Clinical Neurology and Neurosurgery 145 (2016) 19–27

Contents lists available at ScienceDirect

Clinical Neurology and Neurosurgery

journal homepage: www.elsevier.com/locate/clineuro

Focusing on the delayed complications of fusing occipital squama to cervical spine for stabilization of congenital atlantoaxial dislocation and basilar invagination

Pravin Salunke[a,*], Sushanta K Sahoo[a], Sudhir Sood[b], Kanchan K Mukherjee[a], Sunil K Gupta[a]

[a] Dept of Neurosurgery, PGIMER, Chandigarh, India
[b] Dept of Neurosurgery, Joshi Hospital, Jullundhar, India

ARTICLE INFO

Article history:
Received 27 January 2016
Received in revised form 21 March 2016
Accepted 1 April 2016
Available online 4 April 2016

Keywords:
Delayed complications
Occipito-cervical fusion
C1-2 joints
Congenital atlantoaxial dislocation
Basilar invagination
Posterior fusion

ABSTRACT

Objectives: Occipito-cervical(OC) fusion is often practiced for congenital atlanto-axial dislocation (CAAD) and basilar invagination (BI) with claims of good long term outcome. Little has been discussed about the delayed complications following fusing occipital squama to cervical spine (OC fusion). We have described and analyzed delayed complications with OC fusion in our patients that helps us understand the underlying dynamics and biomechanics.

Patients and methods: Twenty seven patients of CAAD and BI underwent OC fusion (between 2008 and 11) after transoral odontoidectomy or direct posterior reduction with OC distraction. OC fusion was achieved using either sublaminar wiring or with precurved rods and screws or contoured loop. One patient was referred after OC fusion with contoured loop and wires with additional C1 laminectomy. The outcome (> 12 months) and delayed complications in these patients were analyzed.

Results: Five types of delayed complications were noticed in 6 of the 28 patients who underwent OC fusion. Five of 6 patients were adults. Vertical redislocation with posterior midline fusion (n = 2), adjacent level angular listhesis (n = 1) and swan neck deformity (n = 1) was seen in cases of OC fusion with sublaminar wires alone. Progressive C1 dislocation was seen in the lone patient who was referred after OC fusion with loop and wires. Vertical and angular dislocation was seen in 1 patient of OC fusion with precurved rod and screws.

Conclusion: Progressive redislocation and adjacent level dislocation are delayed complications seen after OC2 fusion. These complications are more often seen in adults, especially with sublaminar wiring/semi rigid OC fixation.

1. Introduction

The treatment for congenital atlantoaxial dislocation (CAAD) and basilar invagination (BI) poses a surgical challenge because of the critical neurovascular structures the region houses. The treatment options vary from removal of ventrally compressing dens with posterior fusion to the direct posterior reduction through manipulation of joints followed by posterior fusion. The myriad techniques available for posterior fusion add to confusion. These patients often have assimilated arch of atlas or bifid C1. With such associated bony anomalies, the usual practice is to fuse the occipital squama to cervical spine (occipito-cervical fusion or OC fusion) either after transoral decompression or direct posterior reduction and a reasonably good long term outcome is claimed [1,2].

The literature describing complications of OC fusion focuses on instrument failure or hardware misplacement causing neural and vascular compromise [3]. In this manuscript we have described a variety of delayed complications of OC fusion in our cases and analyzed the underlying biomechanics. Highlighting these complications would help us understand the dynamics in bony CVJ anomalies better so that management can be rationalized.

* Corresponding author at: Dept of Neurosurgery, PGIMER, Sector 12, Chandigarh, 160012 India.
E-mail addresses: drpravin_salunke@yahoo.co.uk (P. Salunke), drsushantsahoo@gmail.com (S.K. Sahoo), drsudhirsood@gmail.com (S. Sood), kkmneuro@yahoo.in (K.K. Mukherjee), drguptasunil@gmail.com (S.K. Gupta).

http://dx.doi.org/10.1016/j.clineuro.2016.04.001
0303-8467/

2. Material and methods

The Data includes retrospective analysis of 28 patients (10 children and 18 adults) who were followed up at least 12 months after surgery for irreducible CAAD (IrAAD) with Basilar invagination (BI). A proper informed consent was obtained from the patients. Twenty seven patients were operated by authors over a 3 year period (October 2008 to June 2011) and one was operated elsewhere and referred to us. None of them had reduced completely on preoperative cervical traction starting with one twelfth of body weight gradually increasing it to one eight of body weight. All patients (except one operated outside) had assimilated arch of atlas. The long term clinical outcome (using modified Japanese Orthopedic Association Score (JOAS)) and fusion rates have been briefly described. The delayed complications after posterior fusion and their plausible mechanism have been discussed in details.

Nineteen patients underwent transoral odontoidectomy followed by OC fusion (wiring). An artificial arch was created in occipital squama in these cases, under which the sublaminar wire was passed [4]. Three patients underwent opening of C1-2 joints and reduction followed by OC2 fusion using sublaminar wires alone. The use of sublaminar wiring alone was not due to any bony abnormalities that precluded the use of occipital plates or lateral mass screws. It was due to the limited availability of rigid constructs due to high cost and the comfort of using sublaminar wiring in the past [5]. In 5 patients, direct OC distraction was used to achieve intraoperative reduction followed by OC fusion using precurved rods and screws [6]. One case was operated elsewhere and referred to us after surgery for follow up. Partial reduction was achieved with skeletal traction. She underwent occipito-cervical fusion with contoured loop and wires after intraoperative reduction with OC distraction and addition of C1 laminectomy.

In short, OC fusion was carried out in 28 patients including the one referred from outside after surgery. In none of these patients was the C1-2 joints drilled or tackled with use of spacers.

The patients were followed up at regular interval of 3–4 months and assessed for clinical improvement. At follow up, X-ray CVJ (flexion −extension) was obtained. CT CVJ was acquired at 6–8 months after surgery. The patients were assessed every 6–8 months after 12 months of surgery

3. Results

The age of patients varied between 4 and 45 years. Ten patients were less than 16 years of age. The mean preoperative JOAS was 10.2. The mean follow up was 4 years and mean JOAS at last follow up was 12.6. Fusion was seen in 20 out of 28 patients (71.4%). In 3 patients there was redislocation, but fusion was seen in that position (described below in details). Eight patients showed no bony fusion, but the flexion- extension showed no mobility.

The perioperative morbidity and mortality due to transoral surgery has not been discussed. The perioperative complications included cord compression during passage of sublaminar wires (2 patients), CSF leak and meningitis (1 patient). These perioperative complications have not been analyzed or discussed in details as they are beyond the scope of this manuscript.

There were 5 types of delayed complications unique to OC fusion that occurred in 6 patients that were noticed in the long term follow up. The clinic-radiological findings in these patients have been listed in Table 1. They could be divided into ones due to sublaminar wiring and fusion in hyperextension and those secondary to OC fusion in its physiological position.

The delayed complications secondary to OC sublaminar wiring were of vertical redislocation, adjacent level dislocation and malalignment of subaxial spine. These occurred in 4 patients. In one of them, it was noticed when the patient came back 4 years later with progressive worsening of spasticity and breathing difficulty that was precipitated by a trivial trauma a few months ago. In the interim period he never followed up as he had mild residual spasticity. The immediate postoperative CT CVJ had shown adequate decompression following transoral decompression. The CT CVJ at readmission (4 years post op) showed vertical redislocation with intact construct and good posterior bony fusion. He could not be re-operated due to poor respiration and expired within a few days of presentation (Fig. 1A–F). Similar complication was noticed in another young girl during her routine follow up CT CVJ obtained at 1 year. As there was fusion seen in that dislocated position without progression of clinical symptoms, it was decided to follow her up regularly.

One patient showed progressive adjacent C3-4 angular dislocation that was noticed to be increasing at every follow-up. He had undergone OC2-3 fusion with sublaminar wires and had improved. The radiology showed good construct but progressive angular dislocation. Neck pain was the only symptom (Fig. 2A–D). C3-4 fusion was advised in view of progressive dislocation but did not follow up after 3 years of surgery. On telephonic contact, it was confirmed that he had worsened and died in some other hospital. The other complication noticed was swan neck deformity following OC sublaminar wiring for CAAD. This was detected on radiology at one year follow up (Fig. 3A–C). Restricted neck movements were the only symptom and it was decided to follow up the patient regularly.

Long term complications were also noted following OC fusion with contoured loop or plate and screws (occipital and cervical). Both had undergone OC distraction intraoperatively to achieve reduction. In another patient, precurved plate and screws were used after reduction achieved by OC distraction. At 6 month follow up, vertical and angular dislocation of C2 within occiput was seen with an occipital screw pull out (Fig. 4A–I). Reoperation was advised, but refused as he had minimal residual spasticity (JOAS 15). The follow up is over 3 years and he is maintaining the same status. Fusion is seen in the partially dislocated position

In one patient, operated outside and then referred had undergone OC fusion using contoured loop and C1 laminectomy and had improved symptomatically. At 6 month follow up, there was progressive C1 redislocation both antero-posteriorly and vertically but had minimal symptoms. The construct appeared intact but bony fusion was not seen (Fig. 5A-D). The patient was advised surgery but refused. The patient has JOAS of 11, at 1.5 year follow up and bony fusion was still not present. She has not followed up after 1.5 years.

4. Discussion

The treatment for CAAAD and BI was neural decompression with posterior fusion until recent past. With the arch of atlas often assimilated or at times even absent in CAAD, the cervical spine is often fused to the occipital squama [4]. The focus in congenital atlantoaxial dislocation has shifted to C1-2 joints from removal of bony compression and fusion [7,8,]. The recent trend in management of CAAD is towards reduction of dislocation by facet manipulation and fusion of C1-2 joints [7–10]. The C1 is often assimilated with occiput in CAAD and OC fusion is often carried out.

The occipito-cervical fixation has been described in the past and the procedure has evolved over decades. A few decades ago, holes were drilled in occipital squama and tied to C2 [1]. This was modified by creating an artificial C1 arch in occipital squama and wiring it to C2 [1]. Another method, described the use of metallic contoured loop to fix the occiput to cervical spine [2,11]. This was replaced by a more rigid fixation with plates and screws [1,2]. In fact occipito-cervical distraction has been suggested to reduce basilar