Original Article

Surgical management of os odontoideum: An Algerian center experience

ABSTRACT

Background: Os odontoideum (OO) is a craniovertebral junction malformation of unknown origin. In most times, this lesion is highly unstable demanding surgical management. We present our series of OO surgical management and we discuss clinical, radiological, and management aspects of this pathology via our experience and literature opinions.

Methods: This is a retrospective study of patients operated on at our department between May 2014 and May 2021 for OO. All patients were explored with plane X-rays and computed tomography (CT). In some cases, magnetic resonance imaging (MRI) was necessary. Posterior C1–C2 or C1-C3 fixation with polyaxial screws and rod fixation was used. In postoperative, the patient is asked to put Philadelphia collar for 3 months. Hospitalization periods vary between 3 and 7 days. After discharge, all our patients are followed up regularly in consultation. Control radiographs of the occipito cervical region were performed. After 3 months postoperatively, the CT scan is performed on all our patients to assess the quality of fusion. Patient's follow-up ranges from 4 months to 6 years.

Results: Fifteen patients were included in this study; nine males (60%) and six females (40%); with mean age of 32.5 years old. Ten patients (67%) presented motor weakness, three patients (20%) with neck pain, one patient (6.5%) with torticollis, and one patient (6.5%) presented vertigo. No notable cervical trauma was present in six patients (40%) and in nine patients (60%), a remote history of traumatism was noted. All cases of our series presented mobile OO. Normal thickness of the C2 pedicle was noted in nine patients (60%). In two patients (13%), there was hypoplasia of one pedicle and in four patients (27%) both pedicles. MRI showed direct signs of spinal cord aggression: simple compression, myelomalacia, strangulation, or hypotrophy. C1 lateral mass screw fixation was performed in all patients; and according to C2 morphology: nine patients underwent C1-C2 pedicular fixation, in one patient, bilateral crossing C2 laminar screws technique, in three patients, we skipped C2 to perform a C1-C3 articular fixation, and in two patients, C1-C2-C3 fixations were performed. All patients improved clinically. In one patient, we noted an infection resulting in bad wound healing this infection was successfully treated with no complications. In the patient with bilateral crossing C2 laminar screws technique, CT control objectified 4 mm exceeding of one screw; the patient was reoperated and the screw was slightly pulled back. No other complications were noted.

Conclusion: Congenital origin of OO is always evoked. C1-C2 fixation according to Goel and Harms technique with grafting proved its safety, providing high fixation quality with the acceptable biodynamic outcome. Once treated, the prognostic of OO is in general good, and improvement is observed in most patients with few complications.

Keywords: Cranio-vertebral junction instability, Os odontoideum, spinal malformations, spinal traumas

Lakhdar Berchiche[1,2], Adel Khelifa[1,2], Nadjib Asfirane[1,2], Becherki Yakoubi[1,2], Abdelhalim Morsli[1,2]

[1]Department of Medicine, Faculty of Medicine, Algiers University, [2]Department of Neurosurgery, Mohamed Lamine Debaghine University Hospital (BEO), Algiers, Algeria

Address for correspondence: Dr. Adel Khelifa, Department of Neurosurgery, Mohamed Lamine Debaghine University Hospital (BEO), Algiers, Algeria.
E-mail: drkhelifaadel@gmail.com

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INTRODUCTION

Os odontoideum (OO) is a craniovertebral malformation first described in 1886 by Giacomini,[1-5] it is a rare condition[1,2,5] defined by the presence of a free well-corticated ossicle separated from a shortened odontoid process.[1,2,5] Goel *et al.* define OO as a condition where a "significant" or approximately

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at least half of the presumed odontoid process separated from the rest of the odontoid process and the body of C2.[3] This ossicle might be in normal position in union with the anterior arch of atlas, it is called orthotopic and it can be reducible; when the ossicle is fused to clivus it is called dystopic.[1,2,4] The etiology of OO remains unclear and debatable; two theories exist: congenital and traumatic. For supporters of the congenital theory, OO is a failure of axis embryological segmentation[1,2,4] they find it well understandable that the vestigial intervertebral disc separating the odontoid from the body of C2 is responsible for the formation of OO; then they base on familial observations, identical twins case reports, and some studies on chromosomic aberrations.[1,4] For the supporters of the traumatic theory OO is an unconsolidated Anderson and Alonzo Type II odontoid fracture; in fact, a history of cervical trauma is found in an important part of OO patients.[1,4] For Goel *et al.* based on an observation of 37 cases of the bifid posterior arch of atlas among 190 cases of OO; they came up with the theory that both OO and bifid posterior arch of atlas are natural bone transformations to protect the cord from compression between bones in patients with preexisted atlantoaxial instability.[3] OO could be totally asymptomatic and diagnosed with systematic neck imaging during cervical trauma;[1-5] other presentations include: neck pain, and/or motor deficit due to spinal cord compression in the craniovertebral junction. Intracranial manifestations were reported and are mainly the consequence of vertebrobasilar ischemia, especially gait disturbance[6,7] and consciousness disturbance on neck extension.[8]

METHODS

Patient selection

This is a retrospective study of patients operated at our department from May 2014 to May 2021 for OO. Clinical charts and radiographic data were reviewed in detail. Patients presenting complex cranio-vertebral junction malformation, in most cases managed by occiputo-cervical fixation were excluded.

Radiographic evaluation

Initially, all patients were explored with plane X-rays; in some cases, the diagnosis of OO was directly retained in absence of direct violent cervical trauma with visualization of the free odontoid process; but in most cases, it was evoked in the case of spinolaminar line rupture, an indirect sign of atlantoaxial dislocation. In all cases, occiputo-cervical junction computed tomography (CT) was performed. First CT is used to establish the diagnosis of OO, defining it as free or attached to clivus, and finally to plane surgical strategy based on C2 morphology. Dynamic X-rays are also performed to study atlas mobility. In some cases, magnetic resonance imaging (MRI) was performed.

Operative technique

All patients are put under transcranial traction using Gardner-Wells tongs. Posterior C1–C2 or C1-C3 fusion with polyaxial screw and rod fixation was used. Patients were operated under general anesthesia and orotracheal intubation in prone position under lateral fluoroscopy control. Classic midline posterior cervical spine approach was performed; exposing the occiput, C1 posterior arch and lateral masses, C2 isthmus, facet joints of C3 and C4. We locate the C1 entry point under the inferior arch of C1, on its root on C1 lateral mass, and in the sagittal plane passing by the middle of the C1-C2 facet joint. The C2 entry point was for pedicle fixation or rarely for lamellar fixation. To define the C2 pedicle screw entry point, we divide virtually the C2 isthmus on four quadrants; the entry point is located in the superior medial quadrant. The C2 lamellar entry point is located in the middle of the spinolaminar junction. To define the C3 lateral mass entry point, we divide it virtually into four quadrants, the entry point is located in the inferior medial quadrant. Once the entry points were defined, we first use an automatic drill to decorticate the entry point, then using 24 mm manual drill we prepare the screw hole. The drilling trajectory on C1 is 15° up and 15° medial starting from the entry point and aiming the anterior C1 tuberosity on lateral fluoroscopy. The C2 pedicle drilling trajectory is 25° up and 25° medial starting from the entry point and staying strictly inside the pedicle toward the C2 body; this can be checked with a spatula introduced in the spinal canal with which we expose the medial and the upper borders of C2 pedicle. The C2 lamellar drilling is done under direct visualization of both laminae. The C3 drilling trajectory is 25° up and 25° lateral aiming the virtual superolateral quadrant. Then polyaxial screws of 3.5 mm diameters are used. On C1 screws are partially threaded of 28–32 mm length estimated case by case preoperatively. On C2 screws are totally threaded of 18–24 mm length; and on C3, they are totally threaded of 12–16 mm length. Once all screws are placed rods are cut, set on screws canal, and blocked. Then, an autograft is harvested from the iliac crest and fixed between the C1 posterior arch and C2 laminae and spinous process. All osseous contacts are decorticated in manner to leave only cancellous bone between C1 and the graft then between the graft and C2. Finally, the graft is fixed with classic wiring techniques. The wound is closed as usual.

Follow-up and data collection

In postoperative the patient is kept with Philadelphia collar for 3 months. After that, spine CT is performed to confirm graft fusion, and then, we free the neck from the collar. All patients are regularly examined in our external consultation and periodic plane X-rays are performed. Patients' follow-up range from 5 months to 6 years. All possible complications were searched for at least 4 months postoperatively. Patients with motor weakness were sent to rehabilitation.

RESULTS

Patients' demographics and presentations

Fifteen patients were included in this study; nine males (60%) and six females (40%); with main age of 32.5 years old. Ten patients presented motor weakness (67%), three patients with neck pain (20%), one patient with torticollis (6.5%), and one patient presented vertigo (6.5%). No notable cervical trauma was present in six patients (40%) and in nine patients a remote history of traumatism was noted (60%). This trauma is with a delay of approximately 9 years before diagnosis [Table 1].

Radiographic findings

All cases of this series presented mobile OO. Normal thickness of the C2 pedicle was noted in nine patients (60%); we considered a pedicle of more than 4 mm, a normal size and screwable pedicle. In two patients (13%) there was hypoplasia of one pedicle and in four patients (27%) both pedicles. MRI showed in all cases direct signs of spinal cord aggression: simple compression, myelomalacia, strangulation, or hypotrophy.

Surgical management

Fourteen patients were operated on the first time; and one case was managed previously in another center with simple graft and wiring; after material failure, the patient was sent to our center. C1 lateral mass screw fixation was performed in all patients; and according to C2 morphology: in nine patients, C1-C2 pedicular fixation was performed; in one patient bilateral crossing C2 laminar screws technique; in three patients, we skipped C2 to perform a C1-C3 lateral masses fixation, and in two patients C1-C2-C3 fixations were performed [Table 1].

Patients' outcomes

All patients improved clinically. In one patient, we noted an infection resulting in bad wound healing this infection was successfully treated with no complications. In the patient with bilateral crossing C2 laminar screws technique, CT control objectified 4 mm exceeding in one screw; the patient was reoperated and the screw was slightly pulled back. No other complications were noted [Table 1].

DISCUSSION

A male predominance (sex ratio of 1.5) with main age of 32.5 years old are calculated in our series; we explain these two observations that males in young adult age are more exposed to cervical trauma; this trauma could be remote, explaining the formation of OO itself, or recent which is frequently pushing the patient to consult after the apparition of complaints in patients with preexisting OO tolerated until the trauma happened. Although most of our patients presented the remote history of cervical trauma ranging from 9 months to 16 years; 40% of patients are without noticeable accidents; such observation is more in favor of the congenital theory. Clinical presentations in our series were dominated by progressive motor weakness in four limbs with exaggerated reflexes; signing chronic compression or repetitive aggression of the spinal cord. In other patients-although highly instable-OO was well tolerated and clinical presentations were limited to neck pain, vertigo, and one case of torticollis. Plane radiography is very useful knowing that OO is frequently diagnosed in the emergency room with simple X-rays performed after cervical traumas. Initially, two views are demanded: lateral and face-open

Table 1: Summarizing presented cases in our series of os odontoideum

Cases	Sex	Age	Clinics	Trauma	Delay	Techniques	Complications	Evolution
1	Male	47	Vertigo	No	-	C1-C2	None	Improvement
2	Female	16	Torticollis	Yes	11 years	C1-C2	None	Improvement
3	Female	14	Weakness	Yes	9 months	C1-C2	None	Improvement
4	Female	17	Neck pain	Yes	6 years	C1-C2-C3	None	Improvement
5	Male	47	Weakness	Yes	11 years	C1-C2	None	Improvement
6	Male	54	Weakness	No	-	C1-C2	None	Improvement
7	Female	25	Weakness	No	-	C1-C2 (bilateral, crossing C2 laminar screws)	Reoperated	Improvement
8	Male	12	Weakness	Yes	9 years	C1-C2	None	Improvement
9	Male	40	Neck pain	Yes	14 years	C1-C2-C3	None	Improvement
10	Female	62	Weakness	No	-	C1-C3	None	Improvement
11	Male	17	Weakness	Yes	11 months	C1-C3	None	Improvement
12	Female	11	Weakness	No	-	C1-C2	None	Improvement
13	Male	45	Weakness	Yes	16 years	C1-C3	None	Improvement
14	Male	56	Weakness	No	-	C1-C2	Wound infection	Improvement
15	Male	26	Neck pain	Yes	10 years	C1-C2	None	Improvement

mouth view. In absence of violent trauma, a free ossicle at the top of the axis is an indisputable sign of OO [Figures 1-4]. In other cases, the anterior band of spino-laminar virtual line at the level of C1-C2 [Figure 1] signs an atlantoaxial dislocation, and OO is evoked among other etiologies of craniovertebral junction instabilities. CT is our study of choice because of its accessibility and superiority in the visualization of bone structures; it is first used to diagnosis, it shows the well-corticated ossicle above the shortened odontoid process on orthotopic OO or fused to the clivus in dystopic OO. Second, CT is used to define whether this ossicle is free or attached to clivus. Finally, to plane surgical strategy CT is used to study vertebrae morphology especially atlas lateral masses size and axis pedicles thickness. MRI is helpful to assess the amount of stenosis in the cranio-vertebral junction and its repercussion on the spinal cord. Depending on the degrees of aggression, anomalies on the spinal ranged from myelomalacia (hyposignal in T1 WI and especially hypersignal on T2 WI) to strangulation and hypotrophy of the spinal cord [Figure 1]. Dynamic X-rays are important in treatment planning; performed in flexion-extension, preferentially under medical observation and used to differentiate between mobile and fixed OO.[1-3] Differential diagnosis is made with a fresh odontoid fracture in case of recent violent cervical trauma; or with persistent ossiculum terminale were the ossicle is small and concerns only the odontoid part above the anterior arch of C1. Surgery is uniformly indicated for mobile or compressive fixed OO; however its indication in fixed noncompressive OO is debated; in fact, in those cases, clinical and radiological observation can be proposed with dynamic X-rays every year and MRI every five years.[1] Surgery consists of posterior fusion with or without posterior decompression. Posterior decompression is indicated in the case of nonreducible compression after the failure of transcranial traction; it consists of resection of the posterior arch of the atlas. Anterior decompression might be indicated in some cases of anterior nonreducible compression.[1] We always prefer C1-C2 fixation, because of its least biodynamic impact, high rate of fusion, and its safety compared to other techniques.[9] The surgeon can perform the original technique of plate and screws described by Goel and Laheri in 1994,[9] or one of its variants especially polyaxial screws and roads described by Harms and Melcherin 2001,[10] screw fixation in atlas via posterior arch and lateral mass described by Tan *et al.* in 2003[11] or bilateral crossing C2 laminar screws technique described by Leonard and Wright in 2006.[12] Mostly, we perform posterior C1–C2 fusion with polyaxial screw and rod fixation according to Goel and Harms technique [Figure 1 and 2]. C1 lateral mass screwing was performed in all patients. C2 pedicle screwing is preferable assuring strong fixation passing along pedicles to the thickness of the C2 body; unfortunately, it is not always doable due to pedicle hypoplasia rending is drilling too risky either for the vertebral artery laterally or dura and spinal cord medially [Figures 3 and 4]; in this case, the surgeon must switch to lateral joint fixation of C3 [Figure 3] or bilateral crossing C2 laminar screws [Figure 4]. OO is considered as a permanent unstable lesion and no consolidation is hoped after fixation so putting a bone graft is a good option, giving greater, and more sustainable stability[13] compared to simple fixation always under the risk of material failure (screw and road break). We prefer harvesting the bone graft from the iliac crest and we do not have complications related to the bone graft. Other posterior fixation techniques can be performed. Wiring seems to be no longer an option because of its poor biomechanical results compared with

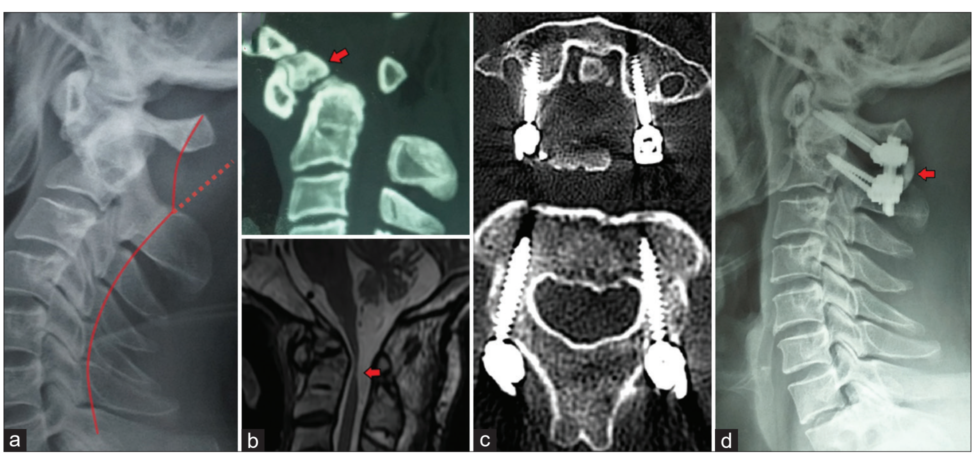

Figure 1: Imaging from case 5; male of 47 years old with Os odontoideum. (a) Preoperative lateral X-rays showing atlantoaxial dislocation cleared by the band of spinolaminar virtual line at the level of C1 (continuous line shows the actual line whereas the discontinuous line shows its normal position). (b) Sagittal computed tomography and magnetic resonance imaging; the arrow in the upper image shows Os odontoideum on computed tomography and the arrow on the lower image shows on magnetic resonance imaging the hypotrophy caused by chronic aggression and compression of the spinal cord. (c) Postoperative axial computed tomography; in the upper image passing through the atlas and in the lower image passing through axis. (d) Postoperative lateral X-rays showing the construct placement, the arrow is pointing to the graft placement

the other available techniques.[13] Occipitocervical fixation is suboptimal option;[14] in fact multi-segmental fusion for mono-segmental instability causes a clear biodynamic poor outcome sacrificing at least 25% of neck flexion-extension by blocking the occipitoaxial and atlantoaxial articulations plus 10% by level in subaxial spine; in addition to 50% of neck rotation by blocking atlantoaxial joint plus 10% by level in subaxial spine;[15] other complications include fixation of patient's neck in exaggerated flexion or extension and obviously with more risk of materiel failure.[16] Transarticular fixation described by Magerl in 1987[13,17] gives a great fusion rate, but it demands an alignment of the atlantoaxial facets[1] which is not always obtained in nonreducible OO; and it seems that up to 20% of patients cannot have a placement of the screw without endangering the vertebral artery.[13] External immobilization with halo vest or collar in postoperative is recommended to increase chances of fusion.[5] All our patients improved clinically; this improvement is based on regain of total or partial motor force, and patient's satisfaction. Few complications were noted in our series, the one case of infection seems to have no relation with the technique and were successfully managed. Although the one case managed by bilateral crossing C2 laminar screws was reoperated to adjust the screws placement; we insist on this technique as a good alternative if pedicle screwing is not possible.

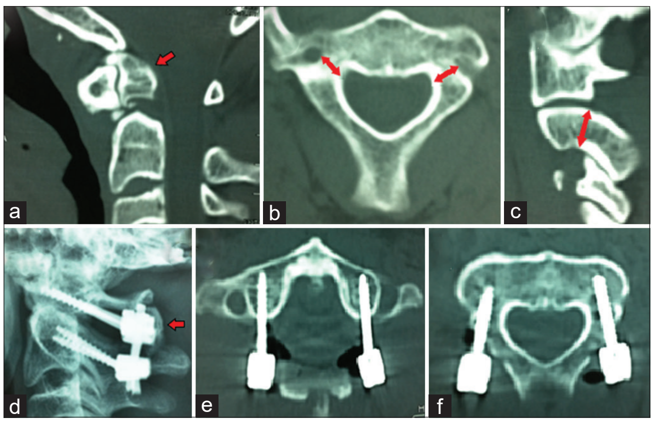

Figure 2: Imaging from case 15; male of 26 years old with Os odontoideum. (a) Sagittal computed tomography; the arrow shows the free well-corticated ossicle. (b) Axial computed tomography passing through C2; the arrows show laterally wide pedicles. (c) Parasagittal computed tomography; the arrow shows the superior to the inferior wide pedicle. (d) Postoperative lateral X-rays; showing C1-C2 fixation, the arrow is pointing to the graft placement. (e and f) Axial computed tomography passing through atlas and axis

CONCLUSION

Although traumatic history is frequently noted, the congenital origin of OO is always evoked. We can observe stable noncompressive lesions, in other cases, surgical management is indicated. C1-C2 fixation according to Goel and Harms technique is our technique of choice; this technique proved its safety, providing high fixation quality with the acceptable biodynamic outcome. Grafting is paramount; once fused; bone graft gives sustainable more lasting fusion with less risk of failure. Once treated, the outcome in OO is in general good; improvement is observed in most patients with few complications.

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Nil.

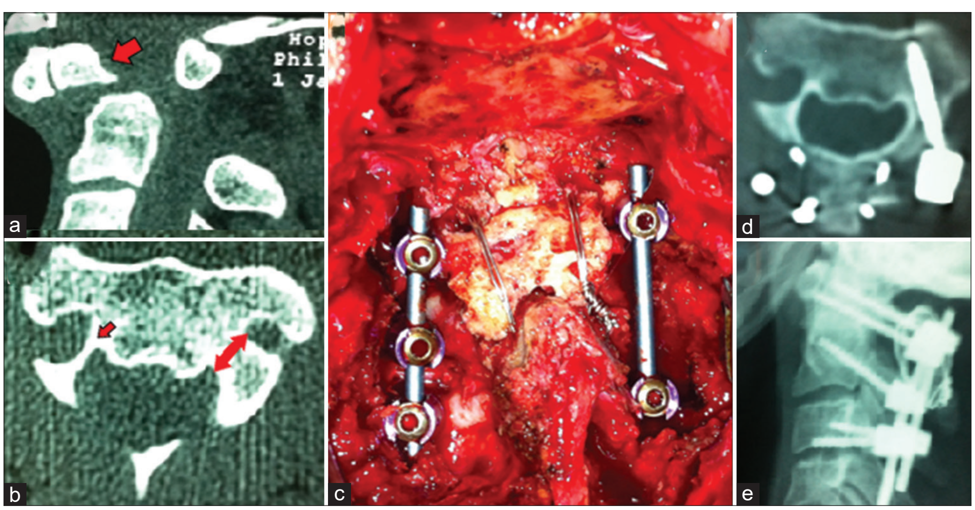

Figure 3: Imaging from case 9, the male of 40 years old. (a) Sagittal computed tomography, the arrow shows the Os odontoideum. (b) Axial computed tomography passing through C2; the arrows show the difference in thickness between the left pedicle where screwing is safe and the right pedicle where screwing is dangerous due to pedicle congenital hypoplasia. (c) Per-operative photography showing left C1 lateral mass, C2 pedicle, and C3 lateral mass screws; on the right side C2 pedicle screwing was skipped; this image shows also the bone graft placement between the posterior arch of C1 and spinous process of C2; hold in place by classic wiring technique. (d) Axial computed tomography passing through C2 showing pedicle screw placement on only the left side. (e) Postoperative lateral X-rays showing the construct placement

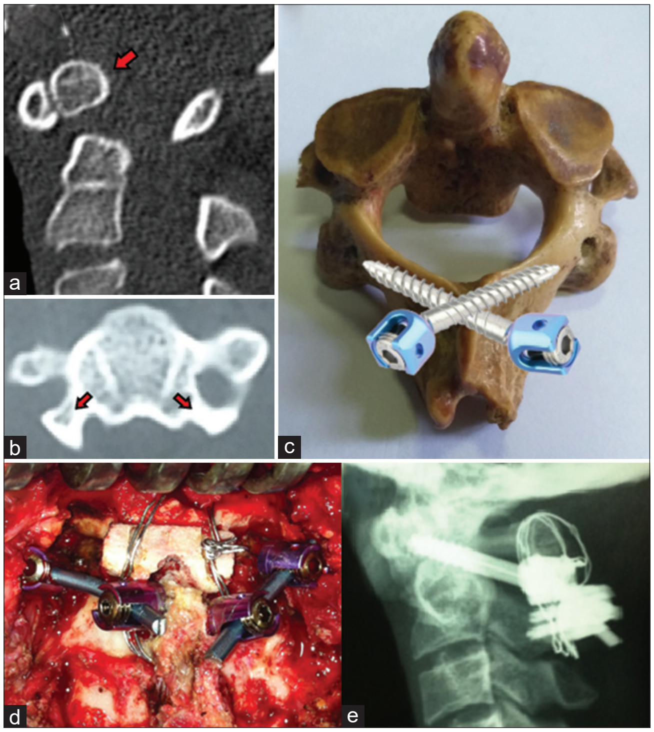

Figure 4: Imaging from case 7; female of 25 years old with Os odontoideum. (a) Sagittal computed tomography showing the Os odontoideum. (b) Axial computed tomography passing through C2; the arrows show the relatively narrow pedicles limiting trans-pedicular screwing. (c) Image of superposition explaining screws placement in bilateral crossing C2 laminar screws technique, our alternative for this case since pedicle screwing is risky. (d) Per-operative photography showing screws placement on C1 lateral masses and laminae of C2; it also shows the bone graft placement between the posterior arch of C1 and spinous process of C2; hold in place by classic wiring technique. (e) Postoperative lateral X-rays showing the construct placement

Conflicts of interest

There are no conflicts of interest.

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