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ATLANTO-AXIAL FUSION IN RHEUMATOID ARTHRITIS

A New Method of Fixation with Wire and Bone Cement

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Twenty-eight occipito-cervical fusions performed over the past 4 years in patients with rheumatoid arthritis are discussed. All of the patients with one exception had signs of neurological involvement preoperatively due to pressure on occipital nerve roots, spinal cord and/or vertebral arteries. A surgical technique using wire, pin and bone cement and permitting early mobilization without external fixation was used and is described in detail. The clinical results were excellent in 21 cases with an additional five patients showing improvement. One patient did not benefit from surgery and one had no symptoms preoperatively. The results are encouraging and the possibility of early mobilization (the day after surgery) is of the utmost importance for this group of patients.

Key words: atlanto-axial luxation; rheumatoid arthritis; bone cement; fusion

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Spontaneous luxation of the atlanto-axial joint is a well-recognized and common complication of rheumatoid arthritis (RA). It is supposed to occur either as a result of loosening of the attachments of the transverse ligament of the atlas or when the odontoid process, shortened by erosion, allows the intact ligament to slip over the tip of the process. The mechanical properties of the joints in this region, the prerequisites for dislocation, and the morbid anatomy of atlanto-axial luxation in RA have been discussed by Werne (1957) and by Ball & Sharp (1971). Cervical luxations of other levels are also quite common in RA (Lidgren et al. 1974) but will not be dealt with in this paper.

The diagnosis of atlanto-axial luxation is based on roentgen examinations. The radiograms must be taken during maximal flexion and maximal extension to investigate the extent of mobility in the joints. The recorded incidence of luxation varies with the selection of patients, the diagnostic criteria, and the radiographic technique. Thus, Conlon et al. (1966) and Mathews (1974) demonstrated luxation roentgenologically in 25 per cent of cases with RA, and this rate is corroborated by our experience. A higher incidence, 37 per cent, was reported by Meikle & Wilkinson (1971)

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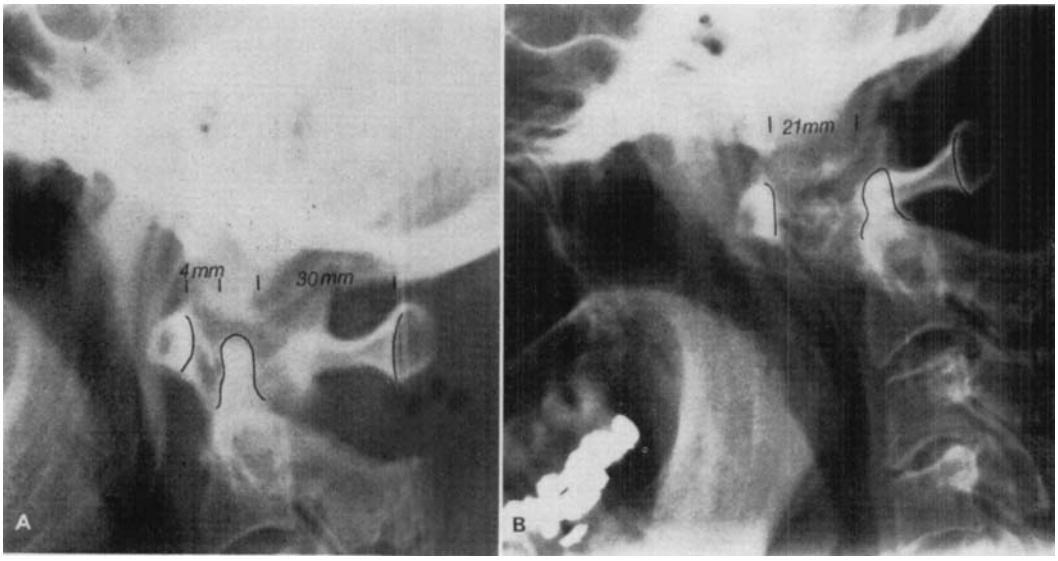

Figure 1. Lateral radiograms of case 26 in maximal extension (A) and flexion (B). The distance between the anterior arch of the atlas and the odontoid process increases from 4–21 mm.

with somewhat wider diagnostic criteria. Martel (1961) found an incidence of 73 per cent in a group of patients complaining of occipital pain.

Three different varieties of atlanto-axial luxation may be found in RA: forward, downward, or backward luxation of the atlas in relation to the axis. Most common is "forward luxation" where skull and atlas move horizontally above the axis. A forward luxation is said to exist if the distance between the odontoid process and the corresponding articular surface of the atlas exceeds 3 mm on the

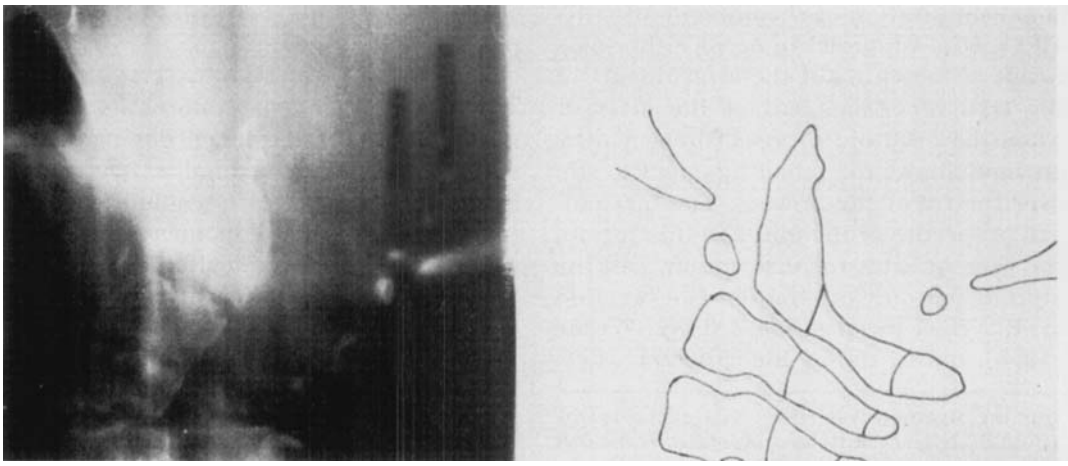

Figure 2. Lateral tomogram of a case with mostly downward luxation, with the tip of the odontoid process protruding into the foramen magnum. (This case was not operated on.)

radiogram during anterior flexion of the head (Figure 1 A and B). Forward luxation is usually unstable and is mostly rapidly and substantially reduced by skull traction.

Less common is "downward luxation" where the anterior arch of the atlas is tilted downwards in front of the axis. The odontoid process then protrudes through the arch of the atlas and the tip of the process usually reaches more than 5 mm above McGregor's (1948) baseline (a line drawn from the upper surface of the posterior edge of the hard palate to the most caudal point of the occipital bone). A preserved odontoid process may then protrude intracranially through the foramen magnum (Brattström et al. 1973) (Figure 2). The posterior arch of the atlas frequently makes a deep indentation in the spinal canal immediately below the foramen magnum. The vertebrae are much less mobile in downward luxation than in forward luxation during flexion-extension of the neck. Reduction during skull traction is insignificant or nonexistent, even if the traction is applied for extended periods of time.

Isdale & Corrigan (1970) have described "backward luxation" in RA but this seems more unusual and such cases are not included in our experience.

Symptomatology

Most patients with atlanto-axial dislocation in RA have no symptoms. However, Smith et al. (1972) found that 30 patients in a series of 150 cases with roentgenologically verified luxation had or developed some symptoms during their period of observation. Rana et al. (1973) found symptoms from the luxation in 37 out of 41 cases. In another series (Stevens et al. 1971) cervical myelopathy was found in two thirds of the cases with atlanto-axial luxation. Mathews (1974) stated that 5 years after diagnosis one third of the patients with forward luxation and half of those with downward luxation had developed long tract symptoms.

The earliest symptoms frequently seem to be caused by a compression of the occipital nerve roots resulting in radiating pain in the occipital region, an occipital rhizopathy. This may precede other symptoms by several years. If the luxation is unstable, the patient may experience crepitations or an unpleasant feeling that the head is slipping backwards or forwards.

Increasing luxation may cause a gradually increasing compression of the spinal cord, resulting in myelopathy and long tract symptoms. This is frequently initially observed by the patient as numbness and weakness in fingers, hands, and arms, and may rapidly progress to pronounced tetraparesis. Severe dislocation may also cause compression of vertebral arteries with signs of brainstem ischemia, such as vertigo or cranial nerve involvement. Spinal cord or vertebral artery compression might ultimately result in respiratory arrest and death (Cohen 1969, Mikulowski et al. 1975, Webb et al. 1968). At present a clear distinction in clinical symptoms between cases with forward or cases with downward luxation of the atlas has not been made.

Therapy

As most patients have no symptoms or only slight symptoms from their atlanto-axial luxation, no therapy is usually required. External support may sometimes reduce or eliminate moderate symptoms. However, in some cases reposition and surgical stabilization may be indicated. In our series, patients were considered eligible for surgery when an atlanto-axial luxation had been verified roentgenologically and when the occipital rhizopathy was unaffected by neck-collar treatment or if collar treatment was not tolerated. Surgery was always chosen if there were

Table 1. Basic data of 28 patients with atlanto-axial luxation treated by occipito-cervical fusion.

Female	23
Male	5
Mean age at operation	59 (27–73) years
Mean duration of disease	17 (5–40) years
Mean duration of occipital rhizopathy	26 (6–80) months
Seropositive	23
Steroid treatment > 1 year	18

signs of spinal cord and/or vertebral artery compression. Only in one patient was instability alone an indication for surgery. In this case movements in the affected joints were excessive during extension-flexion and amounted to 17 mm (case 26 in Table 2).

Choice of surgical procedure

Fusion of the occiput to the upper cervical spine must be the treatment of choice, as it tends to normalize the dislocation anatomically. Various techniques employing bone grafts have been used also in RA (see Discussion). Prolonged immobilization during bed-rest or in traction seems, however, to be a prerequisite for fusion. Extended periods of time in bed are particularly harmful for the RA patient, which is a good reason for using

Table 2. Preoperative symptoms and results of surgery in 28 cases of

Case	Sex	Age (years)	Distance atlas-odontoid proc. (mm) extension	flexion	Downward luxation (see text)	Symptoms. All but one severe neck pain
1	F	47	8	12		Crepitations. "Loose head"
2	F	55	10	15		Hemiparesis
3	F	61	2	12	+	Tetraparesis
4	F	68	2	10		Radiating brachial pain
5	M	61	10	10		Tetraparesis
6	F	64	8	14		Tetraplegic. Bedridden
7	M	54	11	15		Tetraparesis. Vth nerve symptoms
8	M	49	3	10		Tetraparesis
9	F	54	10	10		Tetraparesis
10	F	54	14	20		Tetraparesis
11	F	66	3	6	+	Tetraplegic. Bedridden
12	F	68	5	5	+	Severe pain only
13	F	62	4	11		Tetraparesis
14	F	67	10	10	+	Dizziness. Bedridden
15	F	64	5	5	+	Tetraparesis. Bedridden
16	F	68	0	16		Severe pain only
17	F	48	3	15	+	Tetraparesis. Vth nerve symptoms
18	M	67	3	15		Severe pain only
19	F	63	5	7		Tetraparesis
20	F	59	2	6		Hemiparesis
21	F	69	10	10		Tetraparesis
22	F	66	8	9	+	Radiating brachial pain
23	F	73	7	15	+	Severe pain only
24	F	68	2	13		Hemiparesis. Bedridden
25	F	27	10	10		Crepitations. Psoriasis
26	M	35	4	21		Severe, symptom-free luxation and instability
27	F	67	8	12		Tetraparesis. Vth nerve symptoms
28	F	37	6	9		Radiating brachial pain

Good: painfree; preoperative neurological deficit eliminated or much reduced; no instability on flexion–extension radiograms; the distance atlas–odontoid process the same as preoperatively in extension (± 1 mm).

a technique allowing early mobilization.

This paper reports the experience gained from 28 patients with atlanto-axial luxation, treated with a modification of a surgical procedure initially designed for metastatic disease of the spine. Surgery has been performed as a joint orthopaedic and neurosurgical procedure.

PATIENTS

During the 4 years 1971–1974, 28 patients with atlanto-axial luxation have been treated surgically. The first ten cases were included in a preliminary report (Brattström & Granholm 1973). Patients operated on form a small fraction of a larger number of cases with RA of the cervical spine seen during these years. Twenty-seven of the patients were suffering from severe classical RA whereas one had psoriatic polyarthritis. Some basic data about operated cases are summarized in Table 1.

Preoperative roentgenological and clinical data from the 28 patients operated on are found in Table 2. Instability could be demonstrated roentgenologically in 17 patients, i.e., the distance between the odontoid process and the corresponding articular surface of the atlas changed more than 3 mm between films taken in maximal flexion and in maximal extension of the neck. All patients but one had a severe occipital rhizopathy. Twenty-one patients had signs of spinal cord and/or vertebral artery compression. Several patients experienced a grinding noise in the neck when their heads slid backwards and forwards during neck movements.

rheumatoid arthritis with cervical luxation treated by occipito-cervical fusion.

Observation time (years)	Result	Complications and remarks
4	Good	
3 9/12	Good	
3	Good	
2	Good	Break of wire 1 year postoperatively. Symptom-free.
2	Good	Wound infection, see text.
2	Good	Much improved. Wheelchair bound.
1 9/12	Fair	Wound infection. Cement and wire removed.
1 9/12	Good	
1 9/12	Good	
2	Good	
2	Good	Three stage operation. Wheelchair bound. See text.
1 6/12	Good	Laminectomy of C 1.
1 6/12	Unimproved	Technically unsuccessful. Reoperation after 2 weeks.
1 6/12	Good	Laminectomy of C 1.
4/12	Fair	Laminectomy of C 1. Wheelchair bound. See text.
1 6/12	Good	
1 6/12	Good	
4/12	Fair	Fracture of C 2. Reoperation. See text.
9/12	Good	Fracture of C 2. Reoperation.
6/12	Good	
6/12	Fair	
3/12	Good	
2/12	Fair	Laminectomy of C 1.
2/12	Good	
2/12	Good	Psoriatic arthritis.
2/12	—	Break of wire 1 month postoperatively. See text.
2/12	Good	
1/12	Good	

Fair: less pain, preoperative neurological deficit partly reduced.

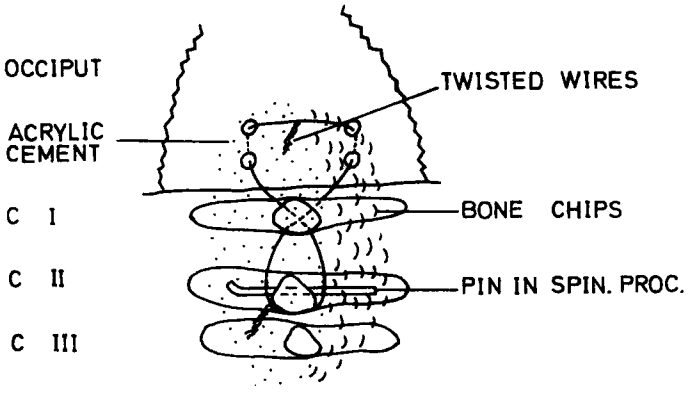

Figure 3 A.

Figure 3. Schematic drawing showing the surgical technique with the application of wire, pin, acrylic cement and bone chips. A: posterior view, B: lateral view, C: cranial view of C 2. See text. The skull and C 1 are pulled dorsally (see arrow in Figure 3 B) when the wire is twisted.

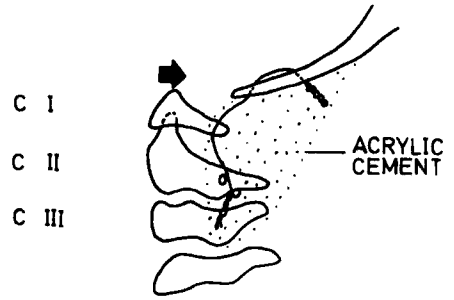

Figure 3 B.

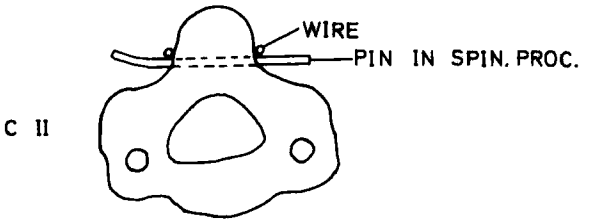

Figure 3 C.

SURGICAL PROCEDURE

Initial experience of the technique prompted a preliminary report on the ten first cases (Brattström & Granholm 1973). Increasing acquaintance with the condition has led to a routine being followed for the treatment of the last 15 cases.

Skull traction

Skull traction is always used. It is started before surgery and is maintained during the procedure. Crutchfield tongs are used and the patient is placed on a Stryker frame. If the radiograms indicate that the luxation is unstable (mainly forward luxation), traction is applied the day before surgery. If the luxation is stable (mainly downward luxation), the patient is kept in traction for a few days before surgery. Any possible reduction of the luxation occurs rapidly, so there seems to be no reason to prolong the period of traction for more than a week before the patient is operated on. If the patient is admitted in acute neurological distress due to spinal cord compression, the first therapeutic measure following roentgenological diagnosis always is skull traction.

Anesthesia

General anesthesia is used and is administered through an endotracheal tube. The tube is introduced with the patient in supine position on the Stryker frame during skull traction. The traction prevents hazardous bending or movements of the neck when the tube is in place, when the patient is turned to the prone position, and during surgery.

Occipito-cervical fusion

From the morning surgery is scheduled, the patient receives Cloxacillin for 10 days, 1 g four times a day. Following intubation, the patient is placed in a prone position on the Stryker frame. With the guidance of previously taken radiograms, the head is moved and tilted in such a position that the atlanto-axial dislocation is reduced as far as possible. One operative field is prepared and draped in the occipito-cervical area and another over the posterior iliac crest, from where bone chips are taken. A straight mid-line incision is made from the external occipital protuberance down to the spinal process of C 4. Muscles and periosteum are removed from the occiput and from the

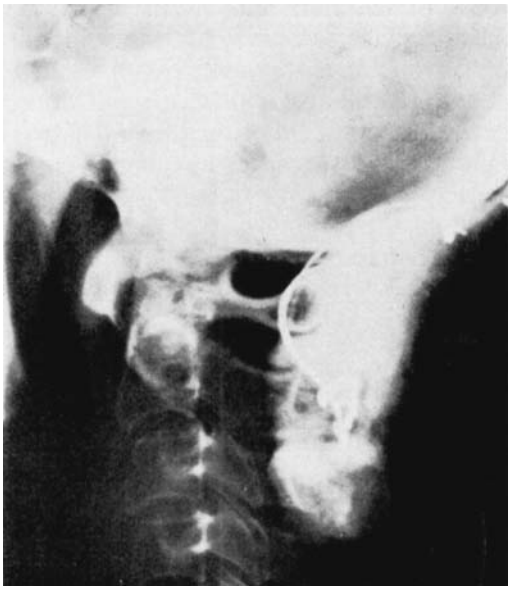

Figure 4. Postoperative radiogram of Case 28.

spinal processes and arches of C 1 and C 2 (occasionally also from those of C 3). The procedure is schematically described in Figures 3 A, B, and C. Four burr holes are placed in the occipital bone and a neurosurgical trephine is suitable for this part of the procedure. Two of the burr holes should be placed as close as possible to the edge of the foramen magnum. The posterior arch of the atlas is dissected free and a nylon ligature is passed between the arch and the dura. A loop of a stainless steel wire is pulled around the arch of the atlas by means of the nylon ligature. The loop is cut, and the two free wires are crossed under the arch. One cranial end of a wire is bent and passed in and out through the left-sided burr holes and the other is bent in the same way through the holes on the right side. The wire ends are then twisted in the midline below the occipital protuberance. If the arch of the atlas is very thin ond osteoporotic, the wires are left un-crossed to spread the mechanical stress over a larger area.

A pin, originally designed to fix a fractured medial malleolus, is passed through the spinal process of C 2, its way through the process being prepared by a clip of a heavy towel clamp. The end of the pin that will later be encased in bone cement is bent slightly to provide a better anchorage to the cement. The function of the pin is twofold: to hold the bone cement in place and to prevent the wire from slipping.

One caudal wire end is now passed dorsally to the pin and twisted with the other wire end, passed in the same way on the other side of the spinal processes. If the spinal process of C 2 is small or very osteoporotic, the wires may be twisted below the spinal process of C 3 with or without a pin through that spinal process. Maximal reduction of the luxation occurs when the wires are twisted. A little more than one half of the exposed bone surface is now covered with bone cement (Orthopaedic Bone Cement Type 1, CMW Laboratories Ltd., Blackpool, England), including two occipital burr holes, the two wire twistings, the spinal processes, and the bent end of the pin. The production of heat in the plastic encasement is counteracted by rinsing with physiologic saline solution. Uncovered bone surface of occiput and laminae is partly decorticated with a dental drill or a chisel and covered with bone chips from the iliac crest. The wounds are closed in separate layers. The patient is then turned to the supine position, whereupon the skull traction tongs and the endotracheal tube are removed. He is returned to the ward in an ordinary patient's bed and is allowed out of bed on the following day in a soft neck collar, which he is supposed to use for 4 weeks (Figure 4).

Downward luxation may need special consideration during surgery. If the luxation is pronounced, the posterior arch of the atlas makes a deep indentation into the dural sac immediately below the foramen magnum. If it is impossible to reduce the luxation, it may then be necessary to perform a laminectomy on the posterior arch of the atlas in order to obtain

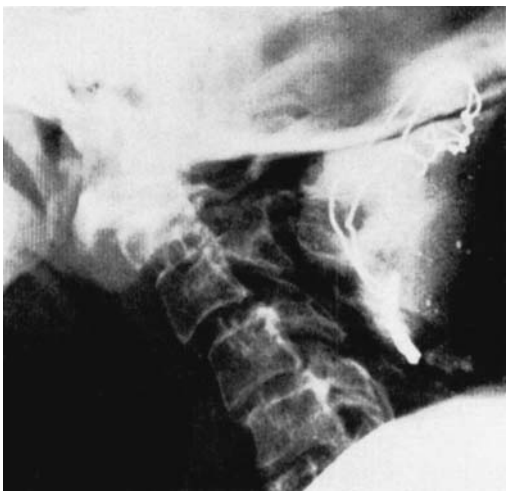

Figure 5. Postoperative radiogram of Case 15. Downward luxation. Laminectomy of C 1. The wire goes from the occiput directly to C 2 and C 3.

decompression of the spinal cord. The wires are then passed from the caudal occipital burr holes straight down to the pin through C 2 and/or C 3 (Figure 5).

RESULTS AND COMPLICATIONS

All patients but one experienced an amelioration of preoperative symptoms and the pain relief was almost complete in 21 cases. Five still have some radiating pain or weakness but describe themselves as improved compared with their preoperative state. Only one patient is completely dissatisfied with the treatment. Particularly gratifying was the neurological improvement in several cases with severe preoperative paresis. The results of surgery are summarized in Table 2.

No deaths occurred at surgery or during the immediate postoperative course. Four patients have died during the follow-up period. Case 5 died 2 years after surgery with bronchiogenic carcinoma. Case 11 was operated on in three stages: 1) transoral removal of odontoid process protruding intracranially through the foramen magnum, 2) after 3 weeks occipito-cervical fusion, 3) after another 2 months anterior fusion at the C 7–D 1 level. She improved and could manage a wheelchair but died 2 years after surgery with chronic urinary tract infection. Case 15 died 4 months after surgery from a pulmonary embolus. Case 18 died suddenly 4 months after surgery from a myocardial infarction with ventricular rupture.

Two cases acquired wound infections, both handled before routine preoperative treatment with antibiotics was instituted. One case was symptom-free from luxation and infection under continuous treatment with Cloxacillin until he died 2 years later from a bronchiogenic carcinoma (Case 5). The fusion material was removed after 1 year in the other patient (Case 7).

The spinal process of C 2 broke a few weeks after surgery in two patients. Both were re-operated and re-fused using a more caudal spinal process or laminae for anchoring the caudal part of the wire (Cases 18 and 19). The wire broke in two cases. In one of these the degree of luxation did not increase, and she has not been re-operated on (Case 4). Surgery is planned for the other case, in whom reluxation has occurred (Case 26).

DISCUSSION

The indications for different types of fusion of the atlas in various diseases have been discussed by Hamblen (1967). Simple instability between the first and second cervical vertebra, such as may occur with fractures of the dens, does not require fusion of the occiput to the upper vertebral spine provided the posterior arch of the atlas is intact. It is then sufficient to fuse the atlas to the vertebral arches below. Fusion of the atlas to the axis and bone cement encasing was used by Alsharif (1969) in two children with atlanto-axial luxation due to RA. RA at the atlanto-axial level is, however, frequently a more widespread disease engaging several joints and the adjacent bone. This necessitates a more extensive procedure, mainly because of the fact that the arch of the atlas usually is thin and osteoporotic ("a little larger than the wishbone of a chicken") (Newman & Sweetnam 1969).

Fusion of the occiput to the upper cervical spine must then be the treatment of choice. This was earlier employed in traumatic injuries by Foerster (1927), who used a fibular graft to stabilize the neck in a case of fracture of the dens. The advantage of iliac bone grafts in the procedure was first recognized by Kahn & Yglesias (1935). There is a large number of reports in the literature from recent years on traumatic injuries treated by occipito-cervical fusion with iliac bone grafts, and also in RA (Crellin et al. 1970, Ferlic et al. 1975, Hamblen 1967, Hauge

1958, Meijers et al. 1974). Prolonged immobilization during bed-rest seems, however, to be a prerequisite for fusion. However, extended periods of time in bed are particularly harmful for the patient with RA, which has been our reason to look for a technique allowing early mobilization.

A suitable technique was initially described for metastatic disease of the spine by Scoville et al. (1967) and consists in wiring of laminae to occiput and acrylic plastic encasement of wires, occiput and spinous processes. The technique employed in the present series of patients is slightly altered. The wires are used also to accomplish maximal reduction of the luxation. It was also felt that it was important to use bone chips to promote bone healing.

One observation from the present series of cases was the difficulty experienced in making an adequate neurological assessment of the patient. Sensory examination is frequently difficult as numerous surgical procedures to joints leave areas around the scar with reduced ability to perceive touch or pin prick. Muscular weakness is caused by medullary or nerve root compression, but is also common in the severely ill RA patient and due to inactivity or stiffness. Reflex activity, finally, may be reduced or absent around severely diseased joints. It was, however, usually possible to separate different qualities of pain from each other. Radiating pain originating from nerve root compression was described as a "new" sensation. In conclusion it is necessary to depend more on the history than on the doubtful value of one neurological examination.

Before this technique was initially employed, it was feared that the patients might be incapacitated by their reduced range of movement in the cervical spine. None has complained of this, however, and it seems as if the reduced range of movement in the fused area has been compensated for by increased movement at lower levels.

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