative ASIA Score was B (Tables 1 and 2).

Cervical spine plain radiograph showed displacement of C-5 vertebrae anteriorly toward C-6 vertebra, with the body of C-5 vertebra in front of the C-7 vertebra and flattening of C-5 and C-6 vertebrae corpus (Fig. 2). CT Image showed spinal cord compression at the C5–6 level (Fig. 3).

After an overall assessment of the patient, surgical intervention was planned. Cervical traction was not applied due to the neglected nature of the case and to avoid further compression of the spinal cord. The single stage-anterior only procedure was planned, with the application of rigid cervical brace and CT-scan evaluation for five months after surgery to evaluate bony fusion. The patient was operated in supine position under general anesthesia. The cervical spine was exposed by a standard leftsided transversal anterior approach. The incision was performed transversally 5–7 cm at the level of vertebral pathology and confirmed with intraoperative fluoroscopy at the 6th cervical spine. After identifying the 5th - 7th cervical body, Caspar pins were put on the upper part of the 5th cervical body and the 7th cervical body. Distraction of Caspar pin was conducted under intraoperative monitoring not to worsen the baseline signal. Maximal distraction was achieved with residual dislocation. Complete corpectomy was performed on the 6th cervical body and partial corpectomy on the lower part of the 5th cervical body. The decompression was performed until the anterior dura mater was evaluated clear from compression, 10 mm diameter of titanium mesh, and bone graft was applied subsequently. Fluoroscopy evaluation was taken to ensure stability, followed by applying 8-hole anterior cervical plate bridging C5-T1. The bleeding control was taken before the drain was installed. The incision was closed in layers, and the operation was done. Post-operative x-ray was performed, as seen in Fig. 4.

The patient was sent to the ICU to stabilize and improve his general condition. Four days postoperatively, the patient began to show improvement in his neurological status. We can objectively see using ASIA Impairment Scale (AIS) C with Karnofsky score of 50%. He needs assistance and frequent medical care. Note that the patient's motoric function was increased significantly, the SF-36 score was 33% (Table 3).



Fig. 4. Post-operative plain radiograph of the cervical spine showed that 5th cervical – 1st thoracal vertebra was stabilized with an anterior cervical plate, and the spine cage.

Table 3

ASIA score, functional outcome, and Karnofsky scale improvement within six months.

Follow up	Pre OP	Post OP	1 M	2 m	3 m	4 m	5 m	6 m
ASIA SCORE								
AIS	В	С	D	D	Е	Е	Е	E
Autonomic	Good	Good	Good	Good	Good	Good	Good	Good
SF36 (%)								
Physical functioning	0	0	15	50	60	70	90	94
Role limitations due to physical health	0	0	20	30	40	60	80	88
Role limitations due to emotional problems	50	60	70	70	80	80	90	92
Energy/fatigue	25	35	35	50	60	70	84	86
Emotional well-being	52	64	67	72	84	86	92	92
Social functioning	12.5	12.5	12.5	34	57	78	82	90
Pain	55	55	55	75	80	86	89	98
General health	20	25	25	48	67	79	90	94
Health change	0	0	5	37	65	83	90	96
Karnofsky performance status scale								
Value	40	50	60	70	80	80	90	90



Fig. 5. Comparison of patient functional status before surgery (left image) and five months after surgery (right image, the patient was able to ride a bicycle).

He slightly regained his ability to move his upper extremity. This neurological improvement progressed steadily until he regained functional muscle strength (4 to 5) in both his upper and lower extremities by the end of the sixth operative week. Two months after surgery, the patient could sit and stand by himself. Three months after surgery, the patient could walk independently with minimal support. Five months after surgery, the patient could ride his bicycle (Fig. 5). The sensations and sphincter control that were unaffected preoperatively remained unaltered.



Fig. 6. Sagittal CT Scan 5 months after the procedure, showing bony fusion of the C6 body.

CT scan evaluation five months after the procedure showed bony fusion at the lesion site (Fig. 6). The abnormality of the cervical spine shape and alignment and café au lait finding on the patient's skin might indicate predisposing congenital etiology. The abnormal spinal anatomy and contour resulted in the unaligned placement of cervical anterior and cage placement, regardless of the patient's excellent function (Figs. 7 and 8).

3. Discussion

Spondyloptosis refers to a spinal dislocation, also known as Grade V spondylolisthesis. This is a condition in which one vertebral body is entirely displaced in the anterior or posterior side of another vertebra. Overall, it occurs more frequently in the lumbar spine than in the cervical spine. The sub axial cervical spine (C7-T1) is the most affected part



Fig. 7. ASIA and Karnofsky improvement within six months.



Fig. 8. Functional outcome improvement within six months.

in the cervical region [9,10]. The etiology of this condition varies, including birth trauma, congenital conditions, corticated defect in pars interarticularis, neoplastic diseases such as neurofibromatosis or aneurysmal bone cyst, as well as vehicle or diving trauma. Spinal cord damage is frequently caused by the displaced apposition of the adjacent vertebral body [11].

We presented a case of a young man suffering from cervical spondyloptosis with bilateral facet dislocation of the 5th - 6th cervical spine, with incomplete spinal cord injury (central cord syndrome) at the level of the 5th cervical spine following cervical manipulation. Overall, 80% of spondyloptosis cases usually result in a neurological deficit. Though injury without neurologic dysfunction may occur, this has rarely been reported in the literature. Generally, in cases where neurological injury does not occur, spontaneous dorsal decompression of the spinal canal allows the cord to move posteriorly after disruption (or fracture) of the posterior neural arch. Since spondyloptosis is uncommon, management has varied on a case-by-case basis [12].

Several studies suggested cervical traction as the initial management of spondyloptosis before surgical intervention [7,9,12]. We believe that uncontrolled cervical traction may cause spinal cord compression because of the retropulsion of disc material behind the spondyloptosis corpus. This, in turn, may cause neurological deterioration [9]. Spondyloptosis is a three-column ligamentous injury, and over-distraction can easily result in further neural damage. Since this was a neglected case, pre-operative reduction of patients using cervical traction was not considered. A rigid cervical collar was applied preoperatively to the patient to further immobilize the cervical segment.

Treatment algorithms proposed by Modi et al. and Dahdaleh et al. recommend an initial trial of closed reduction prior to operative intervention. Modi et al. recommended initial traction in both patient classifications (patients with ASIA A status versus patients with incomplete neurologic deficits or neurologically intact patients). In contrast, Dahdaleh et al. recommended anterior decompression and intraoperative traction if anterior cord compression was present versus bedside traction if no anterior cord compression was present. The decision-making process regarding anterior versus posterior management of a cervical dislocation is challenging and multifactorial. Regardless of the chosen procedure, the ultimate goals are neural decompression, achieving stability, and obtaining anatomic alignment with final solid fusion [7].

Anterior procedure is preferable for treating patients with a large compressive disc herniation or significant burst fracture component. Its versatility in performing direct decompression and removal of compressing mass with anterior support gives better structural and biomechanical management. However, the posterior procedure is the go-to approach to treat patients with multi-segmental fractures or concerns over fracture stability [7]. Combination of anterior and posterior approaches is reserved for comprehensive fracture-dislocations in patients with spinal cord injuries as it provides greater realignment and stabilization [11]. Therefore, most reported cervical spondyloptosis cases were treated with a combined approach or posterior alone approach [9]. In the setting of a long-standing nature's severe degree of displacement, attempting an anatomical reduction was thought to be hazardous. Therefore, it was decided to perform anterior decompression only, cervical corpectomy at the level of C6, deformity correction, cage insertion, bone grafting, and stabilization with a cervical plate to prevent further damage to the spinal cord. This procedure was beneficial in terms of cord release, as evidenced by the neurological recovery. This case highlighted anterior procedure alone could give significant clinical outcome ASIA Impairment Scale (AIS) E or Excellent, SF-36 score 100%, and Karnofsky score was 90% with acceptable stability, alignment, and solid fusion.

4. Conclusion

Cervical spine manipulation might create serious adverse events such as cervical spondyloptosis, moreover if the manipulation was performed in underlying spinal abnormality. The severe degree of displacement and anatomical changes creates challenges for surgeons in managing such cases. This case highlighted that single-stage-anterioronly surgery could be beneficial to the patients. The post-operative follow-up of our case supports the value of a single stage-anterior only technique for managing neglected cervical spondyloptosis with neurological deficit.

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Ethical approval

The study have been approved by ethical committee.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

CRediT authorship contribution statement

Alan Philips Kustianto Putra Resubun, Andi Karsapin Tarsan, Galih Prasetya Sakadewa drafted the manuscript and critically revised the manuscript by Yudha Mathan Sakti for important intellectual content. Zaky Asad Alhaq, Husein Ahmad, Ardicho Irfantian, Sharfan Anzhari, Andrea Kartika facilitated all project-related tasks.

Registration of research studies

None.

Guarantor

Yudha Mathan Sakti

Declaration of competing interest

No potential conflict of interest relevant to this article was reported.

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