Ankylosing spondylitis traumatic subaxial cervical fractures – An updated treatment algorithm

ABSTRACT

Ankylosing spondylitis (AS) is a rheumatologic disease characterized by ankylosis and ligament ossification of the spine with an elevated risk of vertebrae fractures at the cervical level or cervicothoracic junction. AS related cervical fractures (ASCFs) require early diagnosis and a treatment plan that considers the high risk for additional fractures to avoid neurological complications or death. We present the case of a patient with an ASCF and a review of the literature with key recommendations that shape our algorithm for the proper diagnosis and treatment of ASCFs. We present the case of a 29-year-old male with an ASCF at C5-C6 treated initially with a short segment instrumented arthrodesis that required an additional operation to properly stabilize and protect his spine. Based on our experience with this case and a review of the literature, we discuss three recommendations to improve ASCF management. These include the need for early computed tomography/magnetic resonance image for proper diagnoses, combined surgical approach with long-segment stabilization for maximum stability. Delayed diagnosis or revision surgery, both of which are common in these patients who present with a stiffened and osteoporotic spine, may lead to spinal cord injury or neurologic deficits. Our recommendations based on the most recent evidence can help surgeons better manage these patients and decrease their overall morbidity and mortality.

Keywords: Ankylosing spondylitis, cervical fractures, combined surgical approach, imaging, long segment fixation

INTRODUCTION

Ankylosing spondylitis (AS) is a chronic inflammatory condition named after the ankylosis of the apophyseal joints and sacroiliitis seen in affected patients. This is accompanied by gradual ligament ossification resulting in the rigid bamboo spine and osteoporosis, making the ankylosed spine, especially susceptible to fractures.[1] Even minor trauma or no trauma at all can trigger a fracture[2-7] which often goes undetected in this patient population for a variety of reasons.[8] The percentage of bone fractures in AS patients is higher than in the nonaffected population. A majority of these fractures occur in the lower subaxial cervical spine, which must be treated promptly and adequately, considering a third of cervical fractures in this patient population are neurologically devastating and can be fatal.[9] We present the case of a 29-year-old male with AS who was surgically treated for his cervical fracture initially with a short segment instrumented arthrodesis that failed, requiring subsequent stabilization posteriorly with an extended segmental fixation of the cervical spine. We have reviewed the most recent literature on the need for initial anterior and posterior long-segment stabilization of cervical spinal fractures in patients' with AS and provided our algorithm to the approach of these patients

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based on the biomechanics of the fracture in AS and a review of relevant literature.

CASE DETAILS

A 29-year-old male with a past medical history of AS (HLA B27+) presented to the ED after a fall that occurred sometime earlier in the day, leaving him unable to walk along with acute neck pain and radiculopathy. Computed tomography (CT) of the cervical spine showed a hyperextension distraction injury with retrolisthesis of C5 on C6 involving the discoligamentous complex, an avulsion fracture of the C6 endplate and bilateral C6 transverse process fractures with the right extending to the transverse foramina along with fractures posterior through the fused facets at C5-C6 [Figure 1a-d]. Magnetic
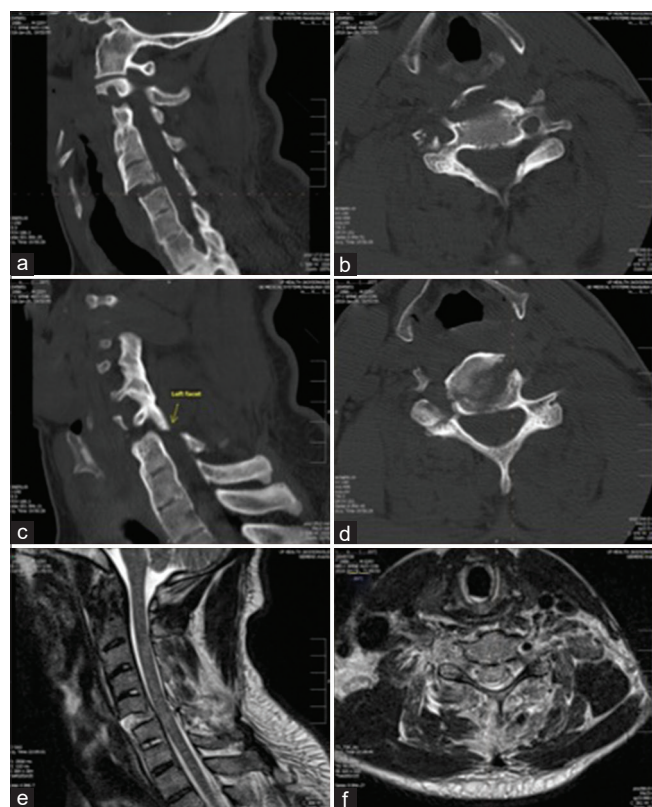
Figure 1: Computed tomography and magnetic resonance image images before 1st surgery. (a and b) Computed tomography cervical spine sagittal and axial view revealing characteristic findings of ankylosing spondylitis with ankylosis of the disc spaces and facet joints evident by ossification of the anterior longitudinal ligament (ALL)/posterior longitudinal ligament (PLL) and facet joints. Injury is a distraction hyperextension with disruption of the discoligamentous complex (DLC), widening of the disc space and the facet joints. (c and d) Computed tomography cervical spine sagittal and axial view with cross-section more lateral at the facet joint revealing and disrupted and near jumped facets with evident subluxation. (e) Magnetic resonance image of the cervical spine T2-WI sagittal section revealing a C5-6 hyperextension distraction injury involving the DLC with disruption of the ALL, PLL, PLC and ligamentum flavum with anterolisthesis of C5 on C6. (f) Magnetic resonance image axial section through the C5-C6 disc space revealing mild hyperintense changes within the cord with stenosis and effacement of thecal space and right VA dissection

resonance image (MRI) revealed acute hyperextension injury at C5-6 with associated anterior and posterior ligamentous disruption at C5-6 [Figure 1e-f]. There was cord impingement at C5-6 as well as cord contusion. MRI of the thoracolumbar spine was negative for any acute injury.

The patient underwent an anterior cervical discectomy and fusion (ACDF) of C5-6 [Figure 2], with some postoperative residual weakness and paresthesia, without any new deficits followed by discharge to a rehabilitation center.

The patient was progressing well when he heard a pop in his neck 2 weeks later. Clinically, he had worsening neck pain with a new radiculopathy involving the right upper extremity in the C6 distribution with sensory changes on the right side. We immediately repeated X-ray imaging which revealed widening of the facet joints posteriorly and increased kyphosis at the site of the ACDF [Figure 3]. Further evaluation with a CT angiogram of the neck revealed worsening vertebral artery dissection on the right side to a complete segmental occlusion along with posterior widening and movement of the prior C5-C6 construct with increased kyphosis and coronal curvature to the right [Figure 3]. Given the worsening clinical findings, evidence of instability on X-ray imaging and the need for heparinizing the patient for the right VA occlusion we decided to perform a posterior C3-T2 arthrodesis. The procedure was a standard posterior cervicothoracic fusion with midline open dissection, exposure of the lateral mass facet complexes, and the entry points at T1, T2 for the pedicle screws followed using appropriate instrumentation. The challenges with AS-patients include the significant blood loss that occurs during the dissection[10] and the loss of posterior bony facet anatomy because of the extensive bony ankylosis with no facet joints being evident. In this case, we used intraoperative image guidance with the Medtronic O-arm

Figure 2: Immediate postoperative X-ray following the anterior cervical discectomy and fusion
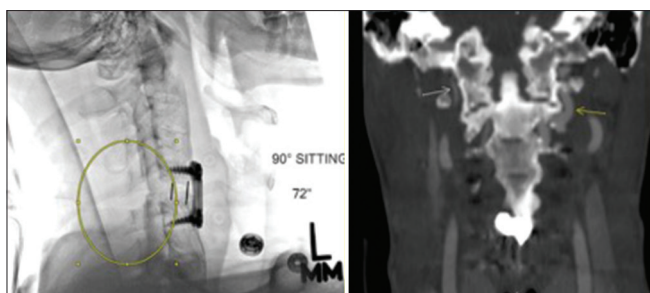
Figure 3: Left: Lateral X-ray revealing increased kyphosis and widening of the facets posteriorly (area within oval). Right: Coronal view computed tomography angiogram of the neck revealing right complete VA occlusion with collateral flow (white arrow) and a normal contralateral left VA (yellow arrow)

and Stealth navigation for identification and insertion of the lateral mass and pedicle screws. Following the posterior cervical stabilization, the patient was placed on a heparin drip and transitioned to antiplatelet therapy for his dissection. The patient continued to improve with rehabilitation, with no further sequelae on follow-up at 6 months.

DISCUSSION

For our patient, an ACDF provided inadequate biomechanical stabilization of his cervical fracture and required an additional posterior long segment fusion to prevent further vascular and neurological injury and associated complications from rotation around the ACDF acting as a fulcrum [Figure 4]. AS patients are at an increased risk for distraction coronal and axial rotation given their rigid, osteoporotic "bamboo" like nature of the spine that can lead to damage of the spinal cord, vascular structures or peripheral nerves resulting in quadriplegia or even death. Fracture risk peaks in the first few years following AS diagnosis and seems to be associated with AS severity.[11]

In the past, nonsurgical treatment was the preferred choice for stable and most unstable fractures with poor long-term outcomes.[12] Treatment options generally consisted of external orthoses, cervical collar, or low weight traction.[1,13] Stabilization with surgical management is now the standard in these patients, as it is associated with lower rates of pseudarthrosis and neurologic deficits.[12] This is significant considering AS patients have a higher frequency of unstable 3-column cervical fractures, where there is greatest risk for spinal cord injury, in comparison to the thoracic, lumbar, or sacral spine.[2,14,15] Cervical spine fractures in AS patients are roughly 50% more common than in other parts of the spine due to the structure and function of the cervical vertebrae.[16] They are generally unstable with a 3.5 times higher associated morbidity and mortality than fractures in non-AS patients.[17]
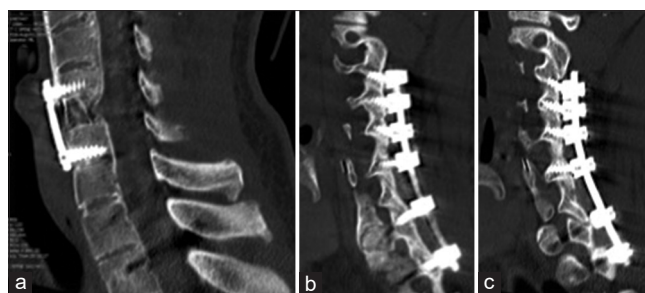
Figure 4: (a-c) Postoperative computed tomography cervical spine images sagittal midline, sagittal through right and left facets revealing good osseous fusion and reduction of the joints, kyphosis

Currently, there are no standard guidelines for diagnosing and treating cervical fractures in AS-patients. Although a treatment algorithm from 2008[18] does exist, we present critical recommendations and emphasize the need for multi-level anterior/posterior or combined fusion-at least 2 levels above and below the fracture line as shown in our case report, literature review, and algorithm.

These recommendations include:
1. Immediate use of diagnostic CT/MRI and angiographic imaging of the cervical vessels due to risk of dissection and thromboembolic events
2. Stabilization through a combined anterior and posterior approach
3. Long segment fixation at least 2 levels above and below the fracture when possible.

Clinicians should have a high index of suspicion for spinal fractures with AS patients presenting following minor trauma or back pain in the absence of a clear medical history. These patients should undergo CT imaging of the spinal axis, and an MRI of the appropriate level based on the clinical exam findings, if possible. AS changes are characterized by formation of cartilage in unusual areas, resulting in calcification of nonbony structures, accompanied by degeneration of the intervertebral discs and facet joints posteriorly. Trauma causes disruption of the fused disc ligament complex or occurs through the fused vertebral body/facet joints resulting in vertebral dislocation causing major deformity and instability.[19,20] Given this 3 column instability, even low-energy trauma can cause fracture and posterior ligamentous destruction in these patients, especially at the subaxial cervical spine and cervicothoracic junction, since fractures are more likely to occur in fused segments and/or at the connection of nonaffected and fused spine.[4,16]

Although plain x-ray radiographs (XR) are the first choice to evaluate AS patients,[21] bony changes and calcifications make XR interpretation difficult, especially at the cervico-thoracic junction and many fractures are therefore left undetected in

this patient population.[22] Anwar *et al*.[23] in their retrospective case series of AS patients over a 12-year period identified a delay in treatment of these fractures with high rates of neurological worsening at their institution. CT scans were able to detect over 60% of the fractures that were not identified on X-ray imaging and is a more sensitive and specific imaging modality in AS. MRI scans can be used as adjuncts in case of suspected neurological injury. A CT angiogram of the neck vessels also provides information about possible injury to the vertebral vessels and should be performed with the initial imaging.

A negative XR should always be followed by CT to detect bony changes and single or multi-level displaced or nondisplaced fractures.[24,25] An MRI is essential if ligamentous destruction or spinal cord injury is suspected.[22] MRI may not be possible, especially in patients with cervical hypokyphosis;[26] however, it may provide information on the presence of cord compression, injury to the cord or contusions that can alter the treatment plan. Posttraumatic epidural hematoma has a 50% higher incidence in AS patients and can also be ruled out with an MRI[27] [Figure 5]. Tavolaro *et al*.[28] conducted a study specifically looking at whether an MRI is
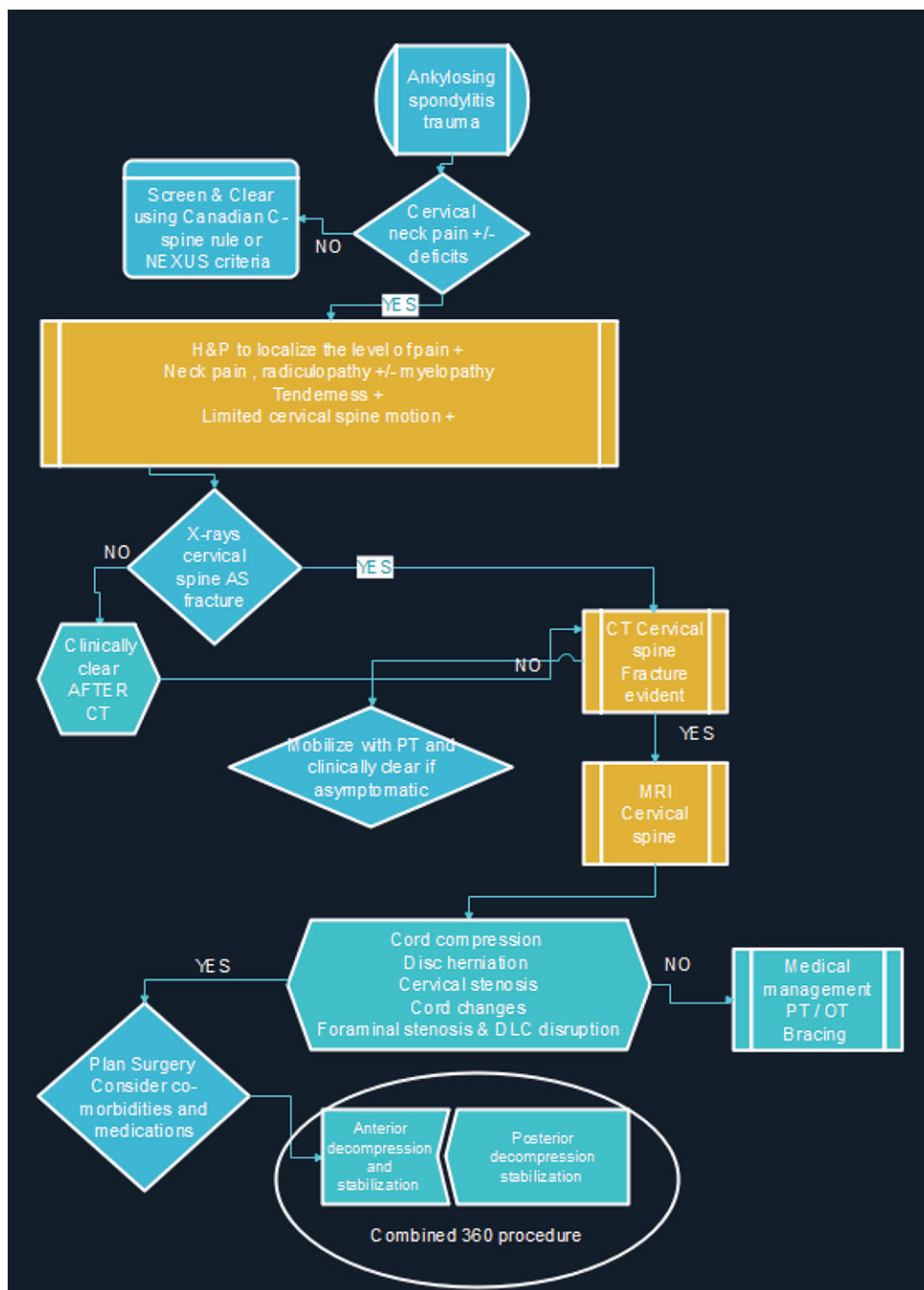

Figure 5: Algorithm for assessment and management of traumatic cervical spine fracture in ankylosing spondylitis patient

necessary in diagnosing these patients in addition to a CT and recommended that it be used routinely in patients with suspected disco-ligamentous injury or those presenting with neurological deficits to assess for a potential cause. Early appropriate imaging is important for diagnosis, surgical planning, avoidance of progressive neurological injury, and to reduce mortality in this high-risk patient population.[4]

Based on the specific location/type of fracture and associated deformities, surgeons can decide whether to use an anterior, posterior, or combined surgical approach when surgically managing these patients. AS cervical spine fractures frequently involve three columns of the spine, causing disruption through the disc space and facet region where hyperostotic fusion has occurred or through the osteoporotic bone in the vertebrae. In a patient with AS and a distraction/hyperflexion or hyperextension injury, the spine has greater load bearing across the fracture line because of the long lever arms that create greater torque forces on the fracture line that acts as a fulcrum [Figure 6a-d]. Torque is defined as the product of the force and the distance of the force from the fulcrum along with the angle and because the cross product takes force that acts in a perpendicular manner, the angle between the upper and lower segment with the fracture line are important, along with bone quality and muscle strength.
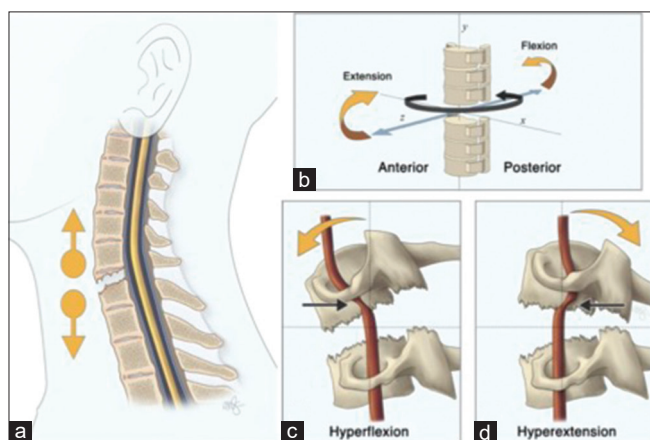
Performing a small segment fusion across the fracture line with an ACDF or posterior instrumentation does not provide the necessary stability to counter the stronger torque forces resulting in construct failure, as was seen in our case. When this is performed anteriorly, due to the complete disruption of

Figure 6: (a) Illustration revealing the destructive nature of the forces with the fracture line passing through the DLC. The lever arms in ankylosing spondylitis are longer due to the ankylosis at adjacent joints increasing torque forces around the fracture line. (b-d) Illustration revealing the forces in the coronal and axial planes that can cause disruption and worsening of the fracture. As the lever arms are long in patients with ankylosing spondylitis, the forces are much greater and short segment forces fusions cannot control for movements in the coronal and axial planes, and the sagittal plane with resultant increased failure rates

the posterior elements, there is likely to be acute, subacute, or chronic delayed failure of the construct and the need for long segment posterior fusion across the fracture line in almost all cases. The fracture site in AS acts as a fulcrum with the spine and cranium above acting as a lever arm and the subaxial spine and below acting as the other lever arm. To balance the cervical spine in AS, the paraspinal muscles have to work harder to support the cranial segment and prevent torque forces from resulting in construct failure.

In the normal cervical spine, every intact spinal level adjacent to the fracture can provide support to the spinal segment, with the lever arms above and below the fracture spanning only a single spinal functional unit. However, in AS because of the abnormal hyperostosis, the level arms are much longer and the torque forces are much higher, giving rise to inability of the muscle to prevent worsening strain/stress on the implant system and its resultant failure.

The anterior approach provides decompression of the neural element and transient stability by holding the fragments together to give the treating team an opportunity to treat any other medical problems in these patients postoperatively. Failure rates of approximately 50% occur due to delayed screw loosening, poor bone quality, and implant mobilization during the follow-up period. This approach also does not resist the tension from injuries in the posterior aspect of the vertebrae contributing to its poor stress resistance.[29] Einsiedel *et al.*[19] reported no longer performing anterior only stabilizations for these reasons and relying on the combined approach. The posterior approach is associated with better clinical outcomes than the anterior approach but is also reported to have a high failure rate due to the lack of support from the anterior aspect of the spine for rotational movements.[29] Therefore, despite patients with AS having associated comorbidities such as those with cardiopulmonary issues, the biomechanics and the need to prevent neurological injury at the cervical spine make the combined 360° approach the treatment of choice for these patients.[4]

Currently, there is no consensus for treating cervical fractures in AS patients. Although anterior spinal fusion was initially the method of choice for these cervical fractures, the literature provides support for a posterior and combined 360° approach in these patients given the associated lower failure rates and reduced morbidity and mortality.[2] However, in terms of biomechanical stability, the combined approach provides the greatest support to stress during rotational movements, forces of torque at the fulcrum which are greater in the AS cervical spine and again hyperflexion/hyperflexion of the spine making it the preferred treatment option.[24,30]

Longo *et al.*[21] evaluated the benefits and complications of anterior, posterior, or combined surgical repairs in patients with AS-related spine fractures. They showed that the combined and posterior approaches provided greater clinical benefit (measured by the Frankel score and clinical neurologic examination pre- and postoperatively, lower postsurgical complications and mortality rate) when compared to the anterior approach. They also concluded that the combined approach may help to decrease the risk of secondary neurological complications in AS patients due to fractures in fused segments given the additional stability.[31]

This was echoed by the German Society for Orthopedics and Trauma (DGOU) and others who have also recommended the combined approach to treat fractures in AS patients secondary to better outcomes and achievement of higher spine stability.[24,30]

Most of the controversy surrounding the use of a combined approach comes from avoiding the need to use an invasive and costly approach when these patients are already at risk for increased complications and mortality.[14,32] However, we believe that the benefit in this case far outweighs any additional risk or cost. The combined approach should be the goal for surgical management of these patients out of an abundance of caution for the increased risk that these patients are faced with.

In addition, long segment posterior fixation in AS-associated cervical fractures is recommended to achieve greater stability and to avoid revision surgery. Kanter *et al.*[18] briefly mentioned this need to include several segments, cephalic and caudal to the fracture site. Given the altered spine of an AS patient, it is important that the primary goal be to ensure complete stabilization rather than limiting the number of fused levels to maximally retain spine mobility. In addition, given poor bone quality, surgery is associated with a high rate of complications such as loosening, pull-out, and screw migration.[33] Long segment fixation will not only provide better radiological outcomes but also protect the patient by ensuring stronger stabilization and a lower incidence of mechanical complications.[34]

Although long segment stabilization from an anterior approach may not be possible for many patients given their presentation and cervical spine morphology, the posterior approach is easier from an operative standpoint and can further decrease the load arm acting on the fracture site, making additional fractures less likely.[35] From a biomechanical standpoint, longer segments of fixation (at least 3 levels) provide the most stability and lowest maximal stress with a combined anterior/posterior approach and then next with a posterior alone approach.[35]

CONCLUSION

Following our case and a review of the current literature, we present three recommendations that shape our algorithm for diagnosing and treating AS patients presenting with cervical fractures. These patients require extensive and careful imaging for a proper diagnosis that then determines their plan of care. Given our goals for treatment, we emphasize the importance of achieving long segment fixation with a combined approach when possible.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardian has given his consent for images and other clinical information to be reported in the journal. The guardian understands that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Conflicts of interest
There are no conflicts of interest.

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