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Rotational dislocation C1–C2 after otoplasty under local anesthesia

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Abstract

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INTRODUCTION

Nontraumatic rotational atlantoaxial subluxation (NTARS) is an uncommon condition in clinical practice. It is mainly observed after infection of the upper respiratory tract, called Grisel, being responsible for 48% of nontraumatic atlantoaxial rotational dislocation. Head-and-neck surgery is the second cause, and adenotonsillectomy is responsible for 31% of NTARS.[1] Although rare, NTARS was reported in the literature after otoplasty[2] and only four reports of patients were found in the researched literature: Dubrana *et al.*, Kelly *et al.*, Durst *et al.* (2012), and Macheboeuf *et al.* (2019).[2,3,4,5] Each author reported one patient with NTARS after otoplasty. All reported patients underwent surgery under general anesthesia.

The goal is to report a rare case of NTARS after bilateral otoplasty, the performed treatment, and follow-up.

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CASE REPORT

A 15-year-old woman presented with head deformity 10 days after bilateral otoplasty under local anesthesia. She presented with a painful torticollis; the head was rotated to the right and tilted to the left side. Computed tomography (CT) showed NTARS [Figure 1].

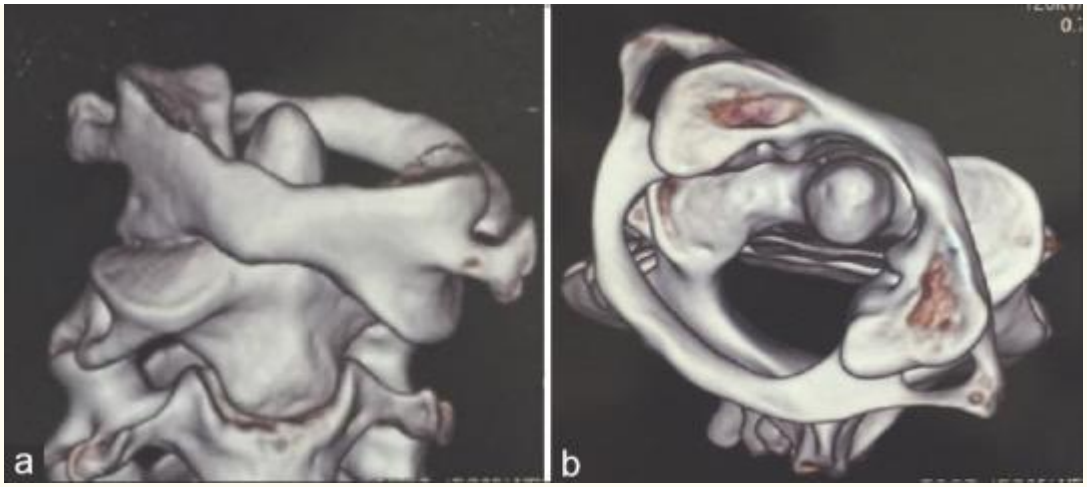

Figure 1

Computed tomography showing the C1–C2 rotational dislocation (a – anterior view and b – axial view)

Reduction was achieved under anesthesia by transoral palpation of the dislocated C1 articular facet associated with manual head traction, followed by cervical halter traction. The anatomical relationship of C1–C2 was confirmed by CT [Figure 2].

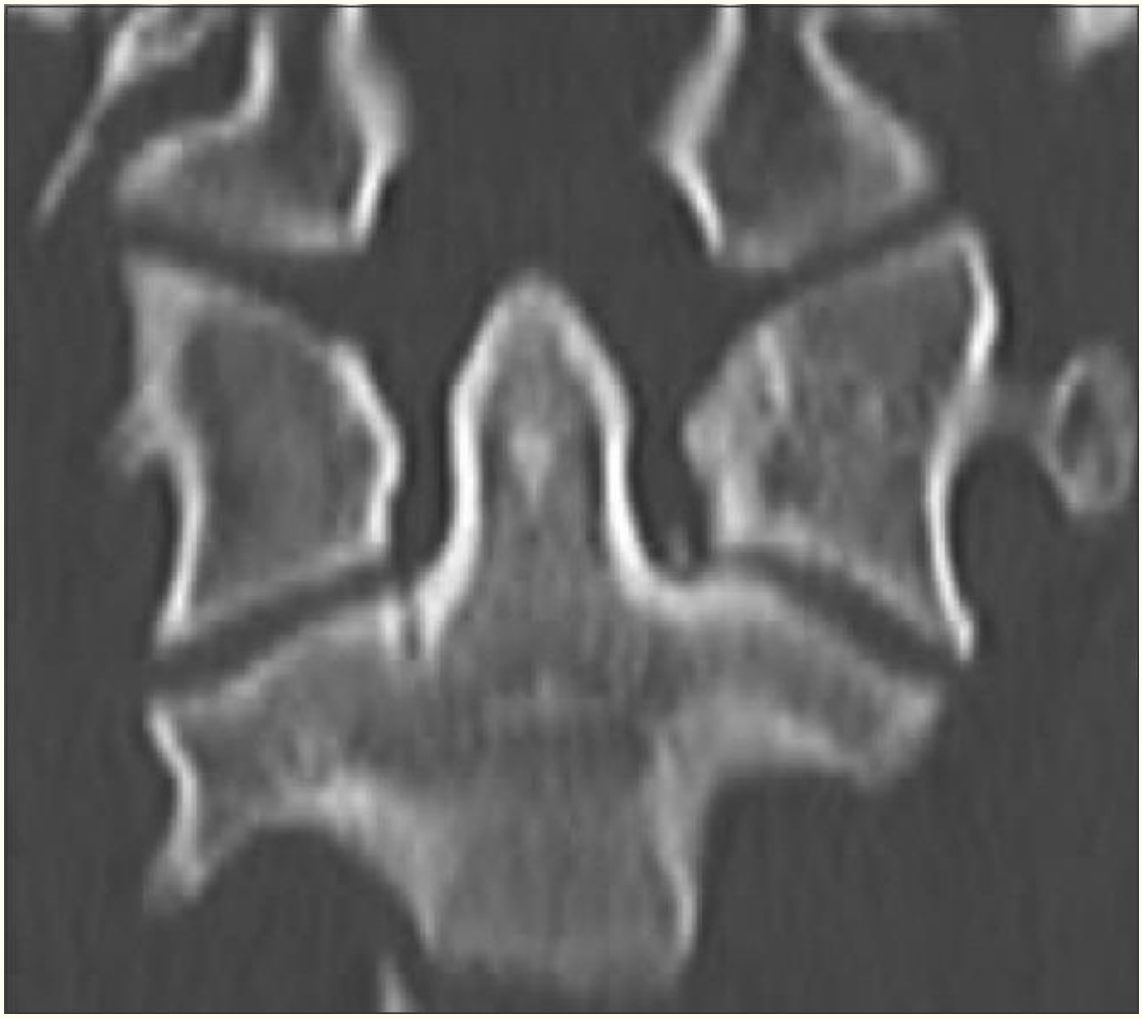

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Figure 2

Computed tomography showing the anatomical reposition of C1–C2 after closed reduction

Two recurrence of the dislocation was observed after two attempts of closed reduction under anesthesia followed by cervical traction and hard collar immobilization. Open reduction and posterior atlantoaxial fixation with C1–C2 sublaminar wire and autologous iliac bone graft was performed, followed by hard cervical collar immobilization for 3 months [Figure 3].

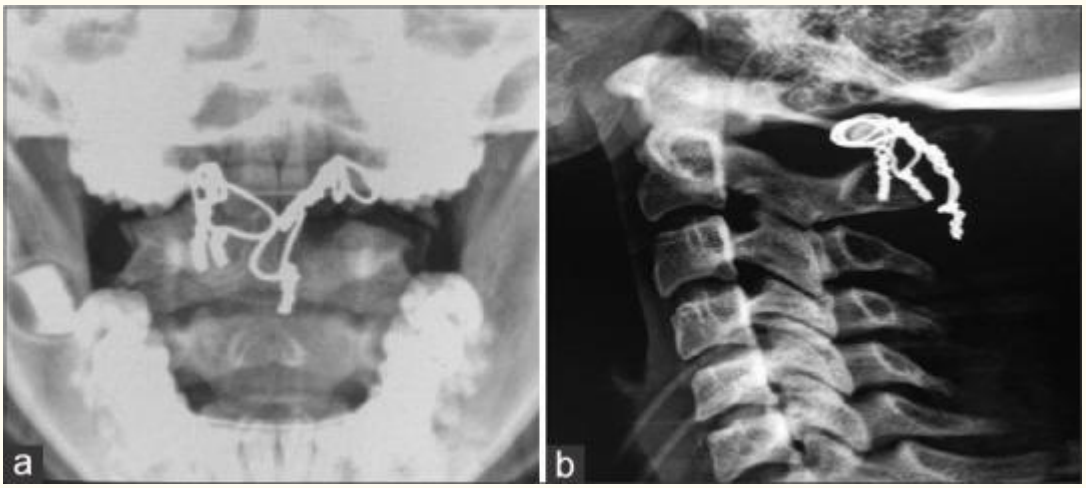

Figure 3

Postoperative radiographs anteroposterior transoral (a) and lateral view (b)

There was neither intraoperative nor postoperative complication. The patient remained neurologically intact, radiographic evidence of bone fusion was observed after 1 year follow-up, and the patient returned to normal activities [Figure 4].

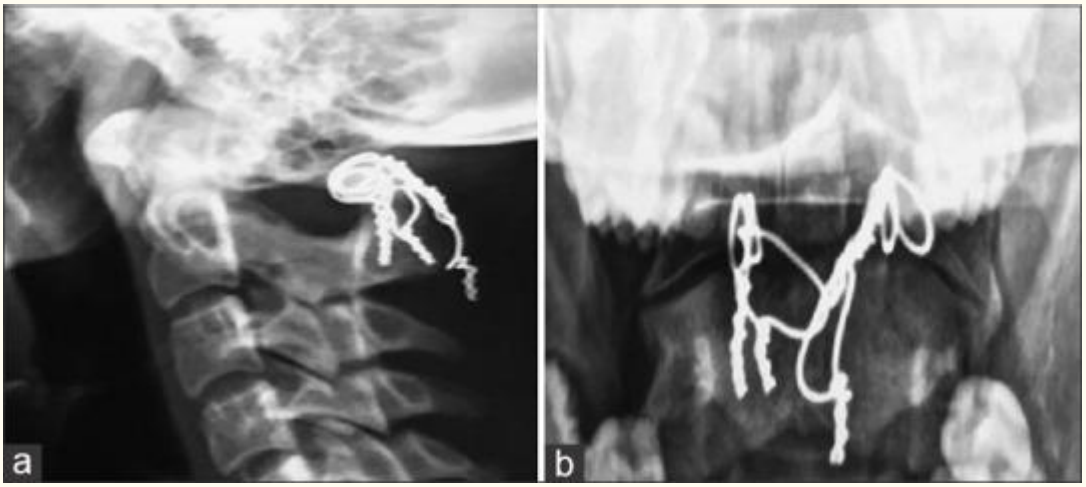

Figure 4

Radiographs of the cervical spine 1 year postoperative, lateral (a) and anteroposterior transoral (b) view

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DISCUSSION

Nontraumatic atlantoaxial rotatory subluxation is a rare condition, and it should be included in the differential diagnosis of a child with painful torticollis after infection of the upper respiratory tract, postadenotonsillectomy, and other surgical procedures, such as pharyngoplasty and ear operation.[6] Torticollis with the head in the oblique position associated with muscular stiffness and reduced range of movements are the typical clinical findings. Usually, this syndrome affects children under the age of 12 years, without predisposition for gender.[7] There are also reports on adults, but it is rare.[7] Neurological complications are rare and reported in 15% of all patients with NTARS.[8]

Laxity of atlantoaxial ligament would be a main cause of instability, and it could explain the physiopathology in patients who underwent surgical procedures as ear operation.[9,10] Marked lateral rotation of the head and extension of the neck during the course of positioning for the surgical procedures are the common features of NTARS in patients without previous history of infection.[2] All reported cases of NTARS were associated with hyperextension of the cervical spine under general anesthesia.[2] However, our patient underwent bilateral otoplasty under local anesthesia although the head and neck was kept in marked lateral rotation and extension during the procedure.

There is no treatment guideline for NTARS, and most articles reported good results with conservative treatment. Surgical treatment is reserved for failed conservative treatment such as nonreducible subluxation or multiple recurrences. Surgical treatment is performed by posterior open reduction and posterior atlantoaxial fixation and arthrodesis with autologous iliac bone graft.[7,10]

The basic principle of NTARS treatment is the reduction of subluxation by traction and its maintenance through external immobilization. Conservative treatment has been able to solve the majority of cases.[10] Manual reposition under anesthesia followed by immobilization was reported with good results.[7,10] This method has the advantage of shortening the treatment period by eliminating the time spent performing traction. In our patient, we choose to start the treatment through closed reduction of subluxation under sedation, using head traction and transoral manipulation of the anteriorly displaced atlas. Instead of minerva cast after manual reposition, we performed traction. Loss l of reposition after manipulation followed by soft collar immobilization was reported by Pilge *et al.*[7]

Considering the instability of C1–C2 joint and recurrence of dislocation, we performed an open posterior reduction and C1–C2 arthrodesis using just wire cerclage. Wire cerclage was the option considering that this procedure can be performed with less exposure of posterior vertebral elements, less dissection, less muscle detachment, and less complications. During follow-up, there was no complication regarding arthrodesis, bone graft healing, and implants. The clinical and radiological outcomes were satisfactory, and after 1 year follow-up, the patient had a normal life and returned to all previous activity.

In the literature, we just found four cases of NTRS after otoplasty.[2,5] The cases reported in the literature showed a good evolution with conservative treatment.

Reduction was stable without recurrence of dislocation, and surgical treatment was not necessary as occurred in our patient who needed surgical treatment to solve the problem.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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