

ATLANTOAXIAL INSTABILITY IN CHILDREN, 26 YEARS OF EXPERIENCE

Abstract

Study design: Retrospective case study.

Objective: To determine the history, clinical and imaging signs, diagnosis, treatment and clinical follow-up.

Summary of background data: The cranio vertebral junction (CVJ) is a complex transitional area between the skull, the upper cervical spine, the brain and the upper cervical cord. It concerns all ages. Instability can be the result of congenital malformations or traumatic injury.

Methods: It is a retrospective study of patient's files in department of paediatric of neurosurgery and the department of pediatric orthopedic of Marseille, during 26 years , Between 16th October 1981 and 16th June 2007. In each medical record, we determined identity, circumstances of instability discovery, clinical and paraclinical sign , diagnosis, therapeutic protocol and patient outcome after treatment.

The inclusion criteria were the medical records with complete information regarding the patients, who were treated for atlantoaxial instability (AAI). Patients presenting clinical instability with incomplete files, have been excluded.

Results: In our retrospective study, 22 children have been treated for AAI, with 10 boys (45%) and 12 girls (55%), aged 5 to 17 years old , mean age was 10.

the main circumstance of lesion was accident 45,5 % , sport accident 27,3%,road accident 18,2%) ,the others circumstances are 55,5%.

The past history included : congenital malformation in 18,2% (Down's syndrome, Klippel-Fiel's syndrome, cardiac malformation), sport accident(9,0%), neurological deficit (13,7%), cervical trauma already treated(4,5%), learning deficit (4,5%), and plagiocephaly (4,5%); for 45,6%, there was no evidence etiology.

The clinical signs were quadriparesis (31,9%), quadriplegia (13,6%), torticoli (9,1%), quadriplegia and priapism (4,5%), headache and dizziness (4,5%), abnormal head position attitude (4,5%). Clinical exam was normal for 31,9%.

Standard X-ray was performed for 50,0%, CT scan for 50,0% and MRI for 40,8%.

These paraclinical examen short that AAI were traumatic in 59,1% (luxation 36,4%, odontoïdum fracture 13,7%, C1C2 dislocation 4,5 % , C2 arch, articular fracture and pseudoarthrosis 4,5%), congenital malformation in 40,9%.

The orthopedic treatment was used for 13,5% of the patients and surgical treatment was used for 86,5% of the patients (posterior approach 72,8 %, anterior approach 9,2%, anterior and posterior approach 4,5%).

Complications were observed in 33% of the patients (consisted of infections 14%, Medulla oblongata compression 5%, basilar impression)

72% of the patients recovered without sequela after 15 years of follow up, 28% had a neurological deficit during the same time .13% conserved their deficit 5 years after treatment.

Conclusion: In many cases, AAI is an insidious affliction in many cases. A neurological deficit is a sinister presentation often leading to significant sequela even after treatment.

Key-words: Atlanto-axial Instability, treatment, children.

I. INTRODUCTION

The cranio vertebral junction (CVJ) is a complex transitional area between the skull, the upper cervical spine, the brainstem and the upper spine cord. The atlantoaxial region in paediatric patients has several anatomical, biomechanical and physiological characteristics that predispose to injury: increased ligamentous laxicity, more horizontally oriented facets, less mature bone ossification, higher inertia and fulcrum of cervical movement. The relatively underdeveloped neck musculature and the higher inertia and torque forces, associated with a larger head to body mass ratio increase the risk of injury to the CVJ and hence atlantoaxial instability (AAI) (3,6,11,16).

The principal causes of instability in children are:

Malformations (Goldenhar's syndrome, skeletal dysplasia, Conradi's syndrome and Klippel-Feil's syndrome, Diseases such as Down's syndrome have a 14-20% incidence of AAI. In Morquio's syndrome, a combination of odontoid, AAI, and cervicothoracic abnormalities occur in 30-50%. The incidence of atlas assimilation is approximately 0.25 % of the population); Trauma, laminectomy, vertebral infections, rheumatoid arthritis, primary tumors (benign and malignant) and metastases (1,5).

The clinical presentation of AAI may be insidious or rapidly evolving, with, sometimes, false localizing signs. The most common deficits are: monoparesis, hemiparesis, quadriplegia, paraplegia (1, 19, 23, 24).

The primary imaging modalities used for evaluation of the craniotovertebral junction (CVJ) include plain radiography, computer tomography (CT), and magnetic resonance imaging (MRI). It identifies the neural abnormalities as well as the osseous compression

The surgical management of AAI in paediatric patients is associated with certain unique challenges. While the indications for internal fixation in children are similar to those in adults, surgical approaches include posterior approaches, anterior approaches or combining anterior decompression and posterior fusion. Most of the data concerning techniques, complications, and outcomes of spinal instrumentation come from experience with adult patients. Cervical arthrodesis in the paediatric age group might limit growth potential and cause secondary spinal deformity. The development of resorbable instrumentation may reduce long-term risks associated with spinal metallic instrumentation in paediatric patients (1, 6).

II. Material and methods

It is a retrospective study of patient's files in the department of pediatric neurosurgery and the department of pediatric orthopedics Marseille. In each medical record, we determined identity, diagnosis, the circumstances surrounding instability discovery, therapeutic protocol and patient outcome after treatment.

Patients' files with complete information treated for AAI surgically or orthopedically have been accepted. We included orthopedic treatment only if reduction and traction under general anesthesia of the instability.

Patients with C1 C2a1 instability with incomplete files or, treated by traction, reduction and Minerva without anesthesia have been excluded.

Diagnosis is obtained after plain radiography, computer tomography and magnetic resonance imaging reveal on facial and parasagittal views of craniotovertebral junction. That gives craniometry parameters and identifies the different lesions. The different lesions characteristics are described in the Table I, (3, 18).

Table 1 Craniometry of the Craniovertebral Junction

| Line/Angle | Anatomic Landmark | Normal Value | Pathologies |
|------------------------------------|---|---|---|
| Chamberlain's line | Posterior pole of hard palate to opisthion | Dens should not extend more than 5 mm above it | Basilar invagination Basilar impression |
| Wackenheim-clivus baseline | Draw along posterior surface of clivus and extend inferiorly | Lies tangent to the odontoid may transect the odontoid in its posterior one-third | Basilar invagination Basilar impression Atlantoaxial dislocation Atlanto-occipital dislocation |
| Clivus-canal angle | Angle between Wackenheim-clivus baseline and line drawn along posterior surface of the odontoid | 150 (flexion) to 180 degrees (extension) | Spinal cord compression if less than 150 degrees |
| Welcher-basal angle | Intersection of nasion-tuberculum line with tuberculum-basion line | Less than 140 degrees | Increased with platybasia |
| Atlantodental interval | Distance between posterior surface of anterior arch of C1 and anterior surface of dens | 3-5 mm | Widened in atlantoaxial instability |
| Atlanto-occipital joint axis angle | Intersection of lines drawn parallel to the atlanto-occipital joint | 124 to 127 degrees | Increased with occipital condyle hypoplasia |

III. Results

This retrospective review concerns 22 children treated for AAI from October 1981 to June 2007 in paediatric neurosurgery with the collaboration of the paediatric orthopedic service of Marseille Timone children hospital. The study duration is 26 years with 22 patients including 10 boys (45%) and 12 girls (55%), with the age ranging from 5 to 17 years with mean age of 10. One 17-year-old patient was included in this study because he had been followed in paediatric neurosurgery all his life. All patients were treated in paediatric neurosurgery.

The AAI was diagnosed in 27,3% of patients after a sport accident, 18,2% after a road traffic accident, 13,6% incidentally (9,1% after radiological Down's syndrome investigation, 4,5% after a MRI for epilepsy investigation), 13,6% after the apparition of a neurological deficit (quadriparesia, quadriplegia), 9,2% after Abnormal head attitudes .Painless torticollis , cervical pain after excess pressure on the head, hearing strange noises during head movement, Hemiatrophy of right side were described in 4,5% of the patients. Table 2

Table 2 . Circumstances of discovery

| Circumstance of discovery | Number of patient | Frequency |
|---------------------------|-------------------|-----------|
| Road traffic accident | 4 | 18,2% |
| Sports accident | 6 | 27,3% |

| | | |
|---|----|--------|
| Abnormal head attitudes | 2 | 9,2% |
| Incidental discovery | 3 | 13,6% |
| Painless torticollis | 1 | 4,5% |
| Tetraplegia | 2 | 9,2% |
| cervical pain after excess pressure on the head | 1 | 4,5% |
| Cervical pain and tetraplegia | 1 | 4,5% |
| hearing strange noises during head movement | 1 | 4,5% |
| Hemiatrophy of right side | 1 | 4,5% |
| | 22 | 100,0% |

Past history

In the history 45,6% had no particular past history, 18,2% of patients had a malformation (Down's syndrome, Klippel-Feil syndrome, cardiac malformation), 13,7% presented with quadriplegia and encephalopathy, 4,5% had a sports accident, 4,5% had a road traffic accident 4,5% had suffered cervical trauma antecedent, 4,5% scholastic delay, and 4,5% plagiocephaly and strabismus, (table 3)

Table 3 Past history

| Antecedents | | Number of patient | % |
|---|--|-------------------|------|
| Malformations | Down's Syndrom | 2 | 9,2% |
| | Klippel-Feil syndrom | 1 | 4,5% |
| | cardiac malformation | 1 | 4,5% |
| Neurological Deficit | Spastic quadriplegia , Encephalopathy post anoxic seizure | 2 | 9,2% |
| | Spastic quadriplegia, Encephalopathy, odontoid Fracture since 8month with Osteosynthesis in other hospital | 1 | 4,5% |
| accident | Rugby | 1 | 4,5% |
| | Road trafic | 1 | 4,5% |
| Cervical trauma with orthopedic treatment, then trans oral Approach, 8 years before | | 1 | 4,5% |
| Scholastic delay | | 1 | 4,5% |
| Plagiocephaly and strabismus | | 1 | 4,5% |

| | | |
|-----------------|----|--------|
| No past history | 10 | 45,6% |
| Total | 22 | 100,0% |

Clinical signs

The clinical examination of 31,9% of patients was normal, 31,9% of patients presented with quadriparesis, 13,6% quadriplegia, 9,1% torticollis, 4,5% quadriplegia and priapism, 4,5% headache and dizziness and 4,5% head abnormal attitude .(table 4)

Table 4: clinical signs

| CLINICALSIGNS | Number of patient | % |
|-------------------------|-------------------|--------|
| Quadriplegia | 3 | 13,6% |
| Quadriparesis | 7 | 31,9% |
| Quadriplegia +priapisma | 1 | 4,5% |
| Torticollis | 2 | 9,1% |
| Headache and dizziness | 1 | 4,5% |
| Head abnormal attitude | 1 | 4,5% |
| Normal examination | 7 | 31,9% |
| TOTAL | 22 | 100,0% |

Investigation:

This investigation associated standard with flexion- extension neck view. Open mouth craniocervical X-rays were performed to best visualize odontoid lesions.

X-ray was done for 50% of patients and noticed luxation (8,9%), C1C2 malformation (8,9%), and C2 fracture, C2 dish-pan fracture , Odontoid agenesis, C1C2 anterior dislocation, C1C2 dynamic instability, cervicocephalic junction malformation were each noticed for 4,5% of patients. 4,5% of patients had a normal result on x-rays but the diagnosis of instability for one patient had been established with MRI.(table 5)

Table 5: radiography result

| Radiography | Number of patient | Effective |
|----------------|-------------------|-----------|
| No radiography | 11 | 50% |

| | | |
|---------------------------------------|----|------|
| Normal | 1 | 4,6% |
| Odontoid agenesis | 1 | 4,6% |
| C1C2 anterior dislocation | 1 | 4,6% |
| C2 dish-pan fracture | 1 | 4,6% |
| C2 fracture | 1 | 4,6% |
| C1C2 dynamic instability | 1 | 4,6% |
| C1C2 luxation | 2 | 8,9% |
| C1C2 malformation | 2 | 8,9% |
| Cervicocephalic junction malformation | 1 | 4,6% |
| Total | 22 | 100% |

Computer tomography scan: This investigation was performed in 50 % of patients, and identified C1C2 luxation (22,7%), odontoid fracture (9,3%). The bipedicular fracture, C2 dish-pan fracture, atlas malformation, and C1 occipitalisation are each noticed in 4,5% of the patients. All of the CT scans were abnormal. (Table 6)

Table 6: CT scan result

| CT SCAN | Number of patient | % |
|-----------------------------------|-------------------|-------|
| No Scan | 11 | 50,0% |
| dish-pan fracture C2 and luxation | 1 | 4,5% |
| Bipedicular fracture C2 | 1 | 4,5% |
| Odontoid fracture | 2 | 9,3% |
| C1-C2 luxation | 5 | 22,7% |
| Atlas malformation | 1 | 4,5% |
| C1 occipital fusion | 1 | 4,5% |
| TOTAL | 22 | 100% |

Magnetic resonance imaging: MRI was performed in 40,8% of the patients. C1 occipital fusion was described in 13,8% of patients. Congenital luxation, spinal compression, stenosis C0C7, spinal anomaly with C1 occipital fusion, spinal anomaly and foramen magnum stenosis with posterior compression, have each been described in 4,5% of the patients.

Only one person had radiography, CT scan and MRI. (Table 7)

Table7: MRI Result

| MRI RESULT | Number of patient | % |
|--|--------------------------|---------------|
| NO MRI | 13 | 59,2% |
| C1 occipital fusion | 3 | 13,8% |
| Cervical spinal congenital stenosis C0C7 | 1 | 4,5% |
| Spina compression | 1 | 4,5% |
| C2 comminutive fracture | 1 | 4,5% |
| Spinal hypersignal | 1 | 4,5% |
| Congenital luxation c1 C2 | 1 | 4,5% |
| Foraminem magnum stenosis with posterior compression | 1 | 4,5% |
| NORMAL | 0 | 0,0% |
| Total | 22 | 100,0% |

Diagnosis

The AAI in our study is caused by traumatism in 60 % of the patients and 40% by congenital malformation.

Traumatic lesions consist of luxation (36,4%), odontoidum fracture (13,7%) ,C1C2 dislocation(4,5%) and arcus posterior atlantis fracture(4,5%)

Congenital malformations lesion include C1C2 complexes (9,2%) , total C1 Occipital fusion (9,2%), C1 arcus posterior occipital fusion 4,5% sprain associated to C1 spinal bifida (4,5%), Atlas malformation 4,5%), Odontoid dens agenesis (4,5%), Pseudo-arthritis C1C2 and instability of dens (4,5%) (Table 8)

Table 8: Diagnostic

| Diagnostics | | Number of patient | % | %Total |
|--------------------|-------------------|--------------------------|----------|---------------|
| TRAUMATIC | Luxation | 8 | 36 ,4% | |
| | odontoid fracture | 3 | 13,7% | |

| | | | | |
|-------------------------|---|-----------|-------------|-------------|
| | C1C2 dislocation | 1 | 4,5% | 59,1% |
| | Arcus posterior atlantis fracture ,and pseudo-arthrosis | 1 | 4,5% | |
| CONGENITAL MALFORMATION | C1C2 sprain and spina bifida C1 | 1 | 4,5% | 40,9% |
| | C1 occipital fusion | 2 | 9,2% | |
| | C1 C2 complexe malformation | 2 | 9,2% | |
| | C1 arcus posterior Occipital fusion | 1 | 4,5% | |
| | Atlas malformation | 1 | 4,5% | |
| | Odontoid dens agensis | 1 | 4,5% | |
| | Pseudo-arthrosis C1C2 and instability of dens | 1 | 4,5% | |
| TOTAL | | 22 | 100% | 100% |

Treatment

The treatment of AAI was orthopedic in 14% of children and surgical in 86%

Orthopedic treatment:

Children with AAI treated by reduction and cephalic traction with 1/10 of their weight without anesthesia during a week were hospitalized in orthopedic department. All patient treated with reduction under anesthesia were treated in the neurosurgical department.

Among the patients treated orthopedically, 4,5% had C1C2 rotation-luxation, 4,5% presented C1-C2 luxation and 4,5% had odontoid fracture with coma .This orthopedic treatment was completed with surgical collar during 3 months .. The patient with coma was died in reanimation from some others multiples lesions.

Surgical treatment:

Surgery was the main theurapetic approach, 86,5 % of our patient were treated surgically (72,8% posterior approach, 9,2% anterior approach, 4,5% combine anterior and posterior approach) and orthopedic in 13,5%.

Occipito cervical osteosynthesis (22,7%), C1C2 fixation wire (22,7%), arthrodesis and osteosynthesis hook (13,7%) are the main posterior approach. Bipedicular fusion (4,5%) and posterior decompression (9,2 %) are the other posterior technical approaches.

Transoral odontoid fixation (9,2%) was the main anterior approach used. (Table 9)

(Fig 1, Fig2, Fig 3)

Table 9: Technical approach

| Approach | Technical approach | Number of patient | % | Total% |
|--|---|-------------------|--------|--------|
| Posterior approach | Osteosynthesis | | | |
| | -arthrodesis and osteosynthesis hook(fig1) | 3 | 13,7% | 72,8% |
| | - occipito cervical osteosynthesis (fig2) | 5 | 22,7% | |
| | - C1C2fixation WIRE(wire steel)(fig3) | 5 | 22,7% | |
| | - bipedicular fusion | 1 | 4,5% | |
| | Decompression | 2 | 9,2% | |
| Anterior approach | Transoral fixation | 2 | 9,2% | |
| Combined Posterior and anterior approach | occipito cervical Osteosynthesis, ablation, etrier cruschfiel, and transoral decompression and fixation | 1 | 4,5% | 4,5% |
| Orthopedic treatment | odontoid fracture with coma | 1 | 4,5% | 13,5% |
| | C1-C2 luxation | 1 | 4,5% | |
| | C1C2 rotation-luxation | 1 | 4,5% | |
| TOTAL | | 22 | 100,0% | 100,0% |

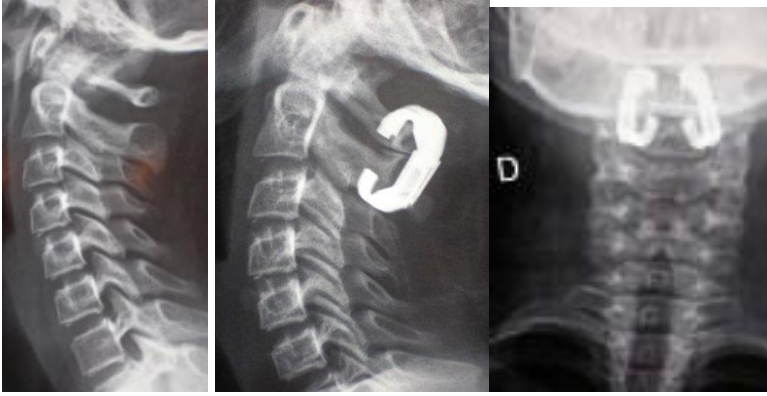


Fig 1 AAI Treated by osteosynthesis hook

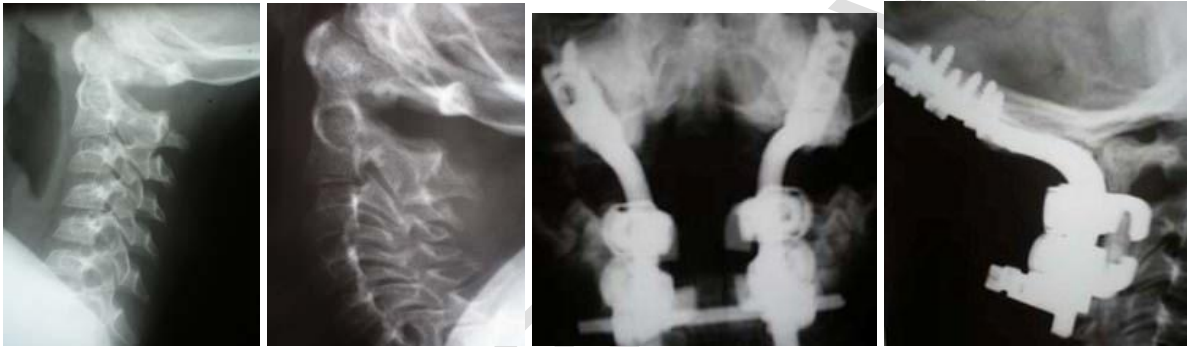


Fig 2 C1 C2 instability with cervico vertebral fixation



Fig 3 C1 C2 instability with C1C2 wire steel fixation

Complications and follow up

Complications were observed in 33% of patients and consisted of infections (14%) including pneumococcal meningitis, lung infections, urinary tract infections and occipital bedsores, 5% of patients had medulla oblongata compression after posterior approach, 5% basilar impression, 5% laterocollis attitude, one patient died as a result of their multiple traumatic lesions.

Treatment of medulla oblongata compression consisted of anterior decompression. Basilar impression was treated by C1 decompression and C2 arthrodesis and jacket halo.

72% of patients had no further complications after 15 years of follow up,

28% had neurological deficits

5% maintained their quadriplegia,

13% maintained their quadriparesis

5% presented with 36 hours of torticollis following on C1C2 luxation and suffered residual hemiparesis with a power of 4/5, 15 years after treatment.

5% suffered from a motor deficiency.

Evaluation of the 26% of patients presenting with quadriplegia at the first examination after 5 years demonstrates a complete regression in half.

IV. Discussion

AAI affects all ages. In our study in children, number of the new case for this illness, the incidence is approximately 0,84%. In some reviews the incidence in children varies from 1 to 10% (11). The anatomical and biomechanical aspects of immature craniovertebral junction, particularly the atlas and axis, are responsible for characteristic spinal patterns in this age group. A disproportionality large head, relatively underdeveloped neck musculature, and shallow occipital-atlas-axis articulation all predispose to AAI (6).

The clinical presentation of neurological deficits can be monoparesis, hemiparesis, paraparesis, and quadriplegia.

At approximately 7 to 8 years old, vertebral form approximates that of adults'. Ligaments and articular capsules become resistant, the articular facets develop vertically and muscles become more developed. Concomitantly in this age, the child becomes more active. This can explain the mean age of our study being 10 years. 45% of patients are 6 to 10 years old. Sport accidents are most common presentation, (27,3%), followed by road traffic accidents (18,2%),

18,2 % of patient had a known congenital malformation syndrome, including: Down's syndrome, mitral congenital insufficiency and Klippel-Feil's Syndrome. The congenital malformation was the principal etiology of the AAI. In some studies, this lesion affects 10 – 20% individuals with Down syndrome (17). The ligamentous laxity associated with atlantoaxial subluxation predisposes to this instability, particularly in Down's syndrome. MENEZES and RYKEN described this mechanism in 15-20% of Down's syndrome patients.

In AAI, the excessive mobility of occipito-atlantoaxial joint may cause repeated trauma of the anterior spinal artery, perforating vessels of the upper spinal cord and the medulla oblongata as well as the vertebral artery. This can lead to spasm or occlusion and attendant neuronal deficit, causing hemiparesis, quadriplegia or paraparesis (1). In our study, the clinical exam was normal for 31,9% of patients. Asymptomatic AAI is described in the literature with a prevalence varying from 14 to 20% in Down's syndrome (17). This incidence cannot be compared with our results, since only Down's syndrome patients were included.

The clinical signs are dominated by quadriplegia (31,9%), paraparesis (13,6%) and the torticollis (9%). The other neurological signs were Head abnormal attitude, Headache and dizziness was described in only one person. The clinical deficit particularly the focalization sign can be the emergency in the C1 C2 instability, sequel may be irreversible or lethal.

Imaging is the main diagnostic exam. In the AAI we studied, standard X-Rays were done for 50% of patients, computer tomography for 50% and the MRI for 40,8%. The standard X-Rays were normal for the 4,5% who presented with odontoid agenesis on CT (Patrick et al); In their study related 9% of odontoid non union diagnosis on CT, had standard normal X-Rays. MRI is the best exam to analyse the musculo-ligamental structures at the spine cord, for bony analysis standard X-Rays and CT scan are more specific and more sensible.

The lesions observed included traumatic lesions (59,5%) and congenital lesions (40,9%). The luxation was described in 36,4% and a C1C2 fracture in 18,5%, C1C2 dislocation in 4,5%; These main lesions of AAI are due to the immaturity and fragility of the muscles, ligament, bones, and joints.

Surgery was the main treatment being performed in 86,6% of patients. 72,8% had a posterior approach. 13,5% received orthopedic treatment. In the Scoll et al study regarding AAI, orthopedic treatment was realized in 67% of the patients. This difference is due to the

restriction of our inclusion criteria to those patient who received reduction under general anesthesia, their study includes all children presenting the C1C2 instability (11).

Surgical treatment was completed with Minerva collar; This maintains the ligament-muscle-skeleton system during the healing period and prevents the cervical cyphosis.

Daniel R. Fassett et al. when studying odontoid synchondrosis in children, initially treated their patients with reduction and external stabilization, with a Halo Vest,.A fusion rate of 93% was found after 3 to 6 months. This technique was very uncomfortable because of this external fixation. Surgery was performed after 6 months if there was nonunion odontoid fracture. In our protocol, surgery is the initial treatment after diagnosis of the odontoid fracture, and our all patients have complete fusion (14).

The follow up was marked by a 33% complication rate (infection, medulla oblongata compression, basilar impression, laterocollis and death). Complication rates were higher when neurologic deficits preexisted before surgical treatment. Only 50% of the patients with quadriplegia had recovered from their neurological deficit at the 5 year follow up. Hence a neurological deficit must be treated as an emergency. Our morbidity rate (30%) after surgery is the same as the Patrick Platzer's et al. although their study included both adults and the elderly. Either in children or in adults, AAI carries with it a high rate of postoperative complications.

V. Conclusion

AAI is rare in children with a prevalence of 0,84% every year in our department. It may occur in a patient presenting after a sports or road traffic accident, or in those with a known congenital malformation (Down syndrome, Klippel-Feil's Syndrome...). Clinical signs are poor but sometimes neurological deficits occur (quadriparesis and quadriplegia). The standard X-Ray, CT Scan and MRI are the main paraclinical investigations. AAI is to traumatic in 59,1% and congenital malformation in 40,9%.The treatment can be surgical with a posterior, anterior ,or combined anterior and posterior surgical approaches. Treatment can also be orthopedic under anesthesia after the failure of reduction without anesthesia. Infections, medulla oblongata compression and laterocollis are the main complications. Quadriparesia and quadriplegia are the most serious complications. Complete recuperation depends on the time delay before initiating treatment and recovery may be incomplete if care is not prompt.

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