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Surgical treatment of adult and pediatric C1/C2 subluxation with intraoperative computed tomography guidance

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Abstract

Background: Surgical treatment of C1/C2 subluxation has evolved significantly over the past 2 decades, from the relatively simpler posterior wiring to more technically demanding instrumentations such as C1 lateral mass screws – C2 pedicle screws, C1/C2 transarticular screws, and occipital cervical fusion. Navigation with fluoroscopy is currently the standard of practice in most centers. However, fluoroscopy at this level carries several major drawbacks, such as blockage by the mandible and inability to produce axial images for assessment of the reduction of rotatory subluxation.

Methods: The authors report a series of 21 patients with C1/C2 subluxation treated surgically with intraoperative computed tomography (ICT) guidance.

Results: There were 7 children and 14 adults. Eight patients underwent C1/C2 fixation with a Harm's construct, and 13 patients underwent occipital cervical fusion. One out of 17 (6%) C1 lateral mass screws has breached the medial wall of lateral mass by 1 mm. Two out of 20 (10%) C2 pedicle screws have breached the foramen transversarium by 1 mm (Neo classification grade 1). The position of all subaxial screws (49 lateral mass screws and 13 pedicle screws) and occipital screws (50 screws) appeared satisfactory. No neurovascular damage occurred in all the patients.

Conclusions: Ninety eight percent of the screws were placed in ideal position with the aid of ICT. Only 2% of the screws deviated from the planned position, but the breaches were not clinically significant and hence no revision was required. This showed that ICT guidance can help to achieve a high accuracy of surgical instrumentation for the treatment of C1/C2 subluxation.

Key Words: Atlanto-axial subluxation, C1/C2, intraoperative computed tomography, navigation, stereotaxy

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INTRODUCTION

Among all conditions of spinal instability requiring surgical treatment, atlanto-axial subluxation is a less commonly encountered entity. However, instrumentation at this anatomical region poses a great technical challenge, owing to the proximity of vital neurovascular structures.[8,10] The margin of error is small and unforgiving. Reported rates of injury to vertebral artery ranged from 1.3% to 4.1% during placement of C1-C2 transarticular screws.[7,19]

Computed tomography (CT) is the gold standard imaging for preoperative planning of surgical approach. The choice of implants, screw length, trajectory, and the method of instrumentation can be made based on CT anatomy.[2] The aim of instrumentation is to achieve maximal construct stability without damaging neurovascular structures. While the planning is routinely done based on CT, execution during surgery is often only routinely guided by recognition of landmarks, fluoroscopy, and surgeon's experience.

Instrumentation based on landmarks alone is subjected to significant inaccuracy even in experienced hands. Fluoroscopy is available in most neurosurgical/spine centers. Nevertheless, it has several limitations when used for instrumentation at the cranio-cervical junction. Fluoroscopy produces 2-dimensional image. Interpretation of screw trajectory requires constant switching between the lateral and anterior–posterior (AP) view. Moreover, the mandible, maxilla, and overlapping of screws significantly degrade the images of C1 and C2 at various angles. The pursuit for improved accuracy has led to the use of intraoperative CT (ICT) and stereotaxy for fixation of the cranio-cervical junction. The purpose of this study is to determine the accuracy of instrumentation with ICT guidance.

MATERIALS AND METHODS

Twenty-one cases of atlanto-axial subluxation with surgical treatment were prospectively recorded from May 2008 to November 2012. All cases were done by single surgeon (RT) and surgical instrumentation in every case was guided by ICT navigation.

Fixation with screws-and-rods construct was performed if both C1 and C2 were suitable for instrumentation. Occipito-cervical (OC) fusion was done for patients with either concurrent atlanto-occipital instability, or if C1 or C2 bone were unhealthy and not amenable to instrumentation. A C2 translaminar screw was inserted instead of C2 pedicle screw if isthmus of C2 pedicle was smaller than 3.5 mm or in the presence of a high riding vertebral artery. All patients in our series were operated in prone position. The head was fixed with a radiolucent Mayfield clamp. CT scan was performed after the patient was appropriately positioned and secured. The scan ensured adequate reduction of the C1/2 subluxation, as well good cranio-cervical alignment in cases where OC fusion would be performed.

The posterior elements of the cervical vertebrae and suboccipital skull were exposed with subperiosteal dissection. A reference array was clamped to a spinous process nearest to the levels to be operated on (usually on C2). An intraoperative CT was then performed, followed by auto-registration. The accuracy of the navigation was verified by placing the pointers on prominent landmarks such as the superior and inferior edges of the spinous process of C2, as well as pins on the reference array.

Entry points of screws were selected based on navigation, and prepared with awl/drill. A reference array was attached to the pedicle probe and registered. The "navigated" pedicle probe was used to find the pedicle track. The position of the probe was tracked by the navigation system throughout the course of insertion. The track was then tapped, followed by insertion of screws based on the same trajectory. For lateral mass screws, the entry points were similarly selected based on navigation. The trajectory was planned using the navigation probe. Drilling, tapping and screws insertion were executed based on the planned trajectory. The site of insertion of the occipital plate was planned with particular attention paid to the anatomy of the dural venous sinuses as they appear on CT. The screw lengths were planned based on the measured thickness of the skull bone, with midline keel fixation being the priority.

Upon completion of instrumentation, a CT scan was done to ascertain satisfactory position of all instruments, as well as excluding an epidural hematoma at the suboccipital area. Revision of screws was done when necessary.

RESULTS

Twenty-one patients with C1/C2 subluxation were included in the study. Age of patients ranged from 6 to 83 years (average 43 years). There were 12 males and 9 females. There were 7 pediatric patients (defined as being under the age of 16 years) and 14 adults. Etiologies of C1/C2 subluxation included congenital, trauma, autoimmune (arthritis), degenerative, infection, and radionecrosis.

Seven patients underwent C1/C2 fixation with a screw-and-rod construct. Thirteen patients underwent occipital cervical fusion. One patient underwent C1 to C5 instrumented fusion (ossification of the posterior longitudinal ligament with concurrent atlanto-axial instability). A total of 153 screws were inserted (103 cervical screws and 50 occipital screws). The type of screws inserted for each levels are shown in Table 1. One out of 17 (6%) C1 lateral mass screws has breached the medial wall of lateral mass by 1 mm. Two out of 20 (10%) C2 pedicle screws have breached the foramen transversarium by 1 mm (Neo classification grade 1).[24] Four translaminar screws were placed in C2. None of the translaminar screws violated the spinal canal. Translaminar screws were placed in cases where C2 pedicles screws could not be inserted (usually limited by pedicle isthmus of less than 3.5 mm). Forty-nine lateral mass screws and 13 pedicle screws were placed in the subaxial spine.

The position of all subaxial screws appeared satisfactory. Table 2 summarizes the details of screw breaches for all the cervical screws. Fifty occipital screws were inserted, out of which 14 has unicortical purchase and 36 has bicortical purchase. Out of the 36 occipital screws with bicortical purchase, 18 did not breach the inner skull table, 9 screws breached the inner skull table by 1 mm, 7 screws by 2 mm, and 2 screws by 3 mm. There was no incidence of extradural or subdural hemorrhage in all patients who underwent occipital screws insertion. Table 3 summarizes the details of screw breaches for all the occipital screws. No neurovascular damage was noted in all the patients. None of the screws needed to be revised. There was no incidence of malfunction of the ICT machine in this series of cases.

Table 1: Summary of the type of operation performed for all patients

Patient No.	Type of operation	Remark
1	Harm's construct	
2	Harm's construct	
3	Harm's construct	
4	Harm's construct	
5	Harm's construct	
6	Harm's construct	
7	Harm's construct	
8	OC fusion	C0-C2-C3
9	OC fusion	C0-C2-C3-C4
10	OC fusion	C0-C1-C2-C3
11	OC fusion	C0-C2-C3
12	OC fusion	C0-C2-C3-C4
13	OC fusion	C0-C3-C4
14	OC fusion	C0-C3-C4-C5
15	OC fusion	C0-C3-C4
16	OC fusion	C0-C3-C4
17	OC fusion	C0-C3-C4-C5
18	OC fusion	C0-C3-C4
19	OC fusion	C0-C3-C4-C5
20	OC fusion	C0-C3-C4-C5-C6
21	C1-C5 posterior fixation	

OC: Occipito-cervical

Table 2: Summary of number of screws with deviation at each cervical level

Cervical screws	Total screws	Deviated screws	Percentage (%)
C1			
LMS	17	1	6
C2			
PS	20	2	10
TLS	4	0	0
C3			
LMS	24	0	0
PS	4	0	0
C4			
LMS	15	0	0
PS	7	0	0
C5			
LMS	8	0	0
PS	2	0	0
C6			
LMS	2	0	0
PS	0	0	0

LMS: Lateral mass screw, TLS: Translaminar screw, PS: Pedicle screw

Example case 1: C1/C2 fixation

A 25-year-old man was involved in a road traffic accident. He was a motorcyclist who skidded and fell from motorbike. He sustained severe head injury requiring intensive care for intracranial pressure (ICP) control. CT of cervical spine showed a rotatory subluxation of C1/C2. He was taken to operating theatre for fixation of the C1/C2 instability after the ICP was under control. As shown in Figure 1, the preoperative CT axial views demonstrated the rotation of C1 relative to C2. The rotatory subluxation could also be appreciated from the sagittal and coronal views. The patient's head was fixed with a radiolucent 3-pin frame. The rotation was reduced under ICT guidance. The reduction was confirmed by ICT in axial, sagittal, and coronal views [Figure 1]. Navigation was especially helpful in guiding the medial angulation of both C1 and C2 screws. Figures 2 and 3 show the screen-shot of the navigation monitor during instrumentation. The trajectories of the screws were super-imposed on the bone images. Navigation led to satisfactory position of all four screws, as shown in Figure 1. Figure 4 is a picture taken intraoperatively after the insertion of screws and rods.

Example case 2: Occipito-cervical fusion

A 9-year-old girl presented with stiff neck and development of right torticollis over 3 months. Examination showed right hypoglossal neuropathy. CT of the cervical spine showed rotatory subluxation of C1/C2. She was treated with halo vest immobilization for 2 months, which resulted in symptom improvement and resolution of the hypoglossal neuropathy. However, the degree of her torticollis worsened after the removal of the halo vest. Repeat CT scan 6 months later [Figure 5] showed a fixed rotatory subluxation of C0/C1 and C1/C2 associated with development of basilar invagination. She underwent OC fusion with occipital plates and screws, C1 lateral mass screw, C2 pedicle screws and C3 lateral mass screws. Reduction was done under ICT guidance. The atlas was partially fused with the occiput, thus preventing a complete reduction of the C0/C1 subluxation. Therefore, a neutral OC position was accepted for fusion. The fixed atlanto-occipital subluxation could be appreciated in the dynamic 3-dimensional CT, as shown in row 2 of Figure 5. A right C1 lateral mass screw was not inserted because of the inability to incorporate this screw along the rod axis due to the fixed C1 rotation. There were a few challenges in this operation. The pedicles of C2 were more narrowed (Right 3.7 mm, Left 3.5 mm) compared with the adult, leaving almost no margin of error in the placement of C2 pedicle screws. Similarly, the occipital bone is also much thinner thus requiring accurate positioning of the occipital keel screws. The availability of ICT navigation significantly increased the accuracy of the instrumentation, and resulted in satisfactory screws placement without damage of the neurovascular structure.

DISCUSSION

Stereotaxy in instrumentation

The advent of stereotaxy is beginning to revolutionize the practice of spinal instrumentation. There is a growing amount of evidences showing that navigated spinal instrumentation is highly accurate.[5,12,25,29,30,32,33,37] The availability of ICT can help to overcome the shortfalls of stereotaxy based on preoperative CT scan. The patient is scanned in the final surgical position, allowing any reduction or intersegmental movement to be updated. This is better than relying on CT imaging obtained preoperatively, which is usually done in a supine position. Intervertebral segmental motion may have occurred when the patient is re-positioned intraoperatively. Auto-registration also obviates the need of time-consuming manual point-to-point registration, thus shortening the set-up time. The use of ICT in surgical treatment of C1-C2 instability is explored in this study.

Table 3: Summary of occipital screws

Occipital screws	Number	Percentage (%)	Cases of EDH or SDH
Unicortical screws	14	28	Nil
Bicortical screws			
No breach	18	36	Nil
1 mm breach	9	18	Nil
2 mm breach	7	14	Nil
3 mm breach	2	4	Nil

EDH: Extradural hematoma, SDH: Subdural hematoma

The restoration of atlantoaxial rotational alignment with intraoperative computed tomography

Restoration of rotational alignment of the C1-C2 junction is important prior to fusion. Spinal alignment is conventionally assessed with fluoroscopy after patient is placed in the final surgical position. Fluoroscopy is usually adequate for the assessment of sagittal and coronal alignment, but its ability to ascertain rotational alignment is limited. In our experience, the assessment of the axis of C1-C2 rotation may not be reliable based on the head position, because a malrotated C1-C2 junction may be compensated by counter-rotation of C0-C1 or the subaxial spine. ICT allows accurate reduction of the rotational angle based on axial images with reconstructed sagittal/coronal views. This is important because failure to fuse the C1-C2 in the neutral axial alignment can lead to permanent torticollis – a significant compromise of function, comfort, and cosmesis.

C1/C2 fixation with ICT

The historical breach rate of C1 lateral mass and C2 pedicle screw ranges from 5% to 21% based on findings of postoperative CT scan.[9,10,18,23,26,36] The difficulty is attributable to the relatively smaller pedicle of C2 and

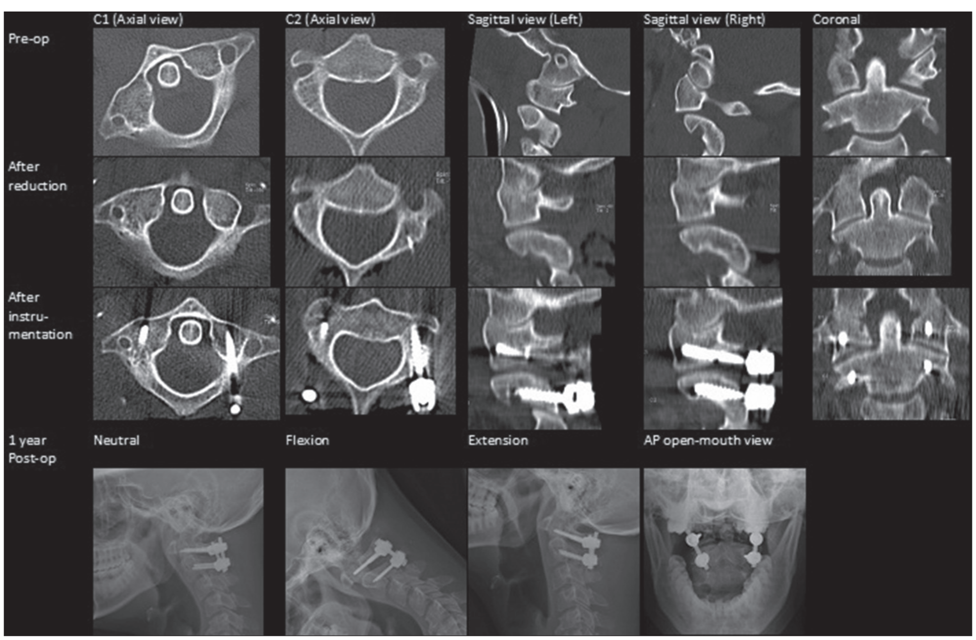

Figure 1: Schematic diagram showing the axial, sagittal, and coronal views of C1 and C2 during the preoperative state, after reduction on the operating table and immediately after instrumentation. The dynamic X-ray 1 year postoperative is also shown

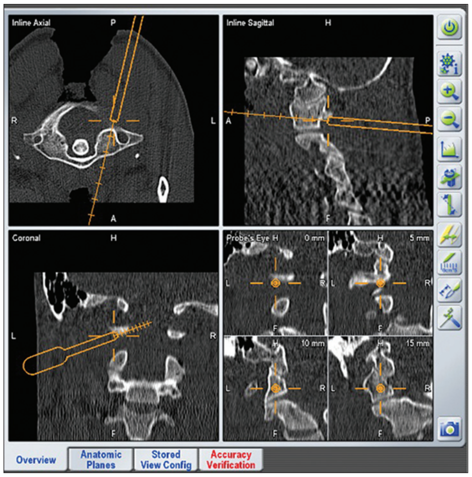

Figure 2: Snapshot of the navigation screen during instrumentation of C1. The axial view was especially helping in guiding the degree of medial angulation of the screw

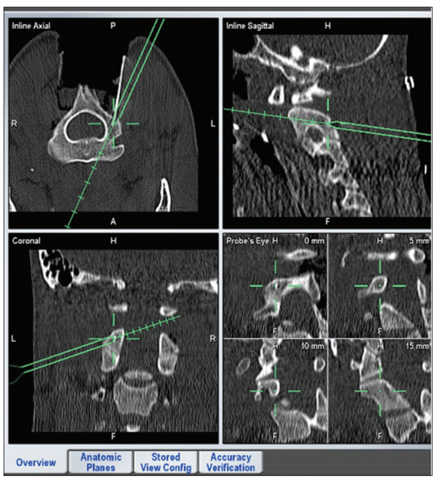

Figure 3: Snapshot of the navigation screen during instrumentation of C2. The axial view was especially helping in guiding the degree of medial angulation of the screw

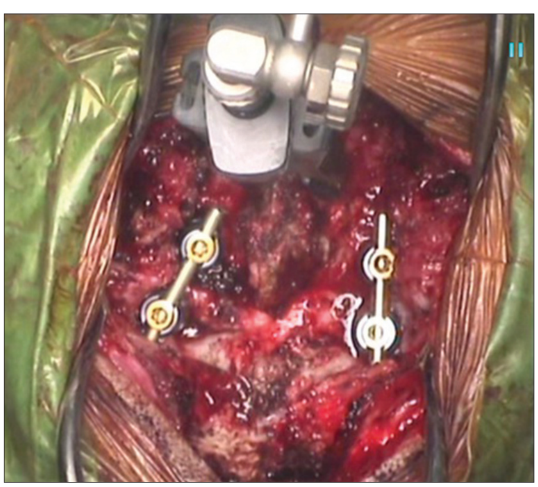

Figure 4: Intraoperative picture taken after completion of instrumentation. The reference array was clamped on to the spinous process of C2

smaller lateral mass of C1, as well as the close relationship of the vertebral artery to the C1-C2 vertebrae. Fluoroscopy was routinely used to ascertain correct entry points of C1 and C2 screws, as well as their sagittal angulations based on the lateral fluoroscopic view. However, the degree of medial angulations of the screws, which was of critical importance to avoid vertebral artery and cord injury, could not be reliably guided by the AP view at this level due to interference from overlapping mandible and teeth. The advent of ICT has circumvented this shortcoming by providing axial CT images during navigation, which clearly shows the position of the foramen transversarium in relation to the pedicle and lateral mass.

When navigated with fluoroscopy, frequent switching between AP and lateral views was required, especially during the critical step of screw insertion. The surgeon could not see both views at the same time. Importantly, a screw that was angulated too medially could only be detected by the AP view of fluoroscopy after it is advanced, and sometimes only after insertion of the entire length of the screw. Neurovascular injury, if any, would have occurred by the time the AP view showed the undesired angulations. If the screw needed to be revised, exchange for a larger rescue screw may not be possible due to limitation of the size of the C1 lateral mass or the C2 pedicle isthmus. Therefore, we found ICT to be superior to fluoroscopy because it allowed navigation based on simultaneous guidance by axial, sagittal, and coronal views. The positions of the screws, or any instruments, were updated in a real-time manner as they were advanced into the vertebrae. In our series, ICT afforded the precision to insert all C1 and C2 screws in a single pass, which technically also translated into greater bony purchase and pullout strength.

Traditionally, a wide exposure of posterior C1, including the lateral mass and sulcus arteriosus, is usually required to delineate the entry point. This process may frequently be hindered by bleeding from the surrounding venous plexuses, and sometimes from the vertebral arteries (VA) at unsuspected locations. We used ICT to define the estimated entry points before exposure of the soft tissue; as such only a limited exposure around the entry point is required. This has resulted in reduced time of exploration, minimal dissection, and reduced blood loss.

A recent study by Yamazaki *et al*. showed a 10% incidence of extraosseous VA anomalies, namely fenestration and persistent intersegmental artery among 100 patients who underwent craniovertebral junction instrumentation.[35] This was significantly more than previously reported incidence of 0.24-0.60%, mainly from studies of patients without cervical vertebral abnormalities.[28,31] In Yamazaki's series, the incidence of VA anomalies approached 37% when there are concurrent craniovertebral junction anomalies, such

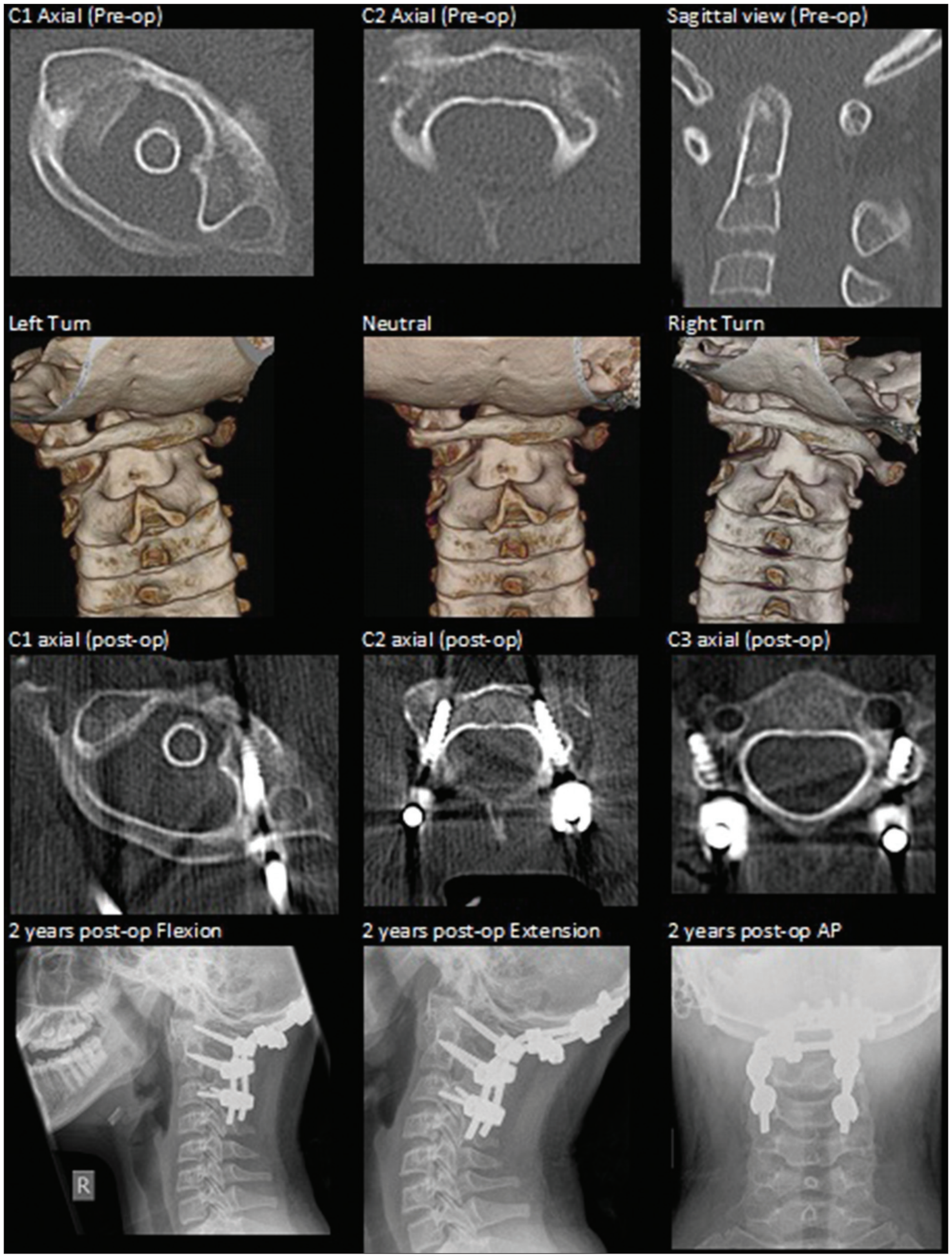

Figure 5: Schematic diagram showing on row 1: Preoperative CT images, row 2: Dynamic 3 dimensional CT, row 3: CT images acquired immediately after instrumentation, and row 4: Dynamic X-rays taken 2 years post-operatively

as os ondontoideum and occipitalization of atlas. The presence of a VA fenestration or persistent intersegmental artery places the VA directly overlying the entry point of a C1 lateral mass screw. In our practice, we used the preoperative magnetic resonance imaging (T2 sequence) or magnetic resonance angiography to locate the position of vertebral artery. In the presence of a VA anomaly, the VA could be explored and mobilized to enable safe insertion of C1 lateral mass screw. A high-riding VA, in contrast, is one that is located more medially, posteriorly, or superiorly. The reported incidence of high riding VA ranged from 10.0% to 22%.[16,20,21] A high riding VA causes narrowing of the C2 isthmus, thus limiting pedicle screws insertion at C2.

Subaxial cervical pedicle screws with ICT guidance

It has been demonstrated that cervical pedicle screws, when compared with lateral mass screws, have a significantly lower rate of loosening at the bone–screw interface, as well as higher strength after fatigue testing.[14] Cervical pedicle screw-rod constructs also demonstrated a greater reduction in axial load transfer through the intervertebral disc than lateral mass screw-rod constructs.[6] However, its use is usually limited by the small pedicle isthmus. Although some cervical pedicles may be wide enough to accommodate the smallest diameter screw available, most surgeons would still avoid inserting pedicle screw because the margin of error is extremely small. Kast, *et al*. reported 30% of cervical pedicle screw misplacement in 26 patients and 94 screws.[15] With improved accuracy, navigation could potentially enable more cervical pedicle screws to be inserted safely, which could result in less number of spinal levels to be instrumented, and reducing the need for second stage anterior fixation.

Role of ICT in occipital plates and screws insertion

The transverse sinus and the torcula mark the superior limit of the occipital fixation. Their location can be accurately determined with ICT, by looking for the posterior edge of the tentorium on sagittal images. We favored occipital plates with a configuration of 2-3 midline screws, and 1 paramedian screw on each side. The midline fixation allows longer screws to be inserted at the thicker midline keel, thus increasing bony purchase. By obtaining good midline keel screw fixation, the OC construct can achieve a better biomechanical strength.[9] The lengths of occipital screws were planned with the preoperative CT scan. The longer screw chosen for the midline has to be inserted exactly at the midline because the skull thickness reduces significantly with even a small deviation from the midline. Excessive protrusion of midline screw risks bleeding from the occipital sinus, which is present in about 38% of patients.[17] ICT navigation has helped to ensure safe insertion of the midline screws. Our series showed that 72% of the occipital screws have bicortical purchase. The maximal tip protrusion was 3 mm. There was no incidence of posterior fossa extradural hemorrhage.

Pediatric cases

Surgical stabilization of the craniovertebral junction in children poses additional challenges compared with adults. Small bony and ligamentous structures, as well as the unusual configuration of vertebral components, and sometimes the presence of congenital malformations have made instrumentation of the craniovertebral junction in children more difficult. In the past, fixations of C1/C2 in children were mainly limited to wiring with a bone graft, because placement of screws at this location was considered too high risk. The fusion rate of wiring is low. Although strong in limiting flexion, wiring has poor strength in limiting extension, rotation and lateral bending. Wineger *et al*. reported in their systemic review of various techniques for occipital cervical fusion, that a screw-rod fixation had the highest fusion rate, with bony fusions occurring in less than 4 months.[34] Therefore, albeit technically demanding, we still prefer fixation with a Harm's construct even in pediatric patients.

Placement of C1 lateral mass screws in pediatric patients is more difficult owing to smaller lateral mass.[13] The benefit of navigation with ICT thus becomes more pronounced in pediatric patients with smaller bony structures. There was no screw deviation in our series of seven pediatric patients, among whom two had C1/C2 fixation and five had OC fusion. Second, a significant number of children with so-called “irreducible basilar invagination” have recently been shown to be reducible under general anesthesia and neuromuscular paralysis. The reduction was demonstrated by intraoperative O-arm (Medtronic) imaging.[22] Following reduction, these children now became candidates for dorsal decompression and stabilization in an anatomically reduced position.

In OC fusion, the thinner occipital bones in children often necessitate multiple screws with bicortical purchase in order to achieve adequate construct rigidity. The recent study of Hwang, *et al*. showed a 20% complication rate arising from occipital screws insertion among 20 children.[11] The complications included vigorous bleeding from dural venous sinuses, cerebrospinal fluid leak, worsening of quadriparesis, wound infection and transient dysphagia. Attempts to achieve bicortical purchase of occipital bone may risk bleeding from the occipital sinus. Hwang concluded that bicortical screw placement in OC constructs might result in a high fusion rate but at the cost of a notable complication rate. The use of ICT navigation is helpful in reducing violation of dural venous sinuses and dural tears. Although the thickness of the midline bony keel could be measured on preoperative CT scan, the thickness usually changes if the entry point is shifted in the cranial, caudal, or lateral direction. Therefore it is hard to predict the screw length on preoperative CT without knowing where exactly the screws are going to be inserted. ICT navigation allows the screw lengths to be measured according to the thickness of skull at the exact point of insertion, thus preventing screw perforation through the dura with possible occipital sinus hemorrhage. With the aid of ICT, we did not experience any significant bleeding from the dural sinuses or any significant extradural hematoma. Our data suggested that up to 2 mm of screw protrusion from the inner skull table was considered safe, as it was not associated with any dura tear or significant sinus bleeding.

Radiation exposure

When compared with fluoroscopy, ICT significantly reduced radiation exposure to operating theatre staffs, including the surgeon.[1] The surgeon's exposure to radiation is virtually zero because he/she is outside the operating theatre during scanning. Nevertheless, the radiation exposure to patient is increased when ICT is used.[3] Using a protocol of 120 kV and 70-130 mAs, our ICT machine produced an average effective dose 5.4 mGy per scan for imaging from occiput to C4. Typically, three scans will be done for each case. One after positioning to ensure adequate reduction, one after exposure and placement of reference clamp (navigation scan) and one scan after instrumentation. Therefore the typical effective dose would be 16.2 mGy per case. In contrast, the highest organ or tissue exposure from fluoroscopy was 6 mGy.[3] Although the radiation exposure to patient has tripled, the absolute increase in lifetime risk of developing cancer remained small. As shown in Bandela's study, the lifetime breast cancer risk, when analyzed based on the BEIR VII report,[4] is 0.055% after exposure to 15 mGy of radiation.[3] Nevertheless, attempts should be made to reduce radiation exposure in pediatric patients. A recent study by Petersen showed that radiation doses for intraoperative 3D CT could be reduced by at least 89% compared with the manufacturer settings while still retaining images clear enough for safe navigation of pedicle screws.[27]

Pitfalls of ICT navigation

The surgeon is responsible for the reliability of the stereotaxy system during the entire course of operation. With even a few millimeter of deviation, stereotaxy may end up misleading the surgeon. In our series, there were no cases where the reference array was displaced accidentally. Verification should always be done after registration. The infrared camera should be placed in a position such that it could detect all the fiducials on the reference array and instruments. Care should be taken not to knock the reference array out of position. If this happen, a CT scan needs to be repeated after repositioning and re-tightening the reference array.

CONCLUSIONS

Instrumentation of C1/C2 instability is challenging owing to the proximity of vital neurovascular structures. Fluoroscopic navigation, which is the current standard practice, has many limitations. In our series, navigation with intraoperative CT has been employed in 21 cases of fixation of atlanto-axial instability. All screws (103 cervical screws and 50 occipital screws) were placed in a single attempt without the need for revision. Ninety eight percent of the screws were shown to be in ideal/planned position on intraoperative CT. Only 2% of the screws deviated from the planned position, but their positions were deemed acceptable and hence not revised intraoperatively. There was no incidence of extradural hemorrhage, cerebrospinal fluid leak or vertebral artery injury. This showed that ICT guidance can help to obtain a high accuracy of surgical instrumentation for the treatment of C1-2 subluxation.

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