International Journal of Surgery Case Reports 90 (2022) 106633

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr

Case report

Occipitocervical fixation: A case report of our techniques and results

Yudha Mathan Sakti [a,*], Zikrina Abyanti Lanodiyu [a], Sonny Wijanarko [b], Zaky Asad Alhaq [b], Galih Prasetya Sakadewa [b], Husein Ahmad [b], Prisilla Desfiandi [b], Rahadyan Magetsari [a]

[a] Staff of Department of Orthopaedics and Traumatology, Dr. Sardjito General Hospital/Faculty of Medicine, Public Health, and Nursing, Universitas Gadjah Mada, Sleman Regency, Yogyakarta Special Region, Indonesia
[b] Resident of Department of Orthopaedics and Traumatology, Dr. Sardjito General Hospital/Faculty of Medicine, Public Health, and Nursing, Universitas Gadjah Mada, Sleman Regency, Yogyakarta Special Region, Indonesia

ARTICLE INFO

Keywords:
Atlanto-occipital joint
Occipitocervical fixation
Craniovertebral junction pathology
Modified Japanese Orthopaedic Assosciation (mJOA) score
Karnofsky score

ABSTRACT

Introduction: Occipitocervical fixation (OCF) can provide good fusion rate to treat various craniovertebral junction (CVJ) pathologies. Biomechanically it gives rigid fixation, good fusion rate, and allows for effective decompression. However, rigid fixation on the mobile occipitocervical junction has shortcomings that affect the post-operative clinical functional outcomes and range of motion. This study aimed to evaluate and elaborate the functional outcomes, range of motions, and radiographic findings in our patients underwent OCF.

Case report: We presented a report of 3 patients underwent posterior decompression procedure followed by occipitocervical fixation. All three patients' clinical outcome was assessed clinically by, Japanese Orthopaedic Association (JOA) score and grading, Karnofsky, range of motion and radiographic cervical alignment evaluation parameters.

Result: All patients have seen improvement (minimal 1 grade in JOA and >30 points of Karnofsky score) in 3 months after the procedure, had a tolerable range of motion limitation, normal range of cervical lordotic and cervical brow vertebral angle (CBVA). Unfortunately, one patient with loss of cranial fixation may be related to history of infection and lack of post-operative wound care.

Conclusion: Our cases conclude that Occipitocervical fixation is a safe technique that provides excellent fusion rate with good functional outcome and tolerable range of motion limitation. Due to its unique anatomy and technically demanding, serial post-operative monitoring evaluation of this procedure is paramount.

1. Introduction

The occipitocervical junction is a complex biomechanical structure that connects the cranium and the upper cervical [1]. Instability of this structure may happen due to congenital, traumatic deformities, infections, tumors, other inflammatory conditions, and following surgical decompression [2,3]. Spinal cord compromise at the level of high cervical spine resulted in chronic pain, cranial nerve dysfunction, paralysis, and even death. These conditions might necessitate a decompression procedure followed by fixation of the occipitocervical junction [3].

Occipitocervical instrumentation is a technically demanding technique but can be successfully completed with close consideration to local anatomy and working knowledge of the different fixation options [4]. The fixation procedure should be performed in a meticulous fashion to avoid injuring essential structures, including the nerve root, the spinal cord, and the vertebral artery [1,2]. Another consideration was the varying thickness of the occipital bone [4]. Hence the unique anatomy and pathological processes of the occipitocervical junction, fixation procedure that may be suitable for use elsewhere in the spine may fail when applied in the occipitocervical junction [4].

Due to advancements in operative procedures and instrumentation techniques, occipitocervical fixation has evolved significantly [4]. Rigid occipital plating with bicortical screws connected via rods to atlantoaxial or subaxial screw fixation has become one of the good alternatives with its remarkable success rate [5,6]. Rigid fixation allows for fusion to occur in order to provide long-term stability of the mobile occipitocervical junction. However, it also has a negative impact on the patients, especially on the cervical range of motions and functional outcomes [6].

This study aimed to evaluate and elaborate the functional outcomes,

* Corresponding author at: Resident of Orthopaedic and Traumatology Division, Department of Surgery, Faculty of Medicine, Universitas Gadjah Mada/Dr. Sardjito Hospital, Jl. Kesehatan No. 1, Yogyakarta 55281, Indonesia.
E-mail address: Orthopaedi426@gmail.com (Y.M. Sakti).

https://doi.org/10.1016/j.ijscr.2021.106633
Received 19 October 2021; Received in revised form 5 November 2021; Accepted 21 November 2021
Available online 25 November 2021

range of motions, and radiographic findings in our patients underwent OCF.

2. Case reports

This was a case report of three patients underwent occipitocervical fixation conducted in the orthopaedic department at Sardjito General Hospital between 2019 and 2020. The subjects were all males at 40, 50, and 57 years of age, respectively. Written informed consent was obtained from the patient for publication of this case report and accompanying images. We evaluate the clinical features and radiological parameters pre-operative, immediate post-operative, and during follow-up examination.

The clinical parameter was measured using Karnofsky and Japanese Orthopaedic Association (JOA) score [7], which assessed pre-operative, immediate post-operative, and follow-up examination. Radiographic parameters measured in this study were cervical lordosis alignment and cervical brow vertebral angle (CBVA) measured in degree. Radiographic evaluation of the patients in the post-operative period was performed in the same manner.

In all patients, adjustment of alignment was made before skin incision. Occipitocervical fication was done with rigid occipital plating with bi-cortical screws (GS medical, Korea) connected to C2 and C3 lateral mass screws (GS medical, Korea) via rods. The posterior arch of C1 was resected in all cases. There was no bone graft placed between the occipital bone and cervical spine lamina. A rigid cervical collar was maintained for three months post-operatively. This case report has been reported in line with the SCARE Criteria [8].

2.1. Case 1

Male, 50 years old, complained of neck pain which became worse in the last two years. He was unable to lie down on his bed because the pain would worsen in the lying position. He had a history of a motorcycle accident ten years before, but no neurological deficit was found afterward. He denied any history of fever, chronic cough, spinal injection, weight loss, seizure, and stroke. There was positive history of diabetes, but he denied any diabetic ulcer.

One month before surgery, he started to feel weak on all his four extremities and getting worse. On physical examination, we found sensory deficit from 1st lumbar level and moderate and severe motor deficit from 5th cervical and 1st lumbar level, respectively. His Karnofsky score was 40, and unable to ambulate. We conducted an x-ray along with routine blood examination and MRI of his spine (Fig. 1).

We then diagnosed the patient with Burst pathologic fracture of the 1st–2nd cervical spine due to suspect specific infection Frankel C, mJOA 12. Then we performed decompression and stabilization, correction of deformity, and open biopsy with spanning occipitocervical junction. We performed decompression in the 1st and 2nd cervical spine, stabilized these levels using an occipital plate, and inserted lateral mass screw 3.5 mm to the 3rd and 4th cervical spine. We also performed deformity correction and maintain the cervical lordosis from 17^0 to 31^0 and coronal alignment from 7^0 to 2^0, pre-and post-operative, respectively. The CBVA was 11° preoperatively and 5° Postoperatively (Fig. 2).

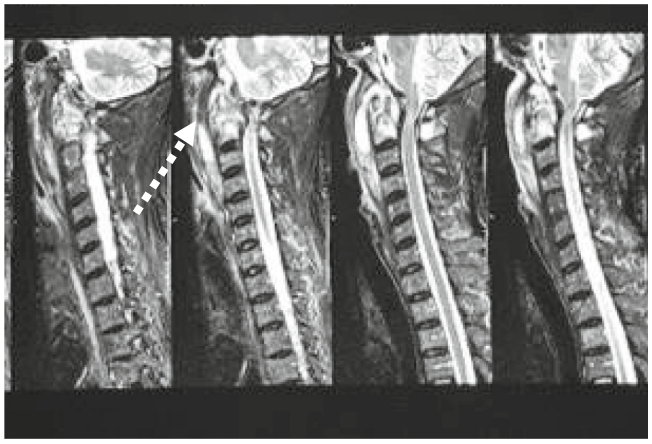

Fig. 1. Initial MRI examination showing the hyperintense structure at 1st–2nd cervical spine level.

The patient came to our clinic for 1-year post-operative follows up. He was able to stand and walk normally, and we found no other neurological deficit. The patient has been in a pain-free state for 1 year. Karnofsky scores assessments were increased significantly from 40 to 90, and patient subjectively satisfies with the result. Cervical lordotic angles were preserved at functional degree (Fig. 3). The cervical flexion was measured at 26° and extension at 10°, right lateral flexion was 9° and left lateral flexion were 10° clockwise cervical rotation was 19° and counter-clockwise cervical rotation was 15°.

2.2. Case 2

Male, 40 years old with history of pain in the cervical region with a tingling sensation in the right arm for one month. He denied any history of trauma, fever, chronic cough, or weight loss before the incident. He consulted to the neurologist and got an MRI examination of his cervical spine (Fig. 4). The result was suspicious of cervical mass, and the patient was referred to a hemato-oncology specialist. Three days before being admitted to Sardjito General Hospital, Yogyakarta, Indonesia. He was unable to wake up from his bed. Right hemiparesis was found in the initial physical examination of this patient, with clonus and pathological reflex appeared on the right lower extremity. There were no autonomic disabilities, including urinating and defecating problems at the initial encounter.

The patient underwent further examination to study the primary source of the tumor. Laboratory examinations (CEA, PSA, CA 19-9 levels) were normal, as well as head and thorax CT scan. MRI studies showed irregular structure over the 2nd–5th cervical spine. The patient was then diagnosed with adult pyogenic vertebral osteomyelitis of the 2nd-5th cervical spine Frankel B mJOA 9. Occipitocervical fixation was then planned for the patient.

We performed a laminectomy, decompression, stabilization, deformity correction, and open biopsy on this patient. Debridement was performed as intra-operative finding showed seropurulent discharge instead of mass/tumor-like structure. The discharge was sent to the clinical pathology department for culture examination, and *Enterococcus faecalis* was found later. Decompression was performed at the level of 3rd–4th cervical spine, and fixation was done at the C4-C6 levels.

The cervical lordosis was found 17° (pre-operative) and 10° (post-operative) and the coronal alignment was found 10° (pre-operative) and 5° (post-operative). The CBVA was -4° (−1 to −5°) preoperatively and it had better result post-operatively) 5° (−1 to −5°). The patient was under intensive care for five days after the procedure (Fig. 5). The patient got a urinary tract infection in the intensive care unit with a bacteria count of his routine urinary examination exceeding 1624.1/μL (normal: 0–100 μL). Collar neck stabilization was applied for one month. Motoric level at his right extremities increased 2 levels above the pre-operative motoric score. Post-operative Karnofsky score was increased from 30 to 50. He was able to sit on the bed and move his right leg. Along with strict rehabilitation protocol, the patient was able to stand with assistance 1 month after surgery.

The patient came back to our outpatient department (Sardjito General Hospital, Yogyakarta, Indonesia) three months after with a history of loss of follow up. He was able to walk with a walking aid and experienced no neck pain. However, he started to complain of exposed metal on his occipital with the wound (Fig. 6). The 6th-month cervical flexion was measured at 6° and extension at 5°, right lateral flexion was 7° and left lateral flexion were 5° clockwise cervical rotation was 7° and

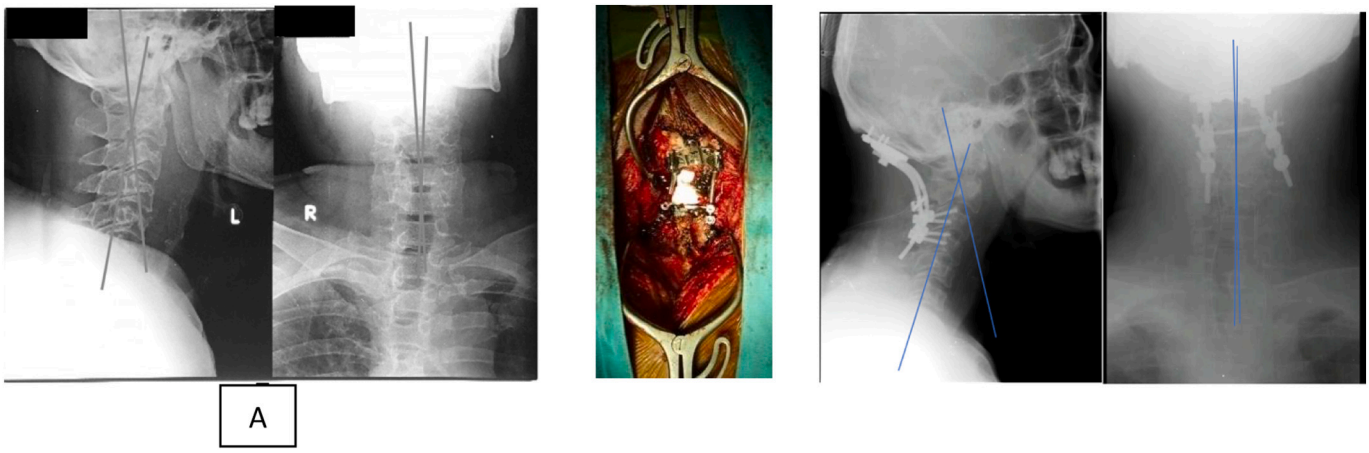

Fig. 2. X-ray images showing pre-operative loss of cervical lordosis and coronal deformity (a) were fixed on post-operative x-ray images (b) intraoperative image of screw placement on cervical spine (c).

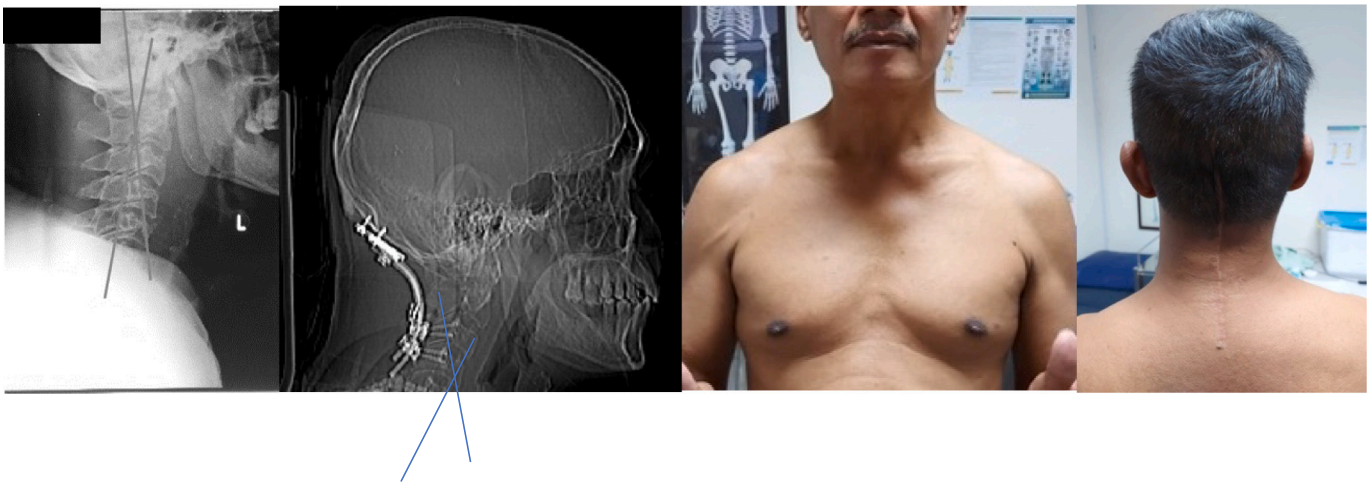

Fig. 3. 1 year follow-up X-ray images and clinical evaluation.

counter-clockwise cervical rotation was 5°.

2.3. Case 3

Male, 57 years old with the chief complaint of weakness of his right upper and lower extremities. Seven months before being admitted to our hospital, the patient complained of numbness and constant tingling sensations throughout his right leg. These complaints worsen over time until the whole right leg is affected. He also complained of weakness of the same arm ever since. These symptoms are felt constantly throughout the day and made it hard for him to work and ambulate. He had a history of trauma on his neck five years ago when he was involved in a motorcycle accident and had a direct impact on the back of his neck, but there were no complaints throughout that time. No history of weight loss, infection, nor significant injury. He denied any other complaints on other body parts, no trouble urinating nor defecating. Physical examination results were within the normal limit. He was able to walk with the help of walking aids. His neurological status showed right sensory deficit from the level of C3 and right motoric deficit from the level of C4. We performed an MRI examination, as seen in Fig. 7. There was normal physiological reflex and no pathological reflex.

We diagnosed the patient with Cervical spondylotic myelopathy of the 1st-4th cervical spine Nurick grade 2 mJOA 13 with a neurologic deficit and performed Laminectomy, decompression, and Occipitocervical fixation. The cervical lordosis was found 21.5° (pre-operative) and 30° (post-operative), and the coronal alignment was found 10° (pre-operative) and 5° (post-operative). The CBVA was −4° (−1 to −5°) preoperatively and 5° (−1 to −5°) Postoperatively (Fig. 8). The patient was discharged 1 week later. The patient was able to walk without a walking aid at 1-week outpatient follow-up. The motor and sensory deficit were improving as well as pain and tingling sensation. The patient's cervical range of motion was also limited at a tolerable range of motion (Fig. 9). The cervical flexion was measured at 33°, extension was 5°, both right and left lateral flexion was 8° clockwise, cervical rotation was 17° and counter-clockwise cervical rotation was 13°. The range of motion of the neck was sufficient to perform daily activity with no disturbance of our patient's gaze and vision.

All of our patients showed good outcome after the procedure based on JOA score with at least one grade of improvement, as seen on Table 1.

3. Discussion

Occipitocervical junction or also known as craniovertebral junction consists of the occiput, os atlas, and os axis [1,4]. The distinct morphology of these bones, aided by the presence of various soft tissues, including ligaments, provides stability while allowing it to retain its wide range of motion [4].

Certain conditions such as trauma, rheumatoid arthritis, neoplastic

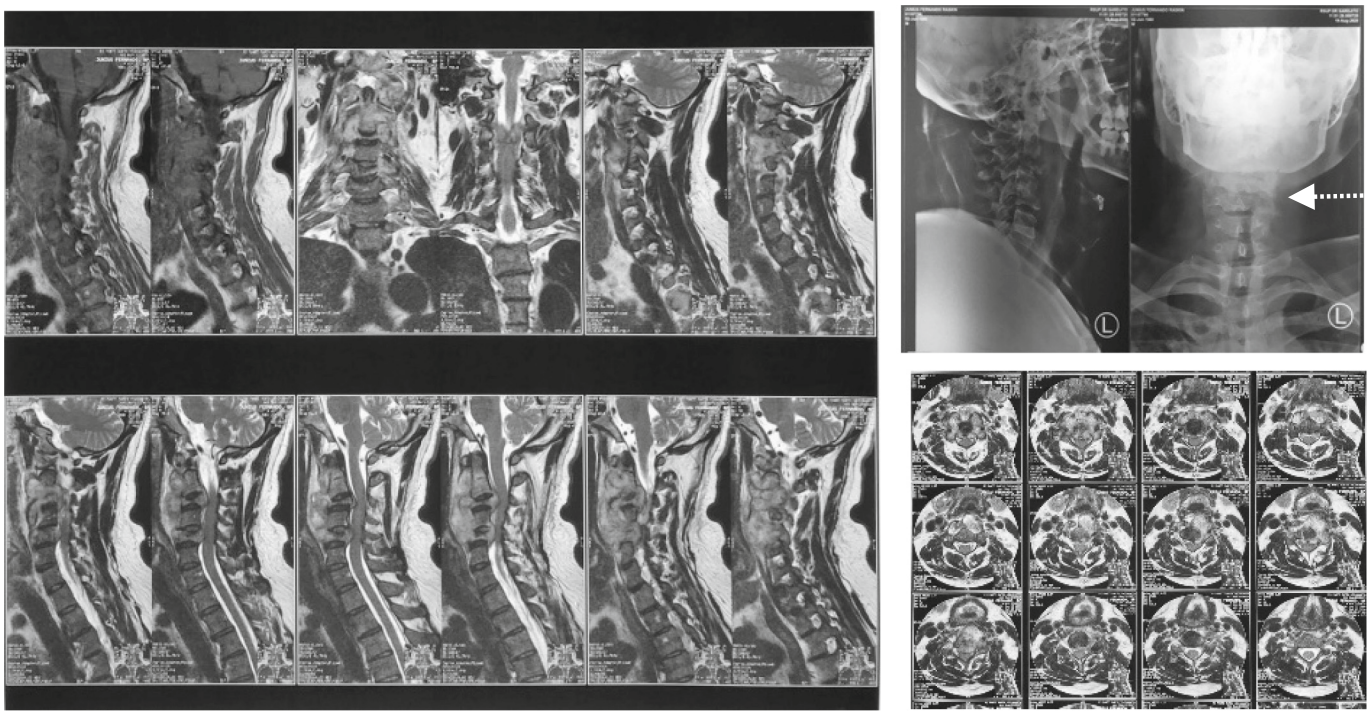

Fig. 4. MRI sagittal T2 showing grey irregular structure over the 2nd–5th cervical spine (left) while bony deformity was also found on AP/lateral cervical spine X-ray (right upper). Axial images showed the extend of the structure (right lower).

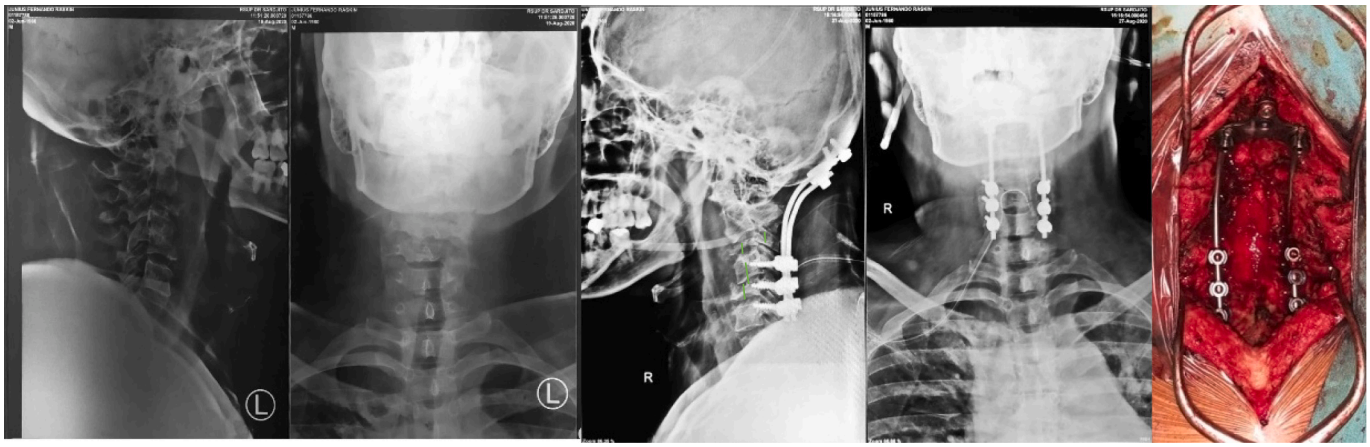

Fig. 5. Pre and post-operative cervical parameter (left & middle) Intraoperative image (right).

disease, infection, congenital anomalies, degeneration, and post decompression may cause instability to the occipitocervical junction and compromise the adjunct spinal cord [1,2]. Two of our cases (Case 1 and Case 3) represented degenerative processes, while the other one was tissue destruction caused by infection. Various etiologic processes have been reported by the previous study as the cause of occipitocervical junction compromise [2,3]. In their study, Martinez-del-Campo et al. reported that out of 120 patients with occipitocervical junction instability, 56 had trauma etiology, followed by 24 cases of congenital anomalies, 13 from rheumatoid arthritis, nine from cervical stenosis with instability, seven from neoplasms, six from hardware failure, three from infections and the remaining two from multiple causes or vascular malformation [1]. Occipitocervical fixation, as an effort to stabilize craniocervical junction, has certain goals, including regaining normal alignment, ensuring adequate neural tissue decompression, and achieving structural stability [2]. Studies from the literature suggested that this particular technique is challenging [1–4].

The patient's position was prone using a fixed head holder to maintain the neck in a slightly flexed position to open the interspinous spaces. The patient's head was supported by a gel frame to avoid any direct pressure to the eye. In this case report, we made the adjustment of cervical alignment before skin incision. The shoulders were abducted at less than 90°, and the forearm was in a neutral position to avoid direct pressure on the ulnar nerve. We placed soft foam padding under the elbows and pillow under the abdomen to avoid pressure on the vena cava. Intra-operative fluoroscopy was prepared, and the patient's arm were strapped down along the body to maximize lateral fluoroscopy of the cervical spine.

In our case series, all patients present with neurologic deficits. One presents with weakness of all extremities. The other two patients presented with tingling sensation and numbness, which were then followed by motor weakness. All three patients did not have a complaint on urination and/or defecation function. Neck pain has always been regarded as the chief complaint in occipitocervical instability. In the study by

Fig. 6. 4 months follow-up, complication, and post six-month ROM evaluation.

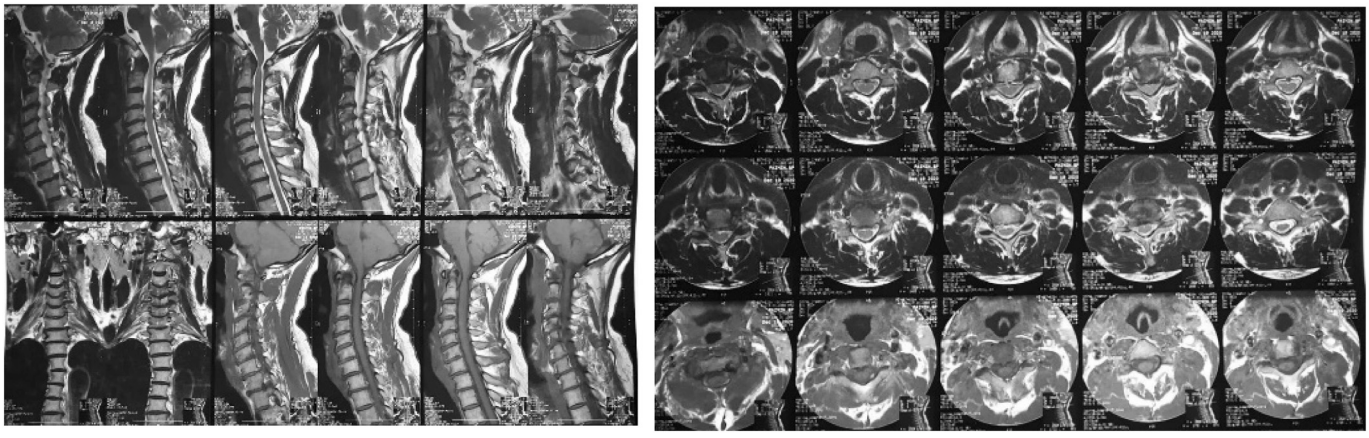

Fig. 7. Pre-operative MRI showed degenerative condition which compromised the spinal canal.

Zidan et al. [3] all ten patients have a complaint of neck pain, while Martinez-del-Campo et al. [1] reported that 83 of 120 patients (69%) presented with neck pain. In this case series, all three patients had a history of neck pain.

Occipitocervical fixation has long been hailed as one of the suitable treatments to achieve biomechanical stabilization and bone fusion, adequate neural tissue decompression, and to avoid injuring the nerve root, the spinal cord, and the vertebral artery [3]. Since its initial

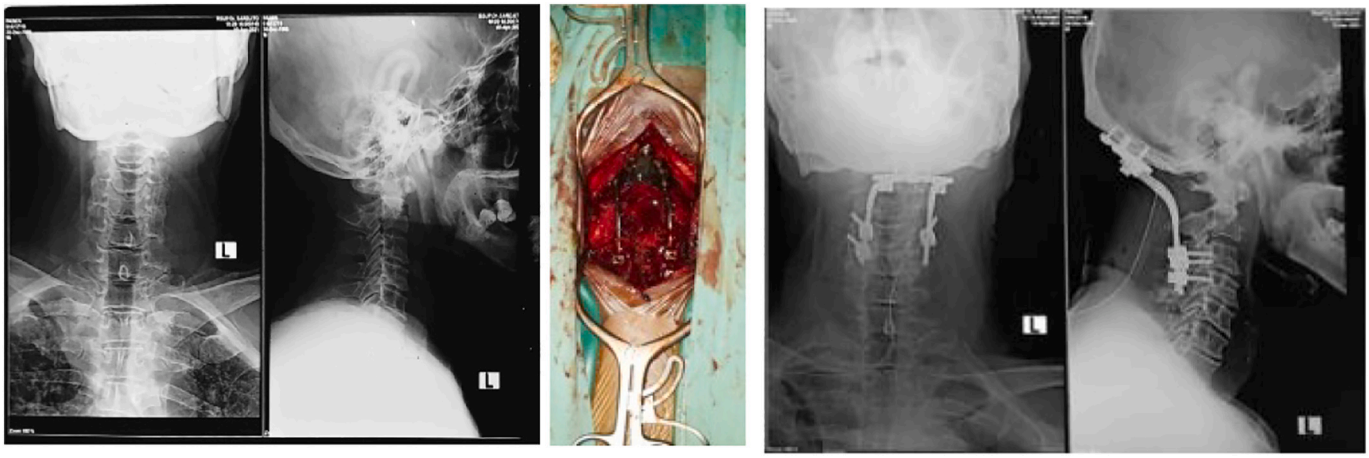

Fig. 8. Pre-operative, intraoperative, and post-operative X-ray showing placement of the fixation.

Fig. 9. Post-operative range of motion of case 3.

Table 1
JOA score before and after occipitocervical fixation.

	Pre-operative	Post-operative
Case 1	11 (grade 2)	17 (normal)
Case 2	8 (grade 2)	12 (grade 1)
Case 3	10 (grade 2)	15 (grade 1)

conception reported by Foerster in 1927 using nasal bone graft, Occipitocervical fixation has evolved [3].

The fusion rate for most methods for Occipitocervical fixation is more than 90% within four months. In the inflammatory disease group, the greatest improvement in neurological status was when screw/rods were used, whereas the tumor disease group was more suited with posterior wiring/rods. In the trauma group, best pain improvement followed screw/plate construct [4,5]. Due to their different usage and advantages, there has not been a method regarded as the best. The chosen method for Occipitocervical fixation and the method of surgery should be based on patient conditions, such as type of instability, the integrity of posterior cervical elements, the extension of decompression needed, comorbidities, individual anatomic variation, and also the surgeons' familiarity with the techniques [2].

Occipitocervical fixation has been shown to improve the clinical state of patients. Zidan et al. [3] reported that there is complete resolution of neck pain in all cases. At the last follow-up, five patients had improved in Frankel grade (three from Frankel grade C to grade D, one from Frankel grade A to B, and the other one patient from Frankel D to E), while the other five patients remained in the same Frankel grade. A similar result was also reported by Martinez-del-Campo et al. [1]. 95% (55 of 58) of nontraumatic patients have neurological improvement with 36% regained complete neurological function and 57% recovered >3 mJOAS categorical points. In trauma patients, 64% (14 of 22) patients had neurological improvement, and 77% (41 of 53) patients regained complete neurological function. Trauma patients have a significantly better improvement than non-trauma as a higher percentage of these patients were neurologically intact before the trauma.

Occipitocervical fixation is not without its risk. Due to its unique characteristics and proximity to many vital structures, it is associated with various complications [4]. Reported complications such as vertebral artery injury, dural tear, cerebrospinal fluid (CSF) leakage, wound infection, nerve or cord injury, screw failure, and bone fusion failure [2,6]. The overall complication rate of these procedures varied from 10% [1] to 52% [4]. In our case report, the only complication met was plate exposure in the second case due to history of infection. The possible cause of this complication might be from lack of wound care due to the patient presenting one month after the surgery. More wound care supervision, such as twice a week, may be beneficial to prevent plate exposure in patients undergoing this procedure.

4. Conclusion

From our cases, we conclude that Occipitocervical fixation is a safe technique that has an excellent fusion rate with good functional outcomes and tolerable range of motion limitation. Due to its unique anatomy and technically demanding, serial post-operative monitoring evaluation of this procedure is paramount.

Declaration of competing interest

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

Acknowledgements

We thank patient family, the surgical team and the nursing staff who were involved in the surgery and patients care.

Not commissioned, externally peer-reviewed.

Sources of funding

The authors declare that this study had no funding resource.

Ethical approval

The study has 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.

Author contribution

Yudha Mathan Sakti, Zikrina Abyanti Lanodiyu, Sonny Wijanarko, Zaky Asad Alhaq, Rahadyan Magetsari drafted the manuscript and critically revised the manuscript by Meirizal for important intellectual content. Galih Prasetya Sakadewa, Husein Ahmad, Prisilla Desfiandi facilitated all project-related tasks.

Registration of research studies

–.

Guarantor

Yudha Mathan Sakti.

References

[1] E. Martinez-Del-Campo, J.D. Turner, S. Kalb, L. Rangel-Castilla, L. Perez-Orribo, H. Soriano-Baron, Occipitocervical fixation: a single surgeon's experience with 120 patients, Neurosurgery 79 (4) (2016) 549–560.

[2] S.H. Choi, S.G. Lee, C.W. Park, W.K. Kim, C.J. Yoo, S. Son, Surgical outcomes and complications after occipito-cervical fusion using the screw-rod system in craniocervical instability, J. Korean Neurosurg. Soc. 53 (4) (2013) 223–227.

[3] I. Zidan, W. Fouad, Occipitocervical fixation in the management of craniocervical instabilities, Alex. J. Med. 47 (3) (2011) 185–192.

[4] C.D. Winegar, J.P. Lawrence, B.C. Friel, C. Fernandez, J. Hong, M. Maltenfort, et al., A systematic review of occipital cervical fusion: techniques and outcomes: a review, J. Neurosurg. Spine 13 (1) (2010) 5–16.

[5] C.D. Winegar, J.A. Rihn, J. Hong, C. Fernandez, B. Friel, A.R. Vaccaro, Cervical spine: contemporary posterior occipitocervical fixation techniques, Curr. Orthop. Pract. (2008) 19.

[6] R. Lall, N.J. Patel, D.K. Resnick, A review of complications associated with craniocervical fusion surgery, Neurosurgery 67 (5) (2010) 1396–1402.

[7] A.F. Joaquim, J.A. Osorio, K.D. Riew, Occipitocervical fixation: general considerations and surgical technique, Glob. Spine J. 10 (5) (2020) 647–656.

[8] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.