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CASE REPORT

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Atlanto-occipital dislocation: four case reports of survival in adults and review of the literature

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Abstract Traumatic atlanto-occipital dislocation (AOD) is a rare cervical spine injury and in most cases fatal. Consequently, relatively few case reports of adult patients surviving this injury appeared in the literature. We retrospectively report four patients who survived AOD injury and were treated at our institution. A young man fell from height and a woman was injured in a traffic accident. Both patients survived the injury but died later in the hospital. The third patient had a motorcycle accident and survived with incomplete paraplegia. The last patient, a man involved in a working accident, survived without neurological deficit of the upper extremities. Rigid posterior fixation and complete reduction of the dislocation were applied in last two cases using Cervifix together with a cancellous bone grafting. Previously reported cases of patients surviving AOD are reviewed, and clinical features and operative stabilisation procedures are discussed.

Keywords Atlanto-occipital dislocation · Cervical spine injury · Posterior cervical fusion

Introduction

Fractures and dislocations of the cranio-cervical junction represent one-third of all injuries to the cervical spine. They are usually caused by high-energy trauma such as traffic accident or fall from a great height [3, 9]. Atlanto-occipital dislocation (AOD), because of its accompanying injuries to the brain stem and lesions to the vascular structures of the neck, is mostly a lethal injury [11, 32, 98]. The diagnosis is often overlooked [74]. Recognition of AOD and therapeutic procedures are hampered by the frequent combination of AOD with traumatic brain injury and/or polytrauma. In this article four adult patients with AOD are presented and two showed a long-term survival and were surgically treated.

Case reports

Case 1

A 22-year-old man fell 12 m from a roof, was unconscious and had a Glasgow Coma Scale (GCS) of three points for an unknown time. Cardiac arrest and apnea occurred after arrival of the rescue team. Following successful resuscitation and intubation, the patient was transferred to the emergency ward of the University Hospital of Zurich without relaxation or sedation. The GCS was unchanged, pupillary light reactions and vital signs were normal. Except for a generalized brain edema in the computed tomography (CT) scan of the head, there were no pathological findings in the clinical and radiological work-up. After insertion of a subdural catheter for intracranial pressure (ICP) monitoring, the patient was transferred to the intensive care unit (ICU). On the first posttraumatic day, clinical neurological examination showed bilateral cranial nerve palsies, absent gag reflex and flabby tetraparesis. Electroencephalogram (EEG) showed a suppression burst pattern and there were no reactions in somatosensory evoked potentials, which was consistent with a high spinal paralysis. Intracranial pressure remained normal all the time. Control CT scan of the head showed a decreased cerebral edema without intracerebral ischaemia or concussions. After an attempt of a wake up, the patient showed a hypoxic aphalic syndrome suspicious of a high cervical spine lesion. Magnetic resonance imaging (MRI) confirmed the diagnosis of an AOD with traumatic lesions of the upper cervical spinal cord and of the lower brain stem. The patient died on tenth day due to severe lesion in the transition from medulla oblongata to spinal cord, verified by autopsy as a severe myelomalacia (total necrosis) in the described anatomic region.

Case 2

A 68-year-old woman was hit by a tram. On arrival of the rescue team, the patient showed a gasping breathing and a GCS of three points with no pupillary light reaction on the left eye. In the emergency ward of our hospital, the patient was orally intubated, sedated, relaxated and developed a haemodynamic instability. After cardiopulmonary resuscitation, clinical and radiological work-up revealed AOD together with a traumatic brain injury and subarachnoidal bleeding. Furthermore, a chest trauma with serial rib fractures, lung contusions on both sides, a pelvic fracture, a floating elbow with an open distal fracture of the humerus, a closed forearm fracture and a fracture of right radius were diagnosed. After immediate insertion of chest tubes, a subdural catheter was placed for ICP monitoring and the left fractured humerus was stabilized by an external fixator. The patient was then transferred to the ICU, where she died the same day due to severe brain and thoracic injuries.

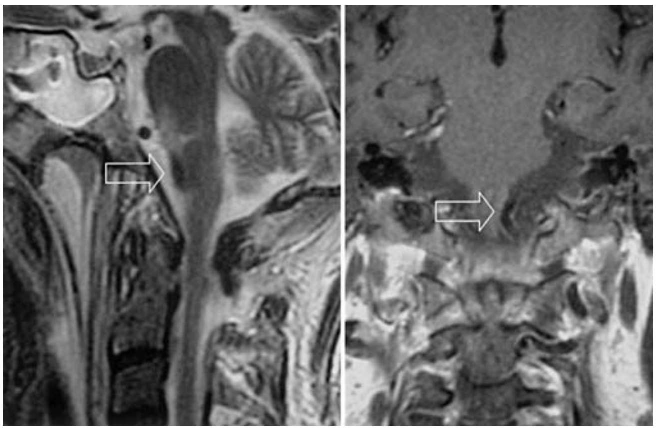
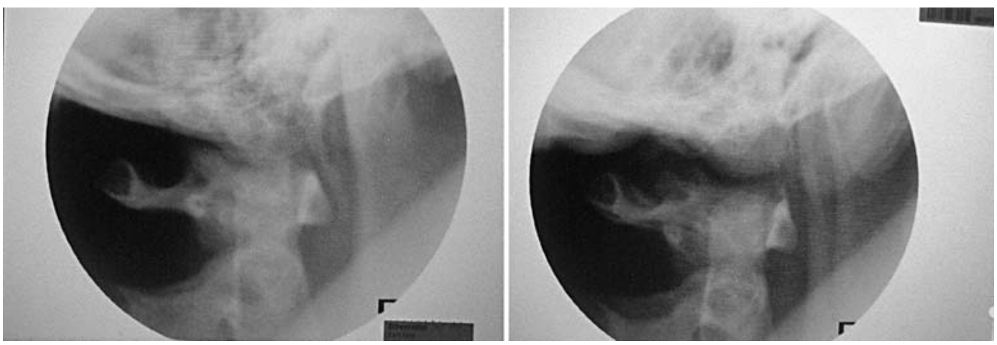


Fig. 1 Sagittal and frontal T2-weighted MRI sequences. Lesion in the pontomedullary junction (*arrows*)

Case 3

A 51-year-old man fell from a motorcycle and collided with a crash barrier. The patient was resuscitated by paramedics until the ambulance arrived. He was orally intubated at the scene showing an GCS of 3 points and no pupillary light reaction on the left eye. Clinical and radiological assessment by conventional X-ray and CT scan of the head revealed a severe brain injury with haemorrhagic concussions on the left temporal side, multiple subarachnoidal haemorrhages, further “shearing injuries” and a ligamentous instability of the right elbow and knee. The CT scan did not show any signs of injuries in the occipito-cervical junction. The ratio according to Powers et al. [73] was less than 0.9. A ventricular catheter for ICP monitoring was inserted and the patient was transferred to ICU. Despite a constantly low intracranial pressure, a failing attempt to wake the patient was a sign of a lesion in the pontomedullary junction. An MRI scan confirmed the diagnosis of an ischaemic lesion in this region (Fig. 1). The injury pattern, the incomplete flabby tetraplegia, the presence of a “locked in” syndrome, bilateral palsy of the *N. abducens* and absent gag reflex indicated AOD. An examination under the image intensifier confirmed the diagnosis (Fig. 2). The neurological findings did not change during hospitalisation. The AOD was stabilized by a dorsal fusion between the occiput and C5 by Cervifix (Stratec, Oberdorf, Switzerland; Fig. 3). Due to patient’s neurologene dysphagia, tracheotomy and endoscopically assisted

Fig. 2 Functional examination under image intensifier demonstrating atlanto-occipital dislocation (AOD)



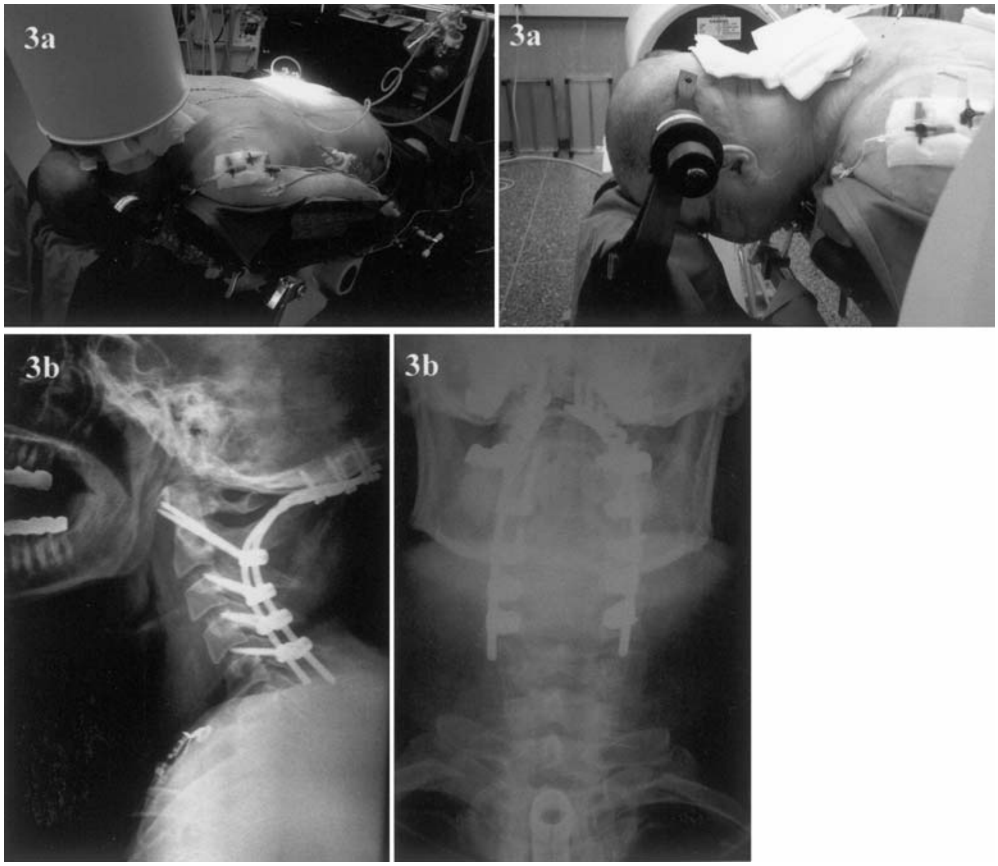


Fig. 3 **a** Positioning of the patient, in prone position, with fixation of the head by a Mayfield clamp. **b** Postoperative X-rays showing stabilization of the AOD by dorsal fusion, from the occiput to C5, with Cervifix and cancellous bone

percutaneous gastroenterostomy (PEG) were performed. Postoperative complications were not observed. Early physiotherapy was applied according to the Bobath scheme. The patient was transferred to other hospital for further neurorehabilitation where he progressively recovered from his neurological deficits. Sixteen months after the accident, he was fully ambulatory without medical aids. The patient, besides an incomplete paraplegia, has a persistent hoarseness and a bilateral palsy of the abducens nerve, for which he has undergone an operative correction.

Case 4

A 35-year-old patient tipped over with a forklift and his shoulder was wedged in by the roof of the engine. At the scene, the initial GCS was 15 points and the patient was able to move his extremities with exception of the right arm. Because of increasing shortness of breath and an inspiratory stridor, he was intubated at the scene by the rescue ambulance and transferred to the University Hospital of Zurich. Clinical work-up revealed a massive swelling at the neck, a subtotal tear of the auricle and a cut at the forearm. Radiological examinations showed fractures of the right clavicle, the left scapula and gave a hint on an injury in the occipito-cervical junction (Fig. 4). The ratio according to Powers et al. [73] was 1.1. The diagnosis of AOD was confirmed by functional X-rays (Fig. 5). During the wake-up, bilateral sensomotor deficiency with palsies of shoulder girdle and the upper extremities were observed. An MRI scan showed a lesion of the vertebral artery and the presence of an epidural haematoma (Fig. 6). The AOD was then stabilised by a dorsal fusion of C0–C5 with a Cervifix (Fig. 7). Postoperative complications were not observed and the neurological deficits of the upper extremities receded. The patient was transferred for further neuro-rehabilitation. Three months after the accident, the patient was fully ambulatory without aids. He still complains of a persistent sensomotor shortage of the right arm at C4–C6. The patient was discharged for ambulatory treatment.

Discussion

A recent study reports an incidence of cervical injuries of 4.3% [39]. The incidence of cervical spine injuries following blunt trauma in children is estimated to be 1.3–1.5% [71, 72]. In 30% of 300 analysed patients with cervical spine injuries the localisation is in the upper region, i.e. between C0 and C2 [9]. Radiological injuries of the upper cervical spine, of which 93% were in the first two

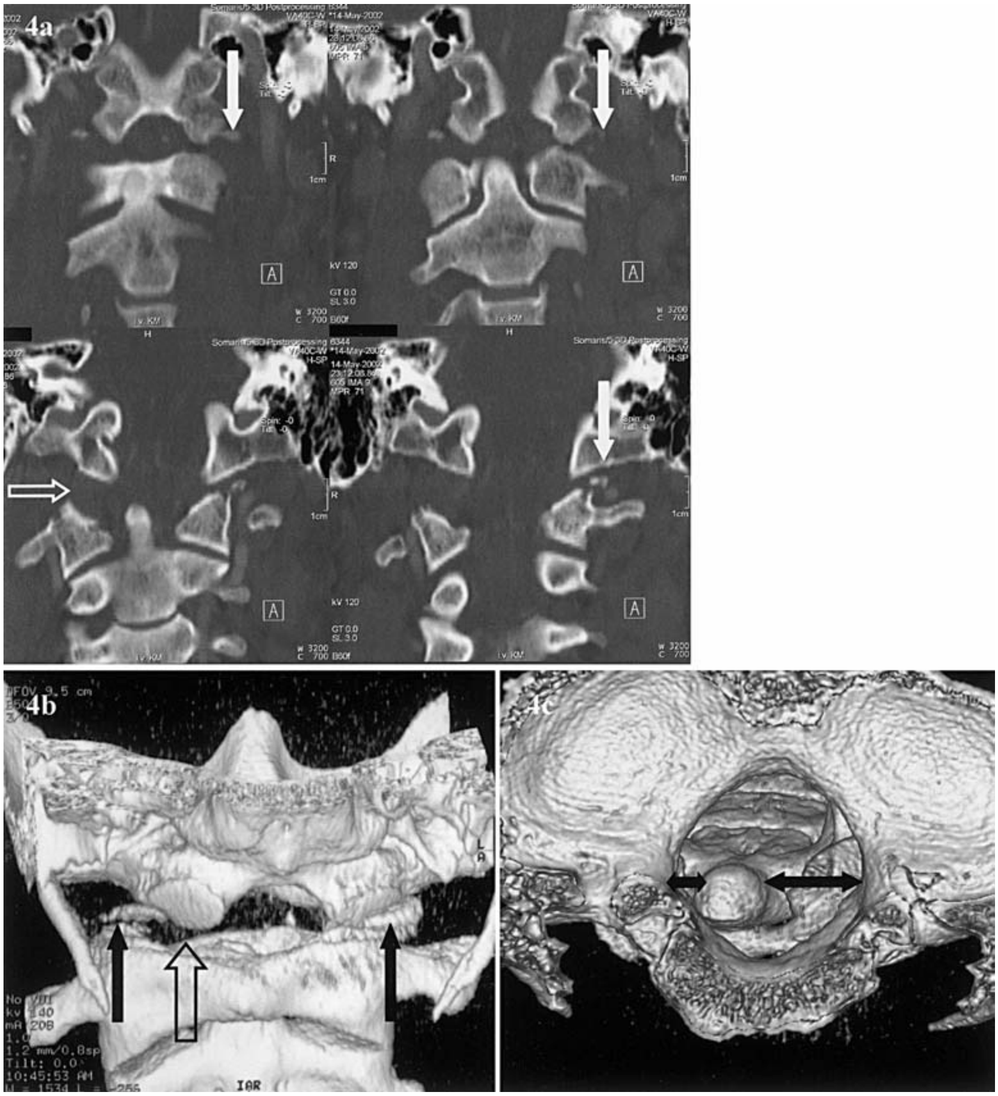


Fig. 4 **a** Atlanto-occipital dissociation in the CT scan demonstrating the subluxation with a rotationary component after first reduction (*open arrow*) and avulsion fracture of the ligaments (*solid arrows*). **b** A 3D reconstruction with an anteroposterior view (*arrows*). **c** Subluxation viewed cranially through the foramen magnum (*arrows*)

segments, were found in 24.4% of 312 deceased traffic victims [3]. Solely ligamentous injuries to the cervical spine were found in only 14 patients of 14,577 with blunt trauma [16]. Injuries to the upper cervical spine are frequent in traffic accidents with the highest prevalence for pedestrians and motorcyclists [86].

Atlanto-occipital dislocation, described for the first time by Blackwood in 1908, is a rare injury [8]. Among 155 examined traffic victims, 12 patients with AOD were found with additional various fractures in the occipito-cervical junction [1]. Others report an incidence of AOD of 8% up to 31% in fatal traffic accidents [11, 98] and of nearly 10% in fatal cervical spine injuries [3]. The AOD may be complete or incomplete. Three different types (ventral, dorsal, axial) of complete AOD were described [88]. The ventral dislocation (type I) is the most common case, the axial dislocation (type II) the most unstable one and the dorsal dislocation (type III) the rarest [42]. In our four cases we found two of type-I (cases 2 and 4) and two of type-II (cases 1 and 3) dislocations. Additionally, lateral dislocations of AOD have been described [90]. Incomplete forms of AOD represent subluxations of the atlanto-occipital junction [42]. The atlanto-occipital, the lateral atlanto-axial and the median atlanto-axial articulations are a functional unit. The overall effect of all joints equals a spherical joint. The rigidity depends mostly on the surrounding ligamentous structures. Among these, the membrana tectoria and the bilateral ligamenta alaria are the main stabilizer [92]. Children and young adults suffer more AOD [11, 24, 64, 79], because of a disparity between the occipital condyles and the articular surfaces of the atlas, a more horizontal plane of the articular surfaces and an increased laxity of the ligamentous structures [12, 45, 52, 80]. Additionally, there are reports that airbag deployment may cause AOD in children [4, 35, 36].

Neurological deficits in up to 33% [32] and vascular complications lead to high mortality. The immediate cause of death may be due to vascular lesions to carotid arteries

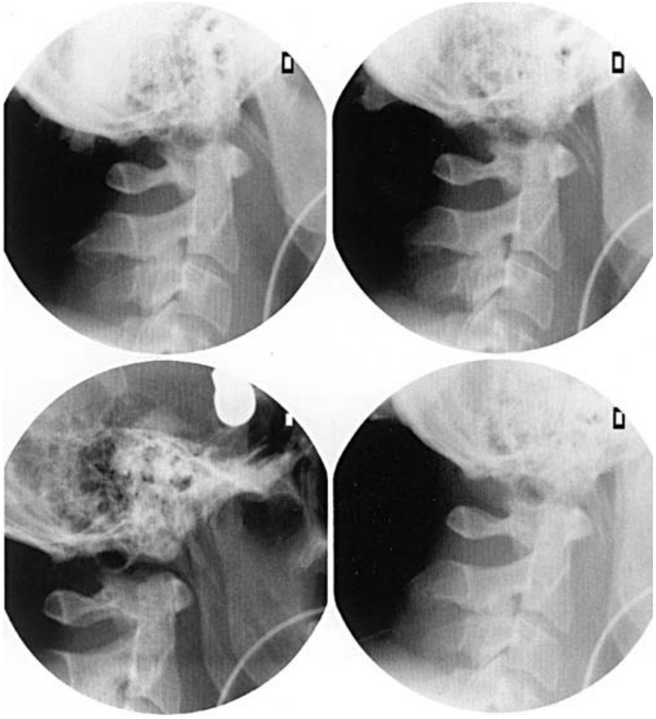


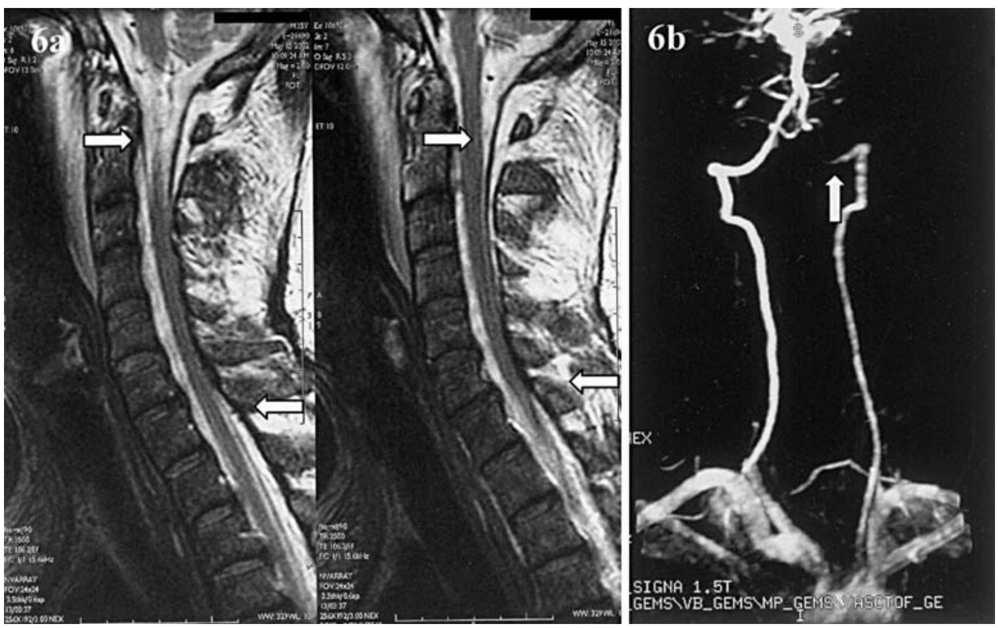
Fig. 5 Functional tests of the atlanto-occipital junction under image intensifier

or vertebral arteries [25, 27, 33, 58, 68, 73] as well as to direct injury to the spinal cord or brain stem [11, 25, 52]. Our patient in case report 4 showed a lack of a vertebral artery due to AOD. Three of our four patients showed signs of direct injuries of the spinal cord or the brain stem. Subdural haematoma as accompanying injury of AOD is described in 16% of cases [1]. A combination with aortic lesions is reported in 25% and with basal skull fractures in 21% [86]. Except our case 4, all patients suffered from severe head injuries. Injuries to the brain nerves, particularly the abducens nerve (N VI) as for example, in our case 4, are occasionally reported in the literature [15, 27, 31, 33]. Out of a group of eight patients with AOD, one patient sustained a rupture of the membrana tectoria with a lethal outcome, and in the remaining seven cases, MRI showed a characteristic stretching with a simultaneous elevation from the clivus and the tip of the dens and a fluid collection [84]. Fourteen of the 79 surviving (18%) patients showed no neurological deficits, and 8 (10%) showed isolated deficits of brain nerves [74].

Absence of neurological deficits has also been reported by other authors [26, 29, 31, 33, 52, 66]. Hemiplegia occurred in 34% and tetraplegia in 38% in patients surviving AOD [74]. Patients with AOD have been reported to show an improvement of neurological deficits [22, 27, 31, 33, 38, 41, 51, 52, 54, 56, 58, 66, 85, 91, 93] and this was also the case with our two patients with long-term survival who both showed an improvement of neurological deficits. Rupture of the trachea [67] and retropharyneal pseudomeningocele [63] were reported as rare complications following AOD.

The majority of patients with an AOD are either immediately dead or survive for few hours only [3, 9, 11]; therefore, this injury was considered potentially lethal [26, 27, 29, 31, 33, 66, 73, 77]. Due to improved pre-hospital care, rapid rescue and shorter transportation time, the number of patients surviving AOD and arriving alive in the hospital is increasing [2, 4, 6, 7, 10, 12, 13, 15, 17, 18, 19, 20, 22, 23, 24, 25, 28, 30, 34, 35, 38, 40, 41, 43, 44, 45, 46, 47, 49, 50, 52, 54, 55, 56, 57, 58, 60, 61, 62, 63,

Fig. 6 **a** An MRI scan of AOD shows an epidural haematoma (*arrows*) with **b** associated lesion of the left vertebral artery (*arrow*)



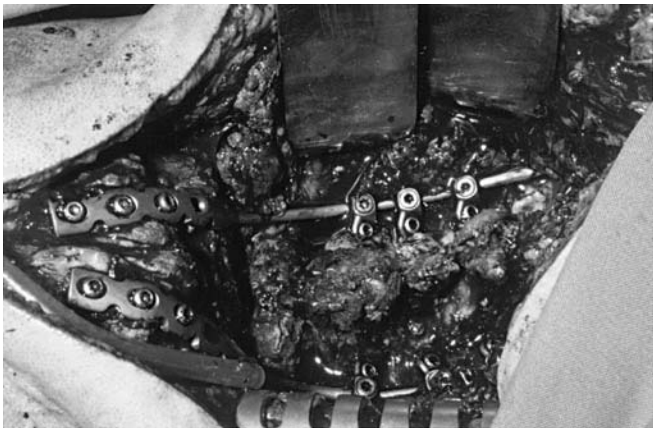


Fig. 7 Dorsal fusion from the occiput to C5 by Cervifix

Table 1 Literature review of adult patients surviving atlanto-occipital dislocation

No. of patients	Reference	No. of cases
1	[3, 5, 6, 7, 8, 9, 14, 19, 26, 33, 40, 43, 47, 48, 49, 50, 51, 55, 62, 63, 66, 67, 73, 76, 78, 79, 85, 87, 90, 91, 93, 95	32
2–5	2, 10, 15, 25, 30, 74	17
>5	23]	7
Total		56

64, 65, 67, 69, 70, 71, 75, 76, 78, 79, 80, 81, 82, 83, 84, 85, 87, 89, 90, 91, 93, 94, 95, 96]. Meta-analysis of the literature between 1948 and present showed 211 patients surviving AOD including 131 (62%) children [2, 4, 9, 12, 13, 15, 17, 18, 20, 22, 23, 24, 25, 27, 28, 29, 30, 31, 34, 35, 38, 41, 44, 45, 46, 47, 52, 54, 56, 60, 61, 62, 64, 65, 68, 69, 71, 73, 74, 75, 77, 78, 79, 80, 81, 82, 83, 84, 88, 89, 93, 94, 96, 97] and 56 (27%) adults (Table 1). For the remaining 24 (11%) cases, the age of the patients was not specified [32, 37, 57].

A complete AOD is accompanied by massive swelling of the head and neck, and radiological diagnosis is, as in our fourth case, usually undemanding. In the presence of a respiratory insufficiency, an irregular cardiac activity up to a cardiac arrest or a hypotension, caused by traumatic lesions of the brain stem, AOD should be suspected [2]. Less prominent dislocations or spontaneously reduced AOD are easily overlooked despite the comprehensive radiological work-up [18, 22, 25, 30, 37, 45, 54, 56]. An analysis of 79 patients with AOD found that in 19 of 50 (38%) children and in 17 of 29 (59%) adults, the diagnosis of an AOD, particularly of an axial dislocation, was missed [74]. We too encountered the same problem in two of our four cases. Diagnostic work-up for AOD consists of a lateral view of the upper cervical spine and the assessment of the ratio according to Powers et al. [73], Harris et al. [42], Dublin et al. [25] or Kaufmann et al. [53]. Functional examination under image intensifier is described [51], combined, if necessary, with monitoring of the somato-sensory potentials [5]. Computer tomography with sagittal, coronary, and three-dimensional reconstruction [21, 40, 60], MRI [13, 14, 21, 37] and angiography [58] are the instruments for the diagnosis and for the assessment of additional injuries in AOD. Avulsion fractures from the occipital condyles or the tip of the dens and a retropharyngeal haematoma may lead to the diagnosis of an AOD [21]. In the CT scan and the MRI in three of our four cases, we found the described radiological signs. Early diagnosis of AOD is mandatory to prevent further impairment of the spinal cord by manipulations during positioning, diagnostic work-up or therapeutic procedures.

After establishing vital functions, particularly of the airways, reposition and splinting are achieved by either a stiff neck, traction in a Gardner–Wells, or a Halo-fixator [7, 27, 29, 33, 56, 62, 67]. For axial dislocation of AOD (type II) traction is not recommended and should be avoided [52]. Complete retention is sometimes impossible due to interposition of fragments or ligamentous structures. Conservative treatment with extension or halo-fixator as further management is recommended in children and in the case of little instability of AOD [2, 28, 49, 56, 63, 73, 83, 84, 93]; however, from the aforementioned literature it is not evident how much instability may be tolerated. In children, a fibrous ankylosis is expected after months of conservative treatment [49, 56, 83, 93]. In adults and with increasing instability, a short dorsal spondylodesis should be performed [15, 34, 68, 82, 93]. The dorsal fusion is achieved by an occipito-cervical tension wire band and augmentation by autologous cancellous bone with an immobilisation in a minerva cast [5, 15, 85]. Fusion by a bone graft from the iliac crest with additional plate fixation (Y-plate, Roy-Camille plate, reconstruction plate) and transpedicular screws allows early mobilization without a cast or with a soft neck for 6 weeks to 4 months [7, 18, 22, 27, 31, 46, 47, 68, 69, 78, 88, 93, 97]. A new technique for stabilization of AOD was described using transarticular fixation with additional occipito-cervical Y-plate fixation [38]. Fusion with the Cervifix system with a bone graft from the iliac crest with or without cancellous bone is an alternative dorsal stabilization procedure [51, 55] and was also applied in two of our four patients. The implant is made from titanium allowing postoperative CT scans with fewer artefacts. The implant design consists of a reconstruction plate in the cranial part and a rod system in the caudal part. An implant removal is not advisable even after complete neurological remission, because mobility from C0 to C2 is lost because of the bony consolidation.

Conclusion

It can be concluded that, due to improved rescue services, decreased transportation time, and increased awareness of the injury, the number of patients surviving AOD increases and that therefore longtime observations are possible [59, 69, 88]. Reviewing the literature, there are 211 documented patients surviving AOD initially and 108 (51%) cases, mainly children and young adults, showed a long-term survival. Diagnosis may be hampered and is frequently delayed, in part due the lack of clinical or radiological evidence, in part due to the additional injuries where a clinical examination is impossible. The extent of neurological deficits in AOD varies from no deficits up to severe neurological deficits caused either by brain stem or spinal cord injuries and by vascular injuries (carotid and vertebral arteries). The therapeutic options range from conservative measures, usually in children, to operative fusion of the cervical spine. The prognosis of primary neurological deficits remains unfavourable; however, there are sporadic reports of neurological improvements after AOD [22, 27, 31, 33, 38, 41, 51, 52, 54, 56, 58, 66, 85, 91, 93].

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