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Management of a case of neglected atlantoaxial rotatory dislocation

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Full Text

Sir,

A case of complex craniovertebral anomaly with severe atlantoaxial rotatory dislocation along with abnormal bone fusion and bone formation is discussed. The role of manual facetal manipulation and attempts at craniovertebral realignment is re-emphasized. A 30-year old farmer was well 2 years ago. He was involved in a fistfight with a friend. One of the fist blows landed on his chin and he suffered significant acute neck pain. Apart from this neck pain, he was well. Following the injury, the patient continued to have pain in the nape of the neck. Approximately 2 months later, he started noticing tilting of his head and chin to the left side that progressively increased in severity. The torticollis gradually increased and was accompanied by the dropping of his head and chin towards the medial end of the left clavicle. He was unable to straighten his neck and experienced continuous excruciating neck pain. He started holding his head up with his left hand to prevent the drooping of his head and also for limiting the neck pain. In order to look straight ahead, he would extend his back and hold the neck and mandible with both his hands. This act resulted in severe pain. Moreover, he could not sleep in a supine posture as it caused breathlessness and neck pain. Thus, he preferred the lateral position. He also lost his job. When he presented for treatment, he had a nearly fixed neck deformity with the chin turned towards the left side, abutting the clavicle, and the vertex rotated towards the right side. His neurological examination was entirely normal.

His investigations included a dynamic computed tomography (CT) scan, magnetic resonance imaging (MRI), and three-dimensional (3D) model construction [Figure 1]. The craniovertebral junction bone malformation neither complied with any known or described syndrome nor simulated any discussed nomenclature. There was an abnormal bone growth of the lower end of the clivus as well as of the arches and facets of atlas. There was severe rotation of the atlas over the axis. The angle of rotation of the atlas along the horizontal plane was 65°. There was vertical or

facetal collapse that resulted in superior migration of the odontoid process. Despite the bone abnormality, there was no neural compression or compromise.{Figure 1}

Surgical treatment was planned with a strategy, as per our published articles, on lateral mass fixation, facet manipulation, and distraction techniques, as well as on the treatment of rotatory atlantoaxial dislocation.[1],[2],[3],[4] The patient was placed in a prone position and the head was placed under traction. Head rotation made the positioning for surgery difficult. The 3D model was used as a surgical guide and was placed in direct view of the surgeon throughout the procedure.[5] Both the atlantoaxial joints were exposed widely. An osteotome was used to distract the facets as widely as was possible. The distraction was done more widely and forcefully on the left side. Using osteotomes simultaneously on both the sides, an attempt was made to realign and reverse-rotate the joints. The abnormality of bone formation had resulted in unnatural twists of the vertebral artery, obscuration of its foramen, and a course that was difficult to trace during surgery. The preoperative imaging and the 3D model assisted in safeguarding the artery and helped in inserting the screws at the optimum site and in the proper direction in the lateral masses of the cervical vertebrae.[6] Large amounts of bone grafts were placed inside the articular cavity on both the sides and a plate and screw fixation was done. Bone was harvested from the iliac crest. Postoperatively, the patient was entirely relieved of his neck pain. He could now lie down and sleep in a supine posture, a position that he was unable to use for almost 2 years. He could now look straight ahead, although he still had some degree of torticollis. Repeat investigations showed that the odontoid process had now descended significantly into its normal position. The rotation of the atlas was reduced to less than 30° [Figure 2]. At a 14-month follow-up, the patient had significant residual torticollis; however, he could look straight ahead without any discomfort. He was essentially asymptomatic and had started working to a limited extent as a farmer.{Figure 2}

Craniovertebral junction is a complex organization of bones and joints that provide the region with a stable as well as a mobile arrangement while permitting the safe passage of the most critical neural and vascular structures. The complexity of the architecture and versatility of its functions places a significant strain on the anatomical organization of the region. Several musculoskeletal and neural anomalies have been recorded that are speculated to be the result of embryonic dysgenesis or of protective natural maneuvering.

The occipitoatlantal joint is credited to be the most stable joint, and the atlantoaxial joint, the most mobile joint of the body. To provide for circumferential movements, the articular surfaces of the atlantoaxial joint are flat and rounded. While this formation provides a ground for a wide range of mobility, it also makes the C1-2 joints susceptible to an enhanced risk of instability and dislocation. In our earlier publication, we speculated that the atlantoaxial joint might well be the most unstable joint of the body. In addition to the generally recognized form of anteroposterior dislocation, atlantoaxial joint can have lateral, vertical, central, axial and rotatory forms of dislocation. Identification of the atlantoaxial instability by the pattern of facetal malalignment; and, a direct operative facet joint handling, has expanded the scope of understanding of the region and has provided an additional methodology for the treatment of these anomalies.

While occipitoatlantal dislocation is distinctly rare, atlantoaxial instability is relatively common.[6] Minor injuries or even a tap on the head have been identified to result in rotatory atlantoaxial dislocation in children.[2] However, in an otherwise healthy adult, only major motor vehicular accidents have been associated with atlantoaxial instability. The exact cause of the craniovertebral junction malformation in our patient, whose work involved heavy physical labor, is unclear, as the fist blow that the patient sustained was highly unlikely to result in such a major distortion and disruption of the craniovertebral junction. It can only be speculated that the 2-year period of avoiding treatment could have led to bone malformation, spinal deformation, and an abnormally asymmetrical bony growth. It is more likely that the patient had an anatomically abnormal craniovertebral junction, the malalignment of which got exacerbated over time and then, the patient developed acutely manifestations as a result of the trauma sustained by him.

The distraught clinical condition of the patient, who was having a grotesque craniovertebral malformation, presented a therapeutic challenge. In addition to the element of severe rotation, the unusual bone growth and abnormal bony fusions made the conceptualization of surgical treatment difficult. Apart from the CT scan and MRI, a 3D model of the region provided a clear anatomical delineation of the bones, the location and alignment of the facets, and defined the relationship of the vertebral artery to the bone complex.[5],[6],[7],[8],[9] Facetal distraction and manipulation under

direct surgical vision and radiological monitoring, as first conceptualized by us in the year 2000 and published in the year 2004,[1] has ushered in a new era, for the treatment of a variety of craniovertebral junction-related issues. Stabilization of the unstable joint is the prime issue in treatment.[10] Wide denuding of the articular cartilage, bilateral facetal manipulation and attempts at craniovertebral realignment, introduction of the bone graft and spacers within the joint cavity, and a firm atlas and axis facetal screw fixation provides an adequate opportunity for a good bone fusion of the C1-2 joints.

In conclusion, facetal manipulation, distraction, manual realignment, and fixation can lead to restoration of even the most severe forms of craniovertebral junction distortion.

Declaration of patient consent

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

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Nil.

Conflicts of interest

There are no conflicts of interest.

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