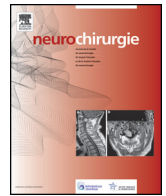




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Short clinical case

The Grisel's syndrome: A non-traumatic subluxation of the atlantoaxial joint

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ABSTRACT

Introduction. – Grisel's syndrome consists in rotational subluxation of C1–C2 following ENT infection or surgery. There is no consensus on management. We present 2 cases requiring surgical treatment in our center.

Case reports. – Two 10-year-old patients presented torticollis with cervical pain resistant to medical treatment, with onset a few months after tonsillectomy. In both cases, radiological assessment, comprising CT scan and MRI, showed Fielding–Hawkins type-3 C1–C2 rotational subluxation, without ligament lesion. After failure of conservative treatment, posterior reaming, realignment, C1–C2 arthrodesis using lateral masses and pars interarticularis screws and bone graft achieved good fusion and immediate spinal stability in all planes of the atlantoaxial complex.

Discussion. – Grisel's syndrome consists in non-traumatic subluxation of the atlantoaxial joint with intact atlantoaxial ligaments. Initial pharyngeal inflammation spreads to the prevertebral fascia via direct connections between the periodontoidal venous plexus and pharyngovertebral veins, inducing fasciitis that leads to abnormal relaxation of the atlantoaxial ligaments and reactional muscle contraction with ankylosis. This phenomenon, appearing gradually and insidiously over a period of a few weeks, creates a frozen joint with ankylosis. Medical treatment with NSAIDs, muscle relaxants, and immobilization is usually sufficient; cervical traction may be needed. Surgical treatment by C1–C2 arthrodesis is indicated in case of failure of medical management or onset of neurologic signs.

Conclusion. – Close collaboration between pediatricians, ENT surgeons and neurosurgeons is essential for early diagnosis and management, which is the main prognostic factor for successful medical treatment, avoiding surgery.

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1. Introduction

Grisel's syndrome consists in non-traumatic atlantoaxial joint subluxation, mainly affecting children. It occurs in a context of ENT infection [1–3] or following head and neck surgery [4]. The mechanism is an inflammatory process extending by contiguity to the muscular fascias and the ligamentous system. First-line treatment combining non-steroidal anti-inflammatory drugs (NSAIDs), physiotherapy and analgesics is usually effective, and surgery is rarely indicated. The surgical options are various, and little described in the literature. The objective of the present study was to report 2 cases of surgical treatment, in order to discuss surgical strategies.

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2. Case reports

2.1. Case 1

A 10-year-old girl was referred to our center for an anomalous head posture progressing for 6 months, with no initial trauma. Six months after tonsillectomy for recurrent pharyngitis, she again developed pharyngitis and fever, with onset of torticollis a few days later. She was treated with oral antibiotics and NSAIDs. The fever and pharyngitis resolved after medical treatment. Initial symptoms included cervical pain and restricted neck motion. Physiotherapy and manipulations were performed by the general practitioner, but did not provide relief.

At examination 6 months after symptom onset, she presented fixed head rotation to the right and >20° head tilt, without neurological complications. She also suffered from dysphonia and dysphagia for solids. CT scan of the upper cervical spine revealed

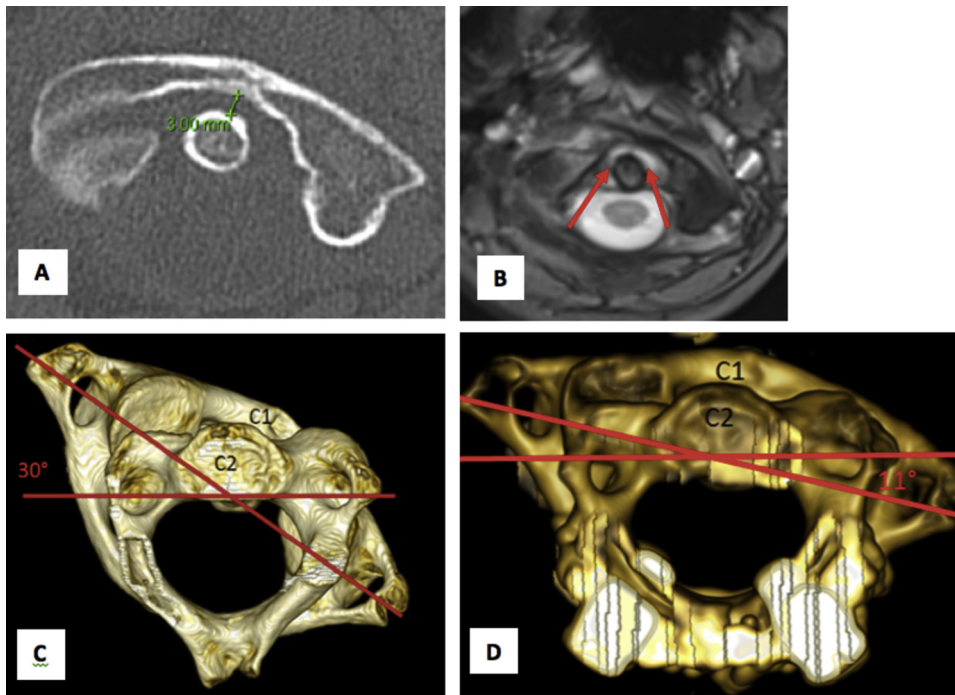


Fig. 1. Case 1, 10-year-old girl with 6 months' persistent torticollis. A. CT scan showing 3 mm anterior displacement of C1 (Fielding–Hawkins type 3). B. MRI T2-weighted sequence, showing no ligament tear. C. 30° preoperative rotation on 3D reconstruction. D. 11° rotation after surgical correction and arthrodesis.

atlantoaxial right rotary subluxation with intact ligaments, confirmed by 3D reconstruction and MRI (Fig. 1). Time to diagnosis was 6 months.

Cervical traction was fitted under general anesthesia with Gardner–Wells tongs and closed reduction under radioscopic control was initially tried. Traction was started immediately, at 5% body-weight, and rapidly increased (after 48 hours) to 15%. Traction in bed-rest was maintained for 7 days; non-enhanced cervical CT scan showed no modification of the subluxation. Due to this lack of clinical and radiological improvement, surgical treatment by open reduction and C1–C2 arthrodesis was necessary.

Cervical angiologic 3D TOF-sequence MRI was performed to determine the exact anatomy of vertebral artery complex. Under general anesthesia, the patient was placed in prone position with head fixation on a Mayfield (Integra®) headrest in neutral position after slight cephalic traction. A posterior approach was performed, and the C1 and C2 posterior arches were exposed subperiosteally. Under microscopic control, the articular facet indentation was exposed bilaterally and drilled. Articular facet reduction was performed under C-arm fluoroscopy to check alignment and position. Reduction of the C1–C2 rotation proved extremely difficult, mostly because of articular deformity and dystrophic ligament calcification. Laminar hooks (Vertex, Medtronic) were placed bilaterally on the posterior arch of C1 and in the inferior lamina of C2, connected by posterior rods. The C1 and C2 laminae were decorticated and lyophilized bone graft was applied. Surgery time was 211 minutes, including installation, radiologic control, and skin-to-skin procedure. There were no intra- or postoperative complications. At 2 months' follow-up, the patient showed complete clinical resolution, with good fusion despite 11° persistent rotation on cervical CT.

2.2. Case 2

A 10-year-old boy presented resistant torticollis with neck pain and >20° left head tilt, following tonsillectomy for chronic tonsillitis and sleep apnea. The patient developed a persistent

cervical lymphadenopathy measuring 4 cm. One month later, he underwent adenoidectomy, with histological diagnosis of large-cell lymphoma. He was treated by rituximab and intrathecal chemotherapy. Six months later, despite orthopedic treatment, the patient presented persistent torticollis and restricted rotational motion. MRI and CT scan were performed 8 months after first symptom onset, showing left-side rotational subluxation of C1–C2 with 4 mm anterior displacement of C1, with intact ligaments. Rotation angle on 3D reconstruction was 24°. Subluxation was type 3 on the Fielding and Hawkins classification, corresponding to a rotated atlas with 3- to 5-mm anterior displacement.

The patient underwent 7 days' cervical traction with the protocol described above. No radiological reduction was obtained, but slight clinical relief was achieved. Surgical treatment was required, consisting in open reduction with C1–C2 arthrodesis. Surgery time was 199 minutes. There were no intra- or postoperative complications. The 2-month follow-up found clinical and radiological resolution, with good fusion and reduction from 24° to 8°.

Table 1
 Therapeutic strategy according to Fielding–Hawkins type in Grisel's syndrome.

Fielding–Hawkins classification	Description	Therapeutic strategy
Type 1	Pure rotation of the atlas in relation to the axis without anterior displacement	Conservative treatment: soft neck brace, NSAIDs, muscle relaxants
Type 2	Atlas rotated with 3- to 5-mm anterior displacement	Conservative treatment, reduction, Philadelphia brace
Type 3	Rotation of the atlas with anterior displacement greater than 5 mm	Closed repositioning, halo extension, cervical traction in bed In case of failure: operative treatment, open reduction, fusion of C1–C2

NSAIDs: non-steroidal anti-inflammatory drugs.

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