Odontoid Synchondrosis Fracture in Preschool Children: Report of Two Cases with Special Reference to a New Reduction/Fixation Technique

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ABSTRACT

Fractures of the odontoid process are among the most common cervical injuries in preschool children, occurring at an average age of 4 years. These fractures are actually physeal injuries of the basilar synchondrosis between the odontoid process and the body of the axis.

Management of the acute odontoid synchondrosis fractures in children is divided into surgical and conservative. Majority of the dontoid synchondrosis fracture heal with external immobilization, but a minority with marked displacement as well as those which are neglected require open reduction and internal fixation.

Herein, 2 boys at the age of 4 years with odontoid synchondrosis fracture are presented. One was admitted shortly after injury and the other after 4 months the acute one was managed with pediatric halo and the neglected one with irreducible atlantoaxial dislocation underwent open atlantoaxial reduction by means of transfacet release and subsequent complete reduction by a new maneuver and instrument, which will be described in detail.

KEY WORDS: Atlantoaxial dislocation, cervical spine, children, odontoid synchondrosis fracture, posterior C1-C2 fixation

INTRODUCTION

Spinal injuries in small children are rare with a reported incidence of 0.2 to 0.5% of all fractures or dislocations and 1.5 to 3% of all traumatic lesions in the spine (4,8,12,14,30,31,36,37). Furthermore, cervical spine injuries are uncommon in children and the estimated annual incidence of pediatric cervical spine injuries is about 7 per 100.000 population and around 1 per 100,000 population for younger than 11 years of age (31). The upper cervical spine is more susceptible region to trauma in the children in this age group (4,8,12,14,15,23,30,31,36,37,44). Among the upper cervical injuries, odontoid remains the most vulnerable part (1,2,13,15,20,21,23-25,28,29,32,34,35,39-41,43-45). In fact, odontoid fractures are apophyseal fractures where the fracture line passes by the synchondrosis, a cartilaginous plate that normally closes at age of 7 years (1, 2, 1013, 33, 40, 41, 45).

If an odontoid synchondrosis fracture is diagnosed early, immobilization after approximation of the fractured odontoid to the axis either with hyperextension of the neck or traction offers the optimal option (1,2,13,15,20,21,24,25,33,35, 39,41,44,45). However, a subtype of acute ones with atlantoaxial instability either due to marked angulation of the dense (more than 30 degree)or significant forward displacement of the fractured odontoid (more than 100%) which indicate instability will require reduction and primary posterior C1-C2 fixation (15, 23, 35). Furthermore, in a small proportion of the children, the injury might be missed being presented with chronic atlantoaxial dislocation either due to failure of external immobilization or as the result of neglecting the fracture and its diagnosis with delay (1,15,35). In this chronic subtype, most of the dislocation are irreducible where the treatment remains a complicated issue requiring a challenging surgery.

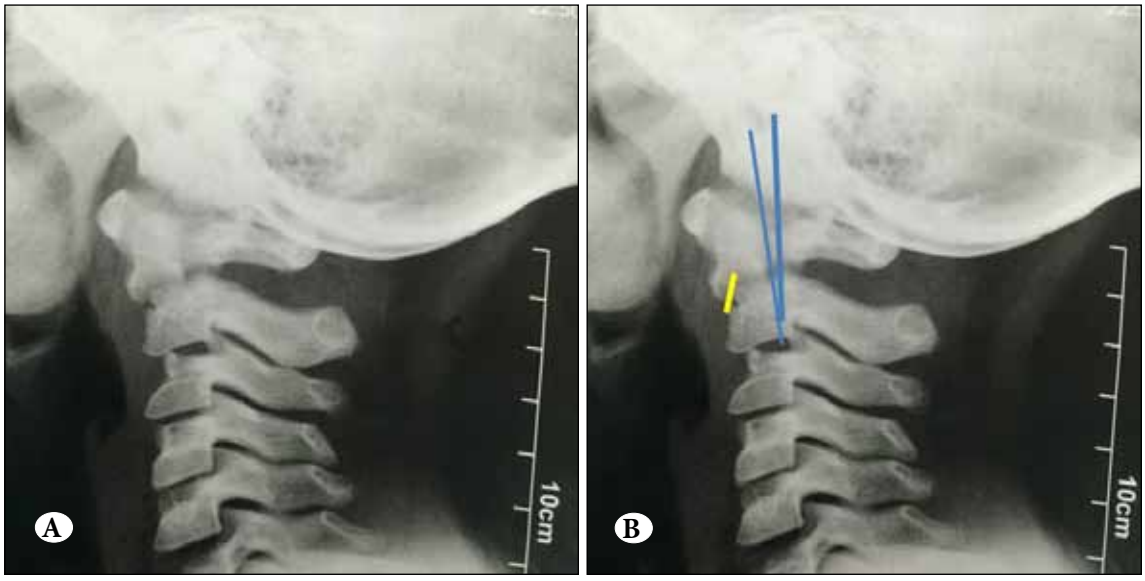

Figure 1: *Lateral radiograph of the Case1, s* ***(A)*** *howing odontoid synchondrosis fracture with mild displacement* ***(B)*** *the fracture angle is 5, and displacement is below 10%.*

Figure 2: *Sagittal reconstructed C.T. scan demonstrates Odontoid synchondrosis fracture.*

Herein, two new cases are presented, one was diagnosed within a few days and the other 4 months after trauma. Alignment of the fractured odontoid to axis was made with closed and open reduction respectively, where fusion could be achieved with a halo vest in the former, in the latter became possible with internal reduction and fixation, both with good outcomes.

A new formulation for irreducible atlantoaxial dislocation due to odontoid fractures and a new technique for either primary C1-C1 fixation or reduction of the dislocation and secondary posterior C1-C2 fixation will be described.

CASE REPORTS

Case 1: A 4-year-old boy sustained trauma in a motor vehicle accident. He was sleeping on the passenger seat without restraint when the driver crashed another car. The casualty remained conscious but complained of neck pain. Therefore, he was transferred to a nearby hospital where an odontoid synchondrosis fracture with mild anterior displacement and angulation was noticed on lateral radiograph. The fracture angle was 5 degrees and its forward displacement was below 10% (Figure 1A, B). He was transferred to our institute and C.T., which was obtained subsequently, confirmed the odontoid fracture (Figure 2). The odontoid fracture was also clearly demonstrated in MRI (Figure 3).

The displaced odontoid fracture could not be reduced with extension of the neck, but it was reduced with traction. After reduction, the child's head and neck were immobilized with halo-jacket (Figure 4). This was preserved for 12 weeks and was removed when union was observed at fracture site (Figure 5). At 6 months follow up, flexion and extension views of the upper cervical spine showed no instability.

Case 2: This 4-year-old boy was involved in a motor vehicle accident. He was sitting on the rear seat unrestrained when his father's car was involved in head-on collision with another car. He was transferred to the nearby hospital. Since, initial assessment did not reveal any serious injury and he was discharged a few hours later. But, after passing 4 months the child started to complain of neck pain. The parents noticed that he holds his head in his hands while looking around or walking. On examination, tenderness on palpation of the upper cervical spine was noted and the range of neck motion was quite restricted.

Radiograph revealed a displaced odontoid fracture, which was not reducible with the extension of the neck (Figure 6). Reconstructed C.T. scan confirmed odontoid synchondrosis fracture with marked displacement. MRI also showed Marked forward displacement of the dense and its angulation along with atlantoaxial dislocation, the fracture angle of 35 degrees and its displacement was 80% (Figure 7). The distance between the spinal process of C2 and its counterpart C1 was significantly increased. With a diagnosis of atlantoaxial dislocation due to odontoid fracture, traction was attempted, starting with 800 mg and increasing it to 6.5 kg gradually. However, the dislocation did not reduced despite prolonged traction. Therefore, open reduction via posterior C1-C2 facet joints release with subsequent C1-C2 internal fixation were considered.

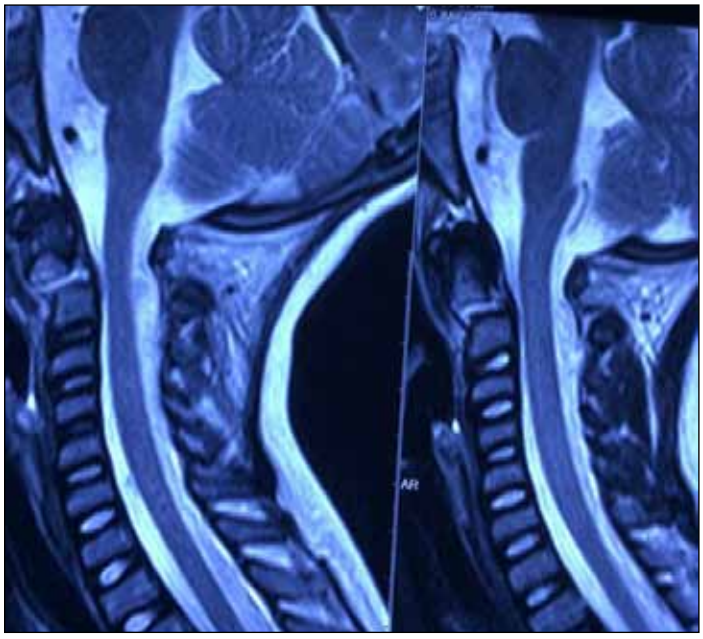

Figure 3: *Sagittal 2-weighted MRI of the cervical spine showing the fracture.*

Under general anesthesia, initially transoral manipulation was attempted while traction was continued, but, the deformity persisted. Thereafter, with the patient in prone position, laminas of C1 and C2 were exposed, Actually C1 facet joint had slipped forward with respect to C2. The capsules of the joints were partially destroyed and replaced by scar tissue. The cartilage and the scar tissues were removed with curettes and small cutting burrs. Later an osteotome like tool was inserted in the corresponding joint on each

Figure 4: *The child in halo vest.*

Figure 5: *Lateral cervical X-ray, at 6-months follow up demonstrates fusion at the site of fracture.*

side. Now,with their simultaneous rotation the joint was opened sufficiently and were freed. Thereafter, two polyester tapes were passed below C1 lamina and two below C2 laminas. Then a rod was placed over the laminas of C1 and C2 between the free ends of polyester bands on each side. Initially, C2 sublaminar bands were tightened on the corresponding rods with the aid of Universal clamps (Zimmer). Then, with tightening of C1 sublaminar bands attached to Universal clamps, on the cranial part of the rods, the atlas ring was pulled back and reduced to its normal position. The excess of the bands were cut and allograft chips mixed with bone marrow aspirate were distributed on the already decorticated posterior elements of C1 and C2 (Figure 8). Fluoroscopy revealed full reduction of the deformity, and relocation of the odontoid process in its normal position.

Figure 6: *Lateral radiograph of the Case2, showing odontoid synchondrosis fracture with marked displacement and angulation.*

At 6-months follow up, the patient was doing well and fusion of odontoid to the body was demonstrated (Figure 9).

DISCUSSION

Traumatic injuries of the cervical spine are uncommon in children and fundamentally different from their adult counterparts (4,8,10,12,14,3,31,36,37). The reason is the greater flexibility and resiliency of the pediatric column, allowing the force of injury to be dissipated more easily over a greater number of segments (4, 8, 10, 12, 14, 3, 31, 36, 37).

Young children are more likely to have upper cervical lesions than subaxial cervical spine. It has been shown that the younger the child at the time of injury, the more likely the upper cervical spine is affected (4,8,10,12,14,3,31,36,37). Eventually, over 50% of spinal injuries in small children affects atlantoaxial region and in particular occurs most often

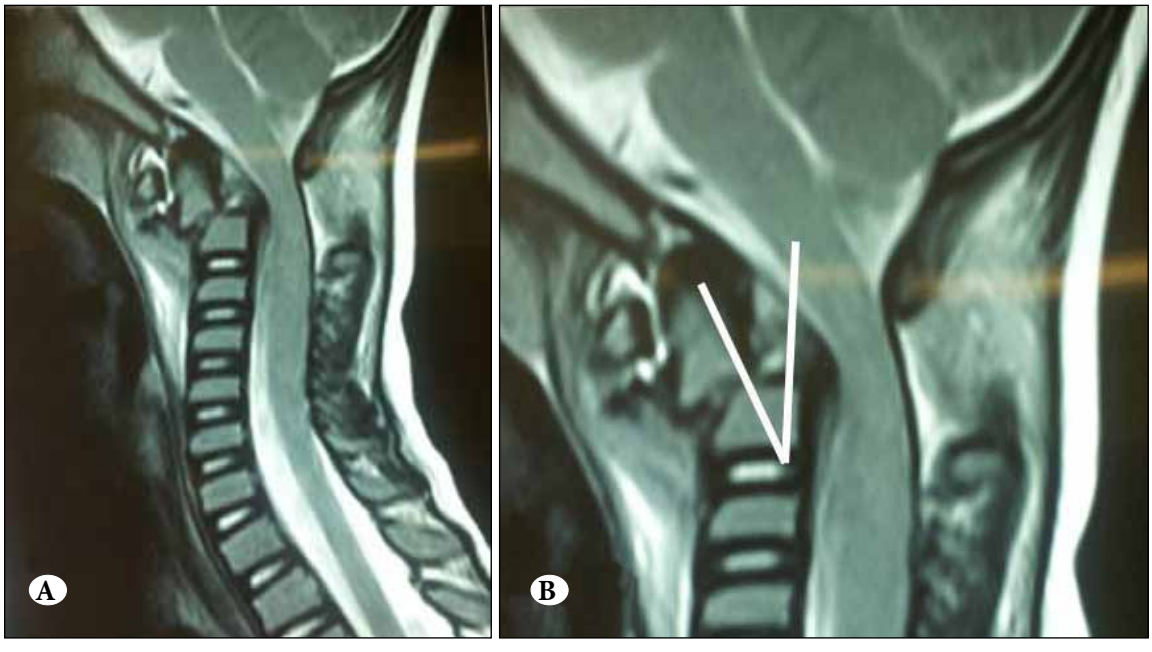

Figure 7: A) *T1-Weighted MRI showing odontoid synchondrosis fracture.* ***B)*** *The fracture angle is 35, and displacement is about 80%.*

at the odontoid level (1,2,10,13,15,20,21,23-25,28,29,33-35,39-41,43-45). Typically, odontoid fractures in children passes through the basilar synchondrosis, a cartilaginous plate that separates the odontoid process from the body of the axis (1,2,10,13,15,20,21,23-25,28,29,33-35,39-41,43-45). The average age at which children sustain odontoid fractures is 4 years (1,2,10,13,15,20,21,23-25,28,29,33-35,39-41,43-45). Most often the fractured part tends to be displaced anteriorly during the flexion movement of the cervical spine.

Figure 8: *Intraoperative photograph, demonstrating polyester bands fixed on the C1-C2 rods by Universal clamps.*

Figure 9: *Post operative lateral cervical radiographs showing proper alignment of odontoid, Universal clamp-rod has fixed C1-C2 arches.*

Obviously, greater force can result in atlantoaxial instability or even dislocation. Because of the instability associated with these lesions, patients are at significant risk for immediate or delayed catastrophic spinal cord compromise (1,13,34,35).

In Western countries, the most common mechanism of such an injury is a head-on motor vehicle accident with a toddler restrained in a backseat by a four-point children's seat harness (25, 31, 33). Both of our patients were sitting on the rear seat unrestraint, quite similar to several other reports from the west (2,44).

Clinical Picture: The clinical features of the children with odontoid synchondrosis fractures are quite variable and can be described in a spectrum. In one side of the spectrum are cases with severe spinal cord injury (15,28,44). While on the other side of the spectrum stands children that are symptom free (2,13). According to Fassett et al 77% of the patients will not experience neurological deficit. In the middle of the spectrum, there are some children with neck pain. Along with pain, limited neck mobility is usually present (25).

The most problematic issue is when the odontoid fracture is missed (1, 15, 34, 35). This mostly occurs in the symptom free cases where the correct diagnosis will require high index of suspicion. In addition, in those who escape and survive from serious multi-organ injuries, odontoid fracture might be easily neglected. In these circumstances, from several weeks to months after the initial accident, neck pain of increasing intensity will be the first clinical feature. Furthermore, the neck appears to be shortened, with development of neck kyphosis, the chin moves closer toward the sternum.

Accordingly, the clinical features of cervical myelopathy will ensue where its severity depends on the degree of atlas displacement on the axis. It is proposed that late myelopathy following odontoid fracture may be more common than hitherto believed and should be considered in the evaluation of progressive disability in the children after initial recovery from severe head injury. Examples are patient of Odent 1999 and of Alp and Crockard in 1990 that delayed diagnosed were made after 6 months and 15 years respectively where both patients had quadriplegia on admission (1, 35). In missed cases with only neck pain as their cardinal clinical feature, the story might be more catastrophic where a trivial trauma may precipitate quadriplegia, respiratory arrest and even death. Osgood and Lund reported the natural history of 55 patients with odontoid fracture, 10 out of these patients

were asymptomatic at the time of initial injury, thus were missed. All these 10 died after second trivial injury (1,35).

Imaging: Prompt radiologic evaluation of children with neck pain or stiffness after any accident that causes a forced hyperflexion of the neck is necessary for visualization of synchondrosis fracture (10, 28, 40, 41, 44).

However, typically, shear stress at the synchondrosis between the body and the odontoid in hyperflexion leads to anterior angulation in 94% and forward dislocation of the odontoid in more than 90% of the children with odontoid fracture (37). Actually, the severity of the fracture can be determined with fracture angulation or its forward displacement (23). The fracture angle is defined as the angle between the perpendicular line of the base of the vertebral body of axis and the tangent line to the posterior surface of the odontoid process (10,15,23).

In the case of doubt, flexion/extension views are helpful. However, computed tomography and in particular, reconstructed sagittal and coronal images can allow to make correct diagnosis (45).

MRI is very helpful in excluding injuries of the posterior ligamentous structures in acute cases. Furthermore, detection of the degree of canal compromise and associated myelopathy are other advantages of MRI.

TREATMENT

The treatment of odontoid synchondrosis fracture in children is quite different in acute and chronic cases. Therefore, each should be discussed as separate issues

Acute odontoid synchondrosis fractures: The management of acute odontoid fractures in children depends on the type of the fracture according to classification described by Hosalkar et al in 2009 (23). Accordingly, odontoid fractures are classified into type I: or fractures which occur before closure of basilar apophysis (before the age of 7) and Type II: which are odontoid fractures that happen after complete closure of basilar synchondrosis (23).

However, acute type I fractures are classified into three subtypes with consideration of the severity, Type Ia, Ib and Ic respectively.

Acute type Ia: is associated with either mild odontoid process displacement or its mild angulation, below 10 degree. The treatment in children type Ia odontoid synchondrosis fractures is conservative where reduction is easily achieved with hyperextension of the neck or traction. This should be followed with immobilization in Minerva cast /jacket or halo vest with high rate of union and success (2, 23, 25, 39-41, 44).

Acute type I b: In this type displacement of the dense with respect to the axis varies from 10 to 100% and its angulation is below 30 degree, Actually, the first line of treatment for this type is also conservative, because they are mostly reducible and usually respond to cranial traction (23,20,28). Reduction by traction should be progressive being started with 8% of body weight with its gradual increase to a maximum of 7 kg.

Close observation and daily sequential lateral cervical radiographs should be obtained in order to monitor reduction and to avoid distraction at the site of fracture according to Mandabach (29).

After optimal alignment, immobilization is a standard method for healing of this sub-type, either in Minerva cast or in a halo-vest (13, 15, 28, 29). Nonetheless, with external immobilization, fusion across the synchondrosis can be obtained in 93% (from 80to100%) of the cases (10, 37).

Because of the capacity of remodeling in children below the age of 3, malunion of the odontoid with respect of the axis and its kyphotic angulation are not problematic but it is regarded a serious sequel of conservative management in older children which deserve appropriate treatment, (3,43) In fact, to date, no data exist to prove the occurrence of spontaneous realignment and remodeling during further growth in older children (3, 43).

Furthermore, it should be added that in type Ia and Ib, a few surgeons has preferred primary direct screwing of odontoid after reduction in order to obviate the necessity of long-term immobilization and possibility of non-union (2,21). It should be added if conservative treatment fails to treat type Ia or Ib and non union occur, secondary posterior fixation will be necessary.

However, in type Ic, which is the most severe subtype of type I odontoid synchondrosis fractures, anterior displacement of odontoid is more than 100%, or angulation is more than 30 degree. In more severe cases, both angulation and displacement are significant. This will eventually result in disruption of posterior ligamentous complex, forward displacement of atlas with its counterpart, the fractured dense and slippage of C1 facet joints, a scenario that is designated as atlantoaxial dislocation.

As a rule, in this subtype, traction should be tried initially to confirm its reducibility, if this fails, the continuation of traction under general anesthesia or manipulation, which are other modes for closed reduction, can be tried (24). Hopefully, dislocation in the majority of the cases with type Ic is reducible.

Eventually, after reduction, with consideration of the associated ligamentous damage, primary surgical intervention with C1-C2 fixation will be mandatory after reduction. C1-C2 stabilization can be best achieved with posterior osteosynthesis and fusion. Various techniques have been described for fixation of reducible atlantoaxial dislocations which varies from C1-C2 wiring technique to transarticular C2-C1 fixation and C1 lateral mass-C2 pedicle screw rod fixation (5, 6, 7, 11, 16, 17, 22, 27, 32).

Subsequently, in an effort to enhance the fusion rate at C1–C2 transarticular C2-C1 screwing and C1 lateral mass C2 pedicle screw of Harms were introduced (5,22). Both these techniques have been used in atlantoaxial dislocation secondary to acute odontoid synchondrosis fracture (5,22). Polyester bands tightened on the short rod by the aid of Universal clamp (Zimmer) which was used in the present case might be proposed as an alternative option for C1-C2 posterior fixation. Eventually, it is very easy technique without the dangers that are hidden in the screwing of C2 and C1 vertebras.

Type II acute odontoid fractures in children: The synchondrosis between the dens and the body of axis normally fuses around the age of seven to 10 years. After closure of the basilar apophysis, pediatric odontoid fractures are designated as type II of Holaskar (23). In this type, pediatric odontoid fractures should be treated in the same manner as they are in adults. Those with minimal displacement should be treated in halo vest and in those with marked displacement, C1-C2 fixation should be done after reduction (34).

Chronic odontoid synchondrosis fractures in children: Chronic odontoid synchondrosis fractures are either due to1) failure of conservative treatment when the initial reduction is lost despite continuation of external immobilization or once the fracture is missed and the diagnosis is made with delay, In both condition the fractured odontoid is displaced significantly or is exhibited with marked angulation and eventually associated with atlantoaxial dislocation. The odontoid might be non-united or mal-united with respect to the axis.

However, surgery of irreducible AAD in children poses a considerable challenge to the treating surgeon because of the patients' immature bone quality, anatomical variability and smaller osseous structures.

Irreducible AAD due to odontoid fractures can be classified in three types, which will ease surgical decision making.

Chronic Subtype I a: Once the fractured odontoid remains non-united to axis and C1 -C2 facets despite slippage are intact (Figure 10A). Treatment in this subtype is reduction following facet release. Actually, the pathology can be reduced via one of the several already described maneuvers after C1-C2 facet releasing. During recent years, our understanding of releasing strategies in irreducible atlantoaxial dislocation has been enhanced by several contributions. Facet releasing plus reduction methods can be done either anteriorly or posteriorly, depending on the experience of the surgeon (9,18,19,26,47).

In a new technique, which has been not described previously, two polyester bands were passed below C2 laminas and two below the arch of atlas. Initially at C2, the sublaminar bands must be tightened on an appropriate 4 cm-length rod by the aid of Universal clamps (Zimmer) on each side. These will act as lever arms and with subsequent tightening of atlas sublaminar tapes on the corresponding rods atlas vertebra will be pulled back resulting in reduction of the dislocation and its fixation simultaneously.

Chronic Subtype I b: The odontoid is mal -fused (mal-united) in displaced angulated position but the facets, despite the slippage are intact (Figure 10B). In this subtype, reduction is not possible without odontoidectomy. Subsequent alignment can be achieved with C1-C2 facet release and reduction via one the previously described maneuvers. Odontoidectomy can be done via different corridors: such as transoral, transoral endoscopic assisted, microsurgical retropharyngeal, transnasal endoscopic and transcervical.

Chronic Subtype Ic: Odontoid process is displaced forward and is mal- fused to axis where the slipped C1-C2 facets are also fractured and engaged (Figure 10C) In this subtype, combination of displaced odontoid process malunited to the axis and C1-C2 facets fractures and fusion coexist. odontoidectomy should be done first. However, because of the engagement or fusion of the facets to each other, facet release is not possible. Therefore, in addition to

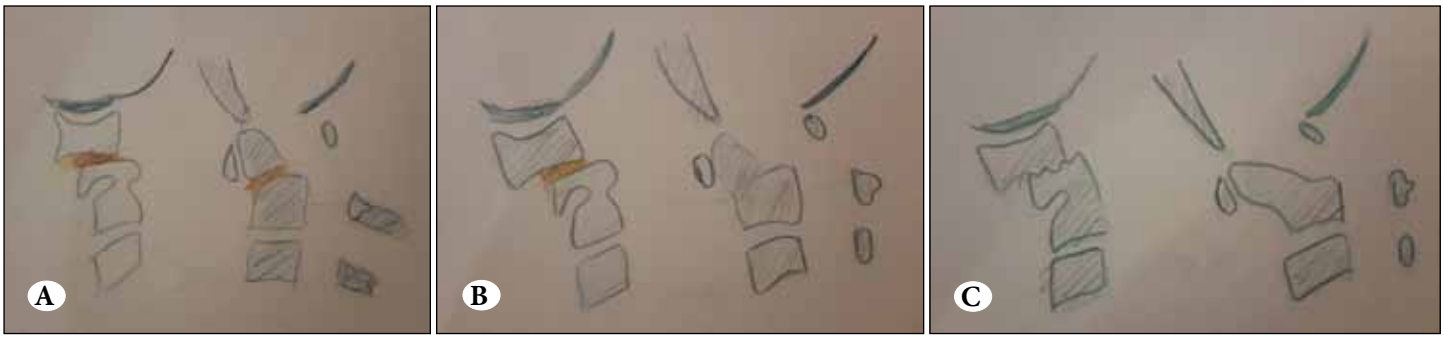

Figure 10: *Schematic drawing of diffrent types of chronic odontoid synchondrosis fracture with irreducible atlantoaxial dislocation.* ***A)*** *Type Ia; the facets are sliped and between the facets scar tissue is seen, but odontoid shows only non-union.* ***B)*** *Type 1b; odontoid is malunited in a displaced position and the facet joints are fixed with scar and granulation tissue.* ***C)*** *Type Ic odontoid is displaced forward and malunited to axis. The facet joints are fractured and are engaged in each other.*

odontoidectomy, occipitocervical fixation plus arthrodesis is the only option. In this type because of constant forward displacement of atlas, the atlas arch laminectomy is mandatory,

Our second patient is clearly chronic subtype Ia, with combination of odontoid non-union fracture and slipped C1-C2 facet joints. Since the formation of the scar tissues within the corresponding facet joints has been the cause of irreducibility, surgical intervention with releasing strategy was indicated. In this case, after atlantoaxial facet release, tightening of sub-laminar polyester band on a rod by a titanium clamp is keyhole of a new system that is called universal clamp system. This instrument has been reported as an alternative for replacing screws and hooks in thoracolumbar region. But to date, it has been not used in cervical spine.

CONCLUSION

Detection of odontoid synchondrosis fractures in recent decades is made more frequently than previously. But, despite of increased awareness, under diagnosed or neglected cases are not infrequent. This is partly due the fact that the clinical feature of the issue is non-specific and partly due to its association with more serious injury such as head trauma which result in unconsciousness for a period of time. Where the treatment is easy and straight forward in those which are diagnosed early, chronic cases pose a great challenge to the surgeon. We hope that the classification described above can be helpful in management of both early and chronic cases. Furthermore, sublaminar polyester band tightened on the rods with the aid of Universal clamp might be employed as an acceptable alternative surgical option, instead of other C1-C2 posterior screw fixation techniques in atlantoaxial instabilities.

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