Management of neglected complex hangman's fracture by reforming the C2 pedicle: new innovative technique of motion preservation at the C1–2 joint in 2 cases

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Hangman's fracture, also known as traumatic spondylolisthesis of the axis, causes widening of the neural canal and thus a low rate of neurological deficits. This low rate is one of the reasons it is neglected and patients present with late neurological deficits. In an effort to preserve motion at the C1–2 joint, the authors devised a new technique of bilateral C2 pedicle reconstruction. They describe the first two cases in the literature of an old hangman's fracture with resorbed C2 pedicles due to chronic fracture, in which bilateral C2 pedicles were reconstructed. One of the two cases (case 2) is the first reported case of severe C2–3 spondyloptosis with C2 displaced up to the level of C4. Case 1 had a follow up of 21 months, while case 2 had a follow up of 12 months. Both patients experienced neurological improvement with evidence of fusion and artificial pedicle formation at last follow-up. Bilateral C2 pedicle reconstruction is a feasible technique that can be used with a good outcome in select patients.

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HANGMAN's fracture, also known as traumatic spondylolisthesis of the axis, is a well-known entity.⁹ It accounts for approximately 5% of all cervical spine fractures,⁷ and results in the separation of the vertebral body and neural arch, therefore causing widening of the neural canal. Patients with hangman's fracture usually show a low rate of neurological deficits; this might be one of the reasons for the fracture being neglected and later detected upon the persistence of chronic neck pain or development of late neurological deficits.^{4,10} There are some case reports available outlining their diagnosis and management.^{8,11,12} To the best of our knowledge, we describe the first two cases in the literature in which bilateral C2 pedicles were reconstructed in old hangman's fractures in which the C2 pedicle was absent due to chronic fracture and reabsorption. One of the two cases (case 2) is the first

reported case of severe C2–3 spondyloptosis with C2 displaced up to the level of C4.

Methods

Two patients who were relatively neurologically preserved presented to our institute. Imaging revealed a complex neglected hangman's fracture with bilateral resorbed C2 pedicles. Conventional treatment would involve fixation, which would have included C1 in the construct. We conceptualized placement of bilateral C2 screws directly into the C2 vertebral body, passing through the region of the pedicle.

The desired outcome was bilateral C2 pedicle reconstruction with adequate reduction and stable fixation, along with preservation of motion at the C1–2 joint. The manage-

ABBREVIATIONS APS = artificial pedicle screw; ASIA = American Spinal Injury Association; CTA = CT angiography; LMS = lateral mass screw; MRC = Medical Research Council; VA = vertebral artery.

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ment of the two patients, including the surgical technique and outcome, is described below. Patient consent was obtained for the use of clinical and radiographic images, and patient details are not identifiable from the text or images. Ethics committee approval was not sought as this paper describes the novel management of severe spondylolisthesis of the axis with commonly used neurosurgical operative techniques.

Results

Case 1

A 14-year-old boy presented to us with a history of fighting with his classmates at school 6 months previously, during which he was shaken around and fell to the ground. He did not inform anyone at the time. He had experienced neck pain since the time of the injury, and started developing progressive weakness in all four limbs, along with paresthesia and numbness below the neck level after a few months. It was only at that time that his parents started seeking medical attention. Upon presentation to our hospital he had limitation in cervical spine movements in all directions. His grade on the American Spinal Injury Association (ASIA) impairment scale on examination was grade D. The patient had spasticity and exaggerated deep tendon reflexes in all 4 extremities, with 4/5 power on the Medical Research Council (MRC) scale across all limb joints. The plantar response was extensor. He was found to have a graded sensory loss to all modalities below the C3 dermatome. Plain radiographs showed an unstable C2–3 spondylolisthesis (Fig. 1A and B). A CT scan showed a grade 3 anterolisthesis of C2 over C3 with anterior translation, severe angular deviation, and bilateral par interarticularis defect. There was reduced disc space at the C2–3 level with a posterior retropulsed C3 vertebra causing severe narrowing of the bony spinal canal (anteroposterior diameter = 5.4 mm). The bilateral C2 pedicles had resorbed. There was loss of cervical lordosis with focal kyphosis at the C2–3 level. CT angiography (CTA) showed bilateral normal vertebral arteries (VAs) in course and caliber (Fig. 1C and D). MRI showed severe cord compression due to the retropulsed C3 fragment (Fig. 1E). The patient was put in cervical traction with no improvement in listhesis.

The patient underwent surgery via a posterior approach under intraoperative CT navigation. Posterior elements of C1–5 were exposed by subperiosteal dissection of muscles. The segment of the VA between the C3 and C1 vertebrae was carefully delineated bilaterally using sharp dissection. Bilateral C2 nerve roots were identified and retracted to expose the C2 body. The point where the absent pedicle would have been attached to the C2 body was chosen as the site of insertion of a body screw. This was further reconfirmed using intraoperative CT. An entry hole was drilled and polyaxial screws were placed bilaterally directly into the body, with the same trajectory as used for normal C2 pedicle screws. The screws passed through the empty space in place of the resorbed pedicles and care was taken to ensure bicortical purchase. Bilateral C3 and C4 pedicle screws were placed under imaging guidance. Reduction of C2 over the C3 vertebral body was achieved. Bone chips from the iliac crest were placed around the

C2 pedicle screws from the posterior surface of the C2 body to the C2 facets, to enable pedicle reformation and posterolaterally around the screw-rod construct to allow for fusion (Fig. 1F–H).

Postoperatively, the patient experienced no new neurological deficits. The patient was discharged with hard cervical collar application advised for 12 weeks. At 3 months' follow-up the patient had improved power in all 4 limbs with reduced spasticity. CT performed at 21 months' follow-up showed reformation of the artificial C2 pedicle around the screw with evidence of posterior fusion (Fig. 2A–D). His current grade on the ASIA impairment scale is grade E, and he is doing well with preserved motion at the C1–2 joint (Fig. 2E and F).

Case 2

A 32-year-old man presented with a history of a road traffic accident, in which he was a pedestrian struck by a four-wheeler. At the time of the accident he was treated for a right temporomandibular joint dislocation with blunt chest trauma, along with fracture of the left tibia and fibula. The patient also underwent forequarter amputation of the left upper limb. Since the accident he had complained of dull aching neck pain with restriction of neck movements, but was not evaluated for these conditions. A few months later he developed mild weakness of his remaining three limbs, along with paresthesia and numbness below the neck level. He came to our institute 1 year after the accident. Upon examination, he was grade D on the ASIA scale. He had spasticity and exaggerated deep tendon reflexes in his remaining three extremities, with 4/5 power on the MRC scale. The plantar response was extensor. He had a graded sensory loss to all modalities below the C3 dermatome. CT of the cervical spine showed a spondyloptosis of C2 over C3, with the C2 vertebral body resting anterior to the superior portion of the C4 vertebral body. Again, there were no visualized C2 pedicles bilaterally. The posterior retropulsed C3 vertebra was causing severe narrowing of the bony spinal canal. MRI showed kinking of the cervical cord by the C2–3 disc along with the C3 vertebral body. CTA showed a dominant left-sided VA (Fig. 3). The patient was put in cervical traction for 2 weeks, with progressive increases in weight up to 5 kg. As the patient was lean and weighed only 40 kg, a further increase in traction weight was not considered. The patient experienced mild reduction, with the C2 vertebral body moving anterior to the upper part of the C4 body. He then underwent anterior soft-tissue release with partial C2–3 discectomy. There was difficulty in reducing the laterally subluxated C2 body and reaching the upper surface of the C3 body because of dense fibrosis. Thus, the patient was again put in traction for a few days with the C2 body reducing to the mid part of C3. The patient was again taken for surgery, in the hope of achieving complete reduction. Anterior soft-tissue release along with C2–3 discectomy was completed. The patient was turned over and posterior soft-tissue release was performed. He was again put in traction during the postoperative period, with almost complete reduction of the listhesis noted after a few days (Fig. 4A–C).

He then underwent surgery via a posterior approach.

FIG. 1. Case 1. **A and B:** Preoperative plain extension (A) and flexion (B) radiographs of case 1 (arrow denotes the absent C2 pedicle). **C and D:** CTA shows bilateral normal VAs. Inset is an axial scan of the C2 vertebral body showing bilateral absent pedicles (arrow denotes the absent C2 pedicle). **E:** Preoperative sagittal T2-weighted MR image shows severe spinal cord compression due to the retropulsed C3 fragment. **F:** Intraoperative CT image taken after placement of bilateral C2 body screws. **G:** Topogram image of the final construct showing reduction achieved by a posterior-only approach. **H:** Postoperative midsagittal CT image showing adequate canal diameter.

Intraoperative CT and neuronavigation was used. Bilateral C2 body screws were placed through the space available in place of the resorbed pedicles (using the same technique as described in case 1 above), followed by C3 lateral mass and C4 pedicle screws. This was followed by rod fixation. Bone chips from the iliac crest were placed around the C2 pedicle screws from the posterior surface of the C2 body to C2 facets to enable pedicle reformation, and posterolaterally around the screw-rod construct to allow for fusion.

The patient was then turned anteriorly, the previous incision was reopened, and bone harvested from the anterior part of the right iliac crest was inserted into the C2–3 disc space. C2–3 vertebral body plating was performed with 4 screws (Fig. 4D–G). The patient was kept on ventilator support in the postoperative period, while undergoing aggressive chest physiotherapy. He was gradually weaned off from the ventilator and extubated. He was discharged with no fresh neurological deficits.

FIG. 2. Case 1. Follow-up imaging of the patient at 21 months. **A and B:** Parasagittal CT images showing bilateral bone formation around the C2 pedicle screws (*arrow*). **C:** Axial CT image showing stable C2 body screws with good purchase at 21 months. **D:** Axial CT image showing bone formation around the C2 pedicle screws. **E and F:** Clinical images of the patient showing preserved neck motion. Figure is available in color online only.

FIG. 3. Case 2. **A:** Preoperative midsagittal CT scan showing severe C2 spondyloptosis over C3, reaching up to the anterior aspect of the C4 body. **B:** Coronal image showing lateral subluxation. **C and D:** Axial CT images showing the C2 body (*arrow*) with absent bilateral C2 pedicles lying anterior to the C3 body. **E and F:** Reconstructed CTA shows bilateral intact VAs with left-side dominance. **G:** Preoperative sagittal T2-weighted MR image showing severe cord compression due to the retropulsed C3 fragment along with the C2–3 disc. Figure is available in color online only.

FIG. 4. Case 2. **A–C:** Serial lateral radiographs showing partial reduction after preoperative traction (**A**), in which the C2 body has moved anterior to the C3 body; after anterior soft-tissue release and traction (**B**), in which the C2 body has moved midway anterior to the C3 body; and after repeat anterior and posterior release and traction (**C**), in which the C2 body has almost completely reduced to its anatomical position. **D:** Intraoperative CT image showing unilateral C3 LMSs and C4 pedicle screws in place with rod held up under the C1 arch for stability of C2. **E:** Unilateral C2 body screw with bilateral C3 lateral mass and C4 pedicle screws with rod placed on one side. **F:** Screw-rod fixation performed in extension, thus reducing C2 completely over the C3 body. **G:** Final construct showing posterior and anterior fixation.

The patient was advised to wear a hard cervical collar for 12 weeks. At 12 months' follow-up, the patient's ASIA scale grade had improved to grade E. A CT scan performed at 12 months' follow-up showed screws with connecting rods seen fixing C2, C3, and C4 vertebral bodies and pedicles with maintained cervical alignment. There was reformation of the artificial C2 pedicle around the screw with evidence of fusion. The C2–3 disc space was obliterated due to arthrodesis (Fig. 5A–D). This was a 900° approach with motion preserved at the C1–2 joints (Fig. 5E–H).

Discussion

Neglected spinal cord injuries have been reported in the literature, the cause being variously reported as overlooked diagnosis¹⁰ or inadequate awareness among pa-

tients in developing countries, thus emphasizing the great importance of creating awareness in preventing neglect.⁴ Clark et al. reported delayed radiological diagnosis in 10% of hangman's fractures based only on radiographic evaluation.⁶

In both cases described in this paper, there were absent bilateral pedicles due to resorption. Screws were placed directly into the vertebral body, followed by bone chips around the screw, with the aim of bilateral pedicle reconstruction. A starter hole was drilled and tapped under direct visualization followed by insertion of a polyaxial screw into the vertebral body. The screws are placed under direct vision into the vertebral body, while bypassing the VA that lies lateral to the absent pedicle. This is necessary to ensure bicortical purchase of the screw (Fig. 6).

Acosta et al.¹ described unilateral C2 artificial pedicle screws (APSS) in 2005. These investigators iatrogenically

FIG. 5. Case 2. Follow-up imaging of the patient at 12 months. **A:** Midsagittal CT image showing adequate canal diameter. *Inset* of axial CT image shows C2 body screws with good purchase. **B:** Coronal CT image showing reduction of the lateral subluxation. **C and D:** Axial CT images showing bone formation (arrow) around the C2 pedicle screw on the right (C) and left (D) sides, respectively. **E–H:** Clinical images of the patient showing well-preserved neck motion. Figure is available in color online only.

drilled off the cervical pedicle to approach tumors ventral to the spinal cord. This was performed after ipsilateral stabilization using lateral mass/pedicle screw-rod fixation. There was no attempt at pedicular reconstruction with bone chips, as in our cases. The authors later described a series of 14 cases.²

Barnes et al.³ tested the biomechanical strength of the artificial cervical pedicle screw placed at the C4 vertebral body level and compared it to the lateral mass screw (LMS). They observed an increase in pullout strength of APSs versus LMSs, although it was not statistically significant. They concluded that there is no difference in the biomechanical performance of LMS and APS fixation techniques in the cervical spine. No studies exist testing biomechanical properties of the C2 APS.

To the best of our knowledge these are the first cases of bilateral C2 pedicle reformation in the literature in the world. Srivastava et al.¹¹ described a case of neglected hangman's fracture similar in morphology to case 1 in our report. They could not achieve a preoperative reduction using traction, so had to use a 360° approach, in which anterior soft-tissue release was followed by posterior and anterior fixation. In contrast, we could achieve adequate reduction of the listhesis after placement of C2 body screws and were able to avoid an anterior approach.

Choi et al.⁵ described a case of an almost completely dislocated C2–3 listhesis, which was managed via posterior fixation. They could not place C2 pedicle screws due

to the removal of the left C-2 pars interarticularis along with the posterior elements of C2, thus including C1 in the construct. This could have been prevented by using our technique described above, thus preserving motility at the C1–2 joint. We could not find any case matching the severity of the spondyloptosis seen in case 2 of our report, and wish to highlight the management of such complex cases through adequate planning.

We have observed several benefits of the technique of artificial pedicle reconstruction. It has allowed segmental fixation even in cases with an absent pedicle or lateral mass and facet complex, thus preserving motion at the C1–2 joint. It completely avoids injury to the VA, passing directly into the vertebral body while remaining medial to the VA. It is not restricted by the pedicle diameter and a wider diameter screw can be safely placed. It is a biomechanically sound alternative to have in the surgeon's armamentarium.

Conclusions

The management of complex neglected cases of hangman's fractures is complicated. Judicial use of traction and careful planning of the operative steps is necessary to ensure a good outcome. Use of intraoperative CT and neuro-navigation has been indispensable in correct placement of difficult screws. Artificial pedicle reconstruction has been shown to be a feasible screw placement option in selected cases.

FIG. 6. Illustration depicting the technique of C2 body screw placement. **A:** Parasagittal diagram showing direct placement of the screw into the C2 body, superior to the C2 facet. **B:** Parasagittal diagram showing the screw position after reducing the C2 body into anatomical position, with bone chips placed in place of the C2 pedicle around the screw. **C and D:** Axial diagrams showing the space left due to the resorbed C2 pedicle (C), and diagram showing bilateral C2 body screws in position with bone chips in place of the C2 pedicle (D). **E:** Coronal diagram showing the entry point of the C2 body screw, medial to the VA and inferior to the C2 nerve root. SC = spinal cord. Figure is available in color online only.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Singh. Acquisition of data: Singh, Agrawal, Sawarkar. Analysis and interpretation of data: Singh, Agrawal,

Sawarkar. Drafting the article: Singh, Agrawal. Critically revising the article: Singh. Reviewed submitted version of manuscript: Singh, Agrawal, Kumar, Verma, Doddamani. Approved the final version of the manuscript on behalf of all authors: Singh. Administrative/technical/material support: all authors. Study supervision: Chandra, Kale.

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