



(SK) Griselov syndróm ako komplikácia po adenoidektómii, kazuistika.

(EN) Grisel syndrome as a complication after adenoidectomy, case report.

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SUMMARY/ABSTRACT

Torticollis in paediatric patients is a relatively common symptom. One rare underlying cause is Grisel's syndrome, a non-traumatic atlanto-axial subluxation that typically develops following nasopharyngeal inflammation or ENT (Ear, Nose, and Throat) surgery. According to the Fielding and Hawkins classification, Grisel's syndrome is divided into four types based on the degree of subluxation between the atlas and axis vertebrae. While types I to III often respond well to manual repositioning, stages III and IV may require surgical intervention. The subluxation results in notable changes to the myofascial structures of the neck, including reduced range of motion (ROM), muscular spasms, trigger points, and muscle shortening—all of which can be identified during physical examination. Our case study focused on the conservative treatment approach for this syndrome. We employed a goniometer to measure and monitor improvements in joint ROM, and conducted manual muscle length tests to evaluate muscle stretching. Pain levels and treatment outcomes were assessed using the Visual Analog Scale (VAS). Physical medicine and rehabilitation played a critical role in the recovery process. Over a six-month treatment period, we successfully restored ROM, eliminated painful movements, and prevented the development of neurological complications or the need for surgical intervention.

KEYWORDS

Grisel's syndrome, range of motion (ROM), physical therapy, atlanto-axial subluxation, torticollis, spontaneous/non-traumatic atlanto-axial subluxation (NAAS), ENT surgical complication

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1 INTRODUCTION

Grisel's syndrome is a non-traumatic, spontaneous subluxation of the atlanto-axial joint, typically occurring as a consequence of pharyngeal inflammation (Bocciolini et al., 2005). The condition was first described in 1830 by Sir Charles Bell in a patient with syphilitic pharyngitis (Bell, 1830). It was later named after the French otorhinolaryngologist Pierre Grisel, who in 1930 reported two cases of nasopharyngeal inflammation leading to the development of torticollis (Grisel, 1930).

The exact pathogenesis of Grisel's syndrome remains unclear. However, it is hypothesized that the inflammatory process may increase ligamentous laxity, resulting in subluxation between the atlas and axis vertebrae (Ciftedemir et al., 2018). Grisel's syndrome has been reported in association with various inflammatory conditions of the pharynx and as a complication following ENT procedures, such as adenoidectomy and tonsillectomy (Al-Driweesh et al., 2020). In this paper, we present a case of persistent torticollis following adenoidectomy, which was successfully treated through a conservative approach using physical medicine and rehabilitation methods.

2 CASE PRESENTATION

An 8-year-old boy underwent adenoidectomy under general anaesthesia. The child was placed in the Rose position, and the procedure was performed endoscopically using a Thomson's adenotome. The sur-

gery was completed without complications. Preoperative inflammatory markers were within normal range, with no clinical signs of ongoing bacterial or viral infection.

On the fourth postoperative day, the patient presented with painful torticollis. Physical examination revealed intense neck pain (Visual Analog Scale, VAS: 75 mm), left lateral flexion of the head with rightward facial rotation (Figure 1). Cervical spine range of motion (ROM) was severely restricted due to fixed torticollis:

- Right lateral flexion: 0° (absent)
- Left head rotation: limited to 5°
- Palpation revealed pain in the left sternocleidomastoid muscle, with shortening of the left upper and middle trapezius, left scalene muscles, and right levator scapulae (graded 2).
- Shoulder asymmetry was observed.



Figure 1 – Patient before and after therapy (own source)

No neurological deficits were detected. Laboratory tests for inflammation remained normal. Imaging studies, including cervical spine X-ray and computed tomography (CT), were performed (Figure 2). Anterior-posterior projection confirmed a classic “cock-robin” position. CT confirmed an atlanto-axial rotary subluxation, further detailed via 3D reconstruction and classified as Fielding Type I (Figures 3–4).



Figure 2 – X-ray Cock-robin position on anterior-posterior cervical spine projection (own source)



Figure 3 – CT scan Three-dimensional reconstruction, showing rotatory atlanto-axial subluxation (own source)

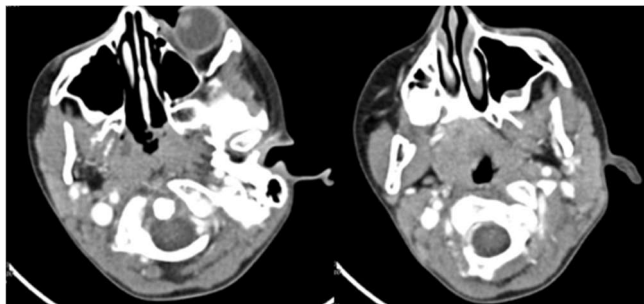


Figure 4 – CT-scan axial view shows atlanto-axial dislocation. Fielding Hawkin I. (own source)

A cervical spine MRI revealed:

- Rotation of C1/C2 segment (25°), with the face turned to the right
- Subluxation in the left intervertebral flexion
- Increased fluid in the atlanto-axial joint and left C1/C2 articulation
- Slightly elevated T2 signal in the C2/C3 disc
- Minor fluid in the right C0/C1 joint
- No soft tissue abscesses or significant lymph node enlargement
- Mild dorsal disc protrusions at C4–C7 (up to 1 mm), without spinal cord compression (Figure 5)

A follow-up MRI after one month showed no significant changes (Figure 6).

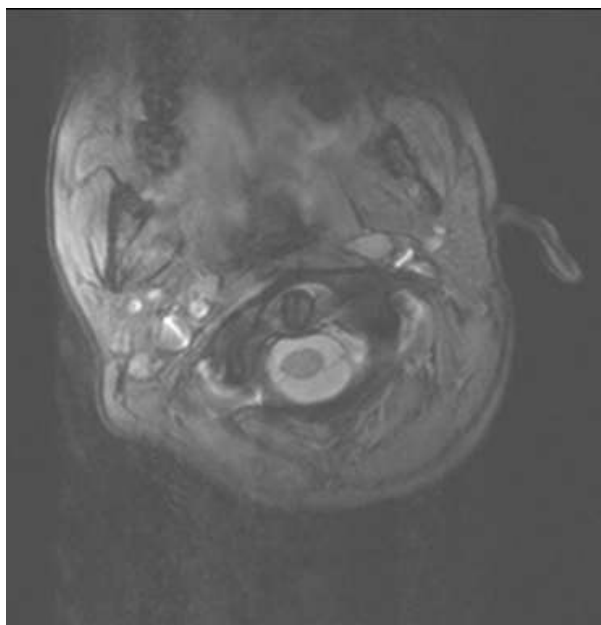


Figure 5 – MRI (own source, department of radiology Uniklinik)



Figure 6 – MRI of the spinal cord + CC transition (Department of radiology Uniklinik)

Initial treatment included antibiotics, spasmolytics, and anti-inflammatory medications, along with bed rest and immobilisation using a cervical collar for one month. Upon admission to the rehabilitation clinic, the patient presented with persistent neck pain and clinical signs of left torticollis. A 6-month rehabilitation protocol was initiated with the primary goals of preventing complications, restoring full cervical ROM, and relieving pain. Progress was monitored using a goniometer for ROM and VAS for pain assessment.

The therapy included:

- Three sessions per week of physical and manual therapy
- Myofascial release techniques to reduce pain and eliminate trigger points
- Cervical joint manipulation via manual traction, performed only after a traction safety test
- Post-Isometric Relaxation (PIR) techniques targeting shortened muscles, especially the upper and middle trapezius, sternocleidomastoids, and scalenes
- Paraffin therapy twice weekly for thermotherapy

Monthly reassessments showed steady improvement:

- After one month:
- Chin-to-sternum distance improved by 1 cm
- Right lateral flexion increased by 10°
- Left head rotation improved by 10°
- Monthly gains of 5–10° in rotation and lateral flexion were documented thereafter

By the end of the 6-month program:

- Symptoms had completely resolved
- VAS score reached 0 (no pain)
- Full restoration of cervical ROM was achieved:
- Lateral flexion: 80°
- Forward flexion: 45°

- Rotation: 90° bilaterally

3 DISCUSSION

Grisel's syndrome is a rare condition, with only a limited number of cases reported in the literature. It primarily affects the pediatric population and is most commonly associated with infections of the upper aerodigestive tract—such as pharyngitis and tonsillitis—or occurs as a postoperative complication of ENT procedures, including adenoidectomy, tonsillectomy, or the insertion of ventilation tubes (Karkos et al., 2007).

According to Al-Driweesh, adenotonsillectomy accounts for approximately 55% of post-surgical cases, followed by pharyngoplasty (11%) (Al-Driweesh et al. 2020). Non-surgical cases have been linked to conditions such as Kawasaki disease and *Mycoplasma pneumoniae*, which were often accompanied by elevated inflammatory markers such as ESR and neutrophilia (Al-Hussain et al., 2024). Other viral causes include Epstein-Barr virus, also associated with Grisel's syndrome (Liu et al., 2020). In 2019, Falsaperla proposed that bacterial pathogens—such as *Streptococcus pyogenes*, *Staphylococcus aureus*, and other infectious agents—may also play a causative role. (Falsaperla et al., 2019).

The pathophysiology of Grisel's syndrome remains incompletely understood. One widely cited explanation is the “two-hit hypothesis,” which proposes that the predisposing factor is increased ligamentous laxity in children, and the second is inflammation-induced muscle spasm in the cervical region, leading to atlanto-axial subluxation (Battiata et al., 2004).

Diagnosis is typically based on clinical examination. A history of recent pharyngeal infection or ENT surgery should raise suspicion of Grisel's syndrome. The classical presentation includes cervical pain and fixed torticollis, often with limited rotation of the C1–C2 joint (Deichmueller & Welkoborsky, 2010). While laboratory investigations may not be diagnostic, inflammatory markers can be elevated in post-infectious cases (Falsaperla et al., 2019). In most cases, symptoms appear within a few days of the triggering event, commonly within the first 5 postoperative days—consistent with our patient, who presented on day four.

Imaging studies are essential for confirmation and classification. X-ray, CT, and MRI of the cervical spine are the most commonly used modalities (Phillips & Hensinger, 1989). CT, particularly with 3D reconstruction, is considered the gold standard for diagnosing atlanto-axial rotary subluxation (Spinnato et al., 2021).

The Fielding and Hawkins classification categorizes the degree of displacement into four types (Fielding & Hawkins, 1977):

- Type I: Fixed rotation without anterior displacement (<3 mm)
- Type II: Unilateral anterior displacement of 3–5 mm
- Type III: Bilateral anterior displacement (>5 mm)
- Type IV: Posterior displacement, often associated with dens fracture or aplasia

The need for surgical intervention generally correlates with the severity of subluxation. Types I and II are typically managed conservatively with immobilisation, anti-inflammatory medications, and physical therapy. Types III and IV, especially those associated with dens abnormalities, often require surgical fusion (C1–C2 arthrodesis). Some authors have reported successful treatment of Type III with halo traction and immobilisation for up to three months. Pilge introduced an alternative approach using manual repositioning under general anaesthesia followed by cervical immobilisation with a Minerva cast. (Pilge et al. 2013)

Early diagnosis and a multidisciplinary approach are critical. Coordination between the pediatrician, physiatrist, ENT surgeon, radiologist, and—when necessary—social workers ensures optimal recovery. As Bevelaqua et al. (2021) emphasize, a biopsychosocial model improves outcomes, particularly in pediatric populations. Similarly, Oláh and Costello (2020) advocate for integrating social services into medical care, especially when long-term cooperation with families is necessary. Building parental trust, as emphasized by Gulášová (2023b), supports daily home-based physiotherapy compliance, which is essential for successful rehabilitation (Gulášová et al., 2023a).

Karkos et al. (2007) conducted a systematic review of 103 cases of Grisel's syndrome reported between 1950 and 2006. Of these, 48% were infection-related and 31% occurred post-adenotonsillectomy.

Most cases were treated conservatively, with surgical intervention as arthrodesis required only in a minority (Karkos et al., 2007). Al-Hussain et al. (2024) concluded that conservative management—including antibiotics and NSAIDs—was sufficient in most cases, resolving symptoms such as neck pain, torticollis, and reduced ROM. Our case supports these findings: early diagnosis combined with structured physical therapy led to full recovery. Notably, even after CT-confirmed realignment of the atlanto-axial joint, residual symptoms persisted due to soft tissue changes, which were successfully addressed through rehabilitation. Guth (2016) emphasized the value of physiotherapy in resolving such functional disturbances, often eliminating the need for prolonged pharmacological treatment (Guth, A. 2016).

The success of conservative management depends on timely diagnosis and early initiation of therapy (Sogoba et al., 2019). Reducing inflammation and addressing infection early are key to preventing complications (Ghodke et al., 2022). Spinnato et al. (2021) noted that ongoing infection or inflammation in the head and neck region may slow recovery (Spinnato et al., 2021). Treatment options beyond antibiotics include muscle relaxants, cervical collars, and traction. Early diagnosis can prevent potentially devastating neurological outcomes (Al-Hussain et al., 2024).

4 CONCLUSIONS

In children presenting with non-traumatic, sudden-onset rigid neck fixation and torticollis, close interdisciplinary collaboration between the pediatrician, physiatrist, and surgeon is essential. Early diagnosis and timely initiation of a tailored treatment plan can significantly reduce the risk of complications and may prevent the need for surgical intervention.

Physical medicine and rehabilitation should be considered an integral component of the management strategy to avoid long-term neurological deficits. In our case, the benefits of a structured rehabilitation program were clearly demonstrated, leading to full functional recovery and resolution of symptoms without surgical intervention.

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