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Evaluation of persistent torticollis following adenoidectomy

Marcel Kraft, MD., Kurt Tschopp, M.D.

Abstract
Post-operative neck complaints are not an uncommon finding following adenotonsillectomy. However, non-traumatic subluxation of the atlantoaxial joint (Grisel's syndrome) should be considered in cases of persistent neck pain and stiffness. An early diagnosis and adequate treatment of this rare condition is mandatory to prevent potentially serious complications. We describe three cases with persistent torticollis in the post-operative period, discuss the pathogenesis and evaluate the clinical management of these patients.

Key words: Torticollis; Adenoidectomy; Surgery; Complications

Introduction

Adenoidectomy with or without tonsillectomy is one of the most common surgical procedures worldwide.[1] Some neck pain and stiffness is reported to occur in up to 10 per cent of patients typically from the fourth to the seventh post-operative day.[2] The most common cause is cervical muscle spasm, but additional considerations have to include cervical adenitis, retropharyngeal and parapharyngeal abscess.[3] However, in cases of persistent cervical pain and torticollis in the post-operative period, more serious causes like cervical spinal subluxation, cervical osteomyelitis and bacterial meningitis should be considered in the differential diagnosis.[4,5]

Although first published by Bell in 1830, non-traumatic subluxation of the atlantoaxial joint is named after Grisel, who described two cases of torticollis nasopharyngien in 1930.[6,7] This condition most frequently affects children and is a rare complication following otolaryngological procedures and upper respiratory infections.[8]

We describe three cases with persistent torticollis following adenoidectomy using the monopolar suction electrocautery for haemostasis, briefly discuss the pathogenesis of post-operative muscle spasm and atlantoaxial subluxation, and evaluate the clinical management of these patients.

Case reports

Case 1

A six-year-old boy underwent an uncomplicated adenoidectomy with myringotomy under general anaesthesia for adenoid hypertrophy and otitis media with effusion. He was placed in the Rose position, the adenoid tissue was removed with a Beck's adenotom, and monopolar suction electrocautery was used for haemostasis. A single dose of intravenous amoxicillin clavulanate was given intraoperatively and the patient was discharged the following day. Four days post-operatively he developed a painful torticollis to the right, difficulty with feeding and fever.

Fig. 1
Case 1 demonstrating a right-sided torticollis with the chin rotated to the left and spasm of the left sternocleidomastoid muscle.

Physical examination revealed spasm of the ipsilateral sternocleidomastoid muscle and a local wound infection (Figure 1). During a four-day course of intravenous antibiotics and local application of hot packs, the pain abated and the torticollis improved. The boy was discharged home on a seven-day course of oral amoxicillin clavulanate. Thirteen days after the initial operation, the patient presented again with new onset of neck pain and stiffness. Computed tomography with three-dimensional reconstruction demonstrated a rotatory subluxation of the right atlantoaxial joint without anterior displacement of the atlas (Figure 2). Flexible nasopharyngoscopy showed granulation tissue as normally seen after adenoidectomy. Intravenous amoxicillin clavulanate therapy was restarted,

From the Department of Otorhinolaryngology, Kantonsspital Liestal, Switzerland.
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FIG. 2

Three-dimensional reconstruction of axial CT scans of Case 1, showing rotatory fixation without anterior displacement of the atlas relative to the axis corresponding a Fielding Type I subluxation.

and a stiff cervical collar was applied. As the torticollis did not subside within one week of conservative treatment, orthopaedic consultation was obtained. The boy was placed under extension in a Glisson's sling for another week (Figure 3). Thereafter, his neck pain and torticollis completely resolved, restoring a full range of painless cervical movement.

Case 2

A six-year-old girl presented to the paediatric emergency room with painful torticollis to the right, bad breath and fever four days after an outpatient adenoidectomy for adenoid hypertrophy. Monopolar suction electrocautery was used for haemostasis during the procedure because of excessive bleeding in the nasopharynx. Physical examination revealed spasm of the cervical muscles and a local wound infection. The patient was discharged home on a 14-day course of oral amoxicillin clavulanate and a close follow-up was performed. Six weeks post-operatively, the painful torticollis had completely resolved, restoring a normal range of motion, and flexible nasopharyngoscopy showed normal conditions in the nasopharynx.

FIG. 3

Case 1 being placed under extension in a Glisson's sling for reduction of atlantoaxial subluxation.

Case 3

A six-year-old girl underwent an uncomplicated adenoidectomy for adenoid hypertrophy on an outpatient basis. In the first post-operative night she developed fever and neck pain. Because of a persistent torticollis, she presented five days later to the referring general practitioner who initiated a 14-day course of oral clarithromycin. Further follow-up was performed at the surgeon's institution. During the oral antibiotic therapy, the neck pain resolved completely, and a full range of cervical movement was restored within four weeks of the operation.

Discussion

Most reviews of post-operative risks following adenotonsillectomy emphasize the presentation and frequency of secondary haemorrhage.[9,10] Only a few articles address less frequent complications like neck complaints.[11,12]

The most common cause of nuchal rigidity or transient torticollis in a post-operative child is a reflex irritation of various neck muscles. The surgical violation of a chronically infected tonsillar bed or adenoid may lead to inflammatory extension into adjacent tissue spaces. Stimulation of fascial planes causes painful contraction of muscle groups that arise from the cervical vertebrae as well as the sternocleidomastoid and trapezius muscle.[3] A similar pathogenesis is postulated for non-traumatic subluxation of the atlantoaxial joint, as most authors agree that peripharyngeal inflammation is essential for the development of this condition.[13] A transport of metastatic effusions from the peripharyngeal tissues to the atlantoaxial articulations has been assumed early.[14] However, identification of the pharyngovertebral veins draining the posterior pharyngeal region gives a possible anatomical rationale for atlantoaxial subluxation. Because of their direct connection with the peri-odontoidal venous plexus and the suboccipital epidural sinuses, these veins provide a haematogenous route for transport of peripharyngeal septic exudates to the upper cervical spinal structures.[15] Some rarely reported complications like cervical osteomyelitis and bacterial meningitis might develop on the same route, as the internal vertebral venous plexus has further connections with the cervical vertebrae and the subarachnoid space.[16] Case 1 in our series showed a diffuse inflammatory enhancement of the epidural space between C2 and C3, supporting the above-mentioned theory (Figure 4).

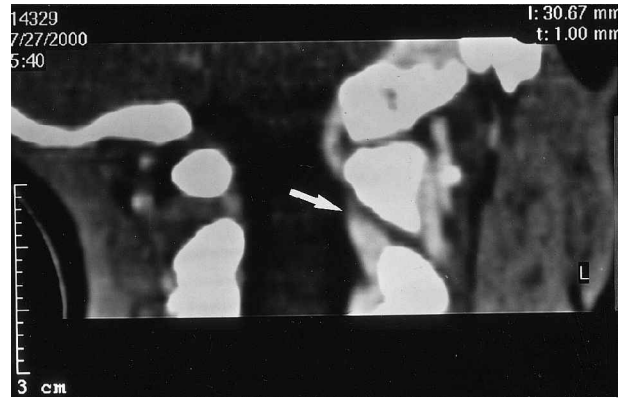

FIG. 4

Coronal CT scan of the upper neck showing a diffuse inflammatory enhancement of the epidural space between C2 and C3 on the left (arrow).

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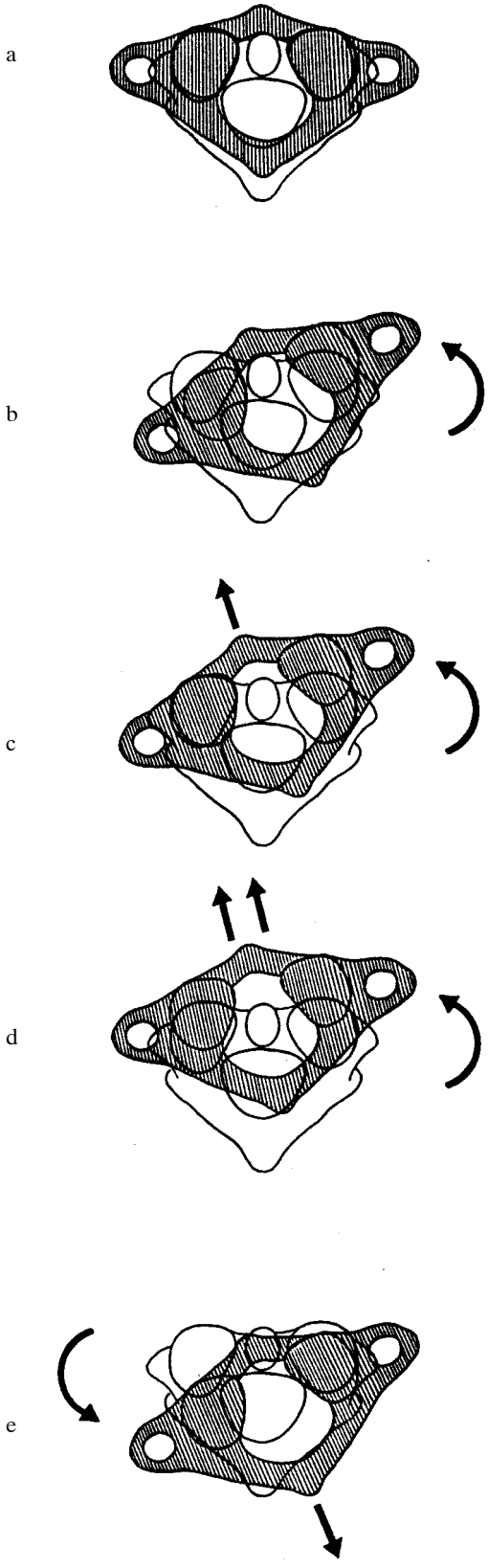

FIG. 5

Modified Fielding classification of atlantoaxial subluxation. (A) Type 0: no fixed subluxation of the atlantoaxial joint. (B) Type I: rotatory fixation without anterior displacement of the atlas (3 mm or less). (C) Type II: rotatory fixation with anterior displacement of the atlas of 3–5 mm. (D) Type III: rotatory fixation with anterior displacement of the atlas of more than 5 mm. (E) Type IV: rotatory fixation with posterior displacement of the atlas.

To understand the further pathogenesis of Grisel's syndrome, a profound knowledge of the joint between the atlas and axis is essential.[17] Its principal movement is that of axial rotation, but flexion–extension and tilting are also possible. Hereby, the tranverse ligament prevents anterior dislocation of the atlas on the axis. The paired alar ligaments assist in this function but primarily prevent excessive rotation. The other ligaments of the atlantoaxial complex are of minor importance to stabilization.[18] While some authors suggested abnormal loosening of the involved ligaments as possible mechanism for the development of Grisel's syndrome, recent investigations have demonstrated the condition to begin rather as a typical torticollis caused by spasm of irritated neck muscles.[8] If long-standing, this may result in distension of the ligaments involved and eventually in atlantoaxial subluxation.[19] The prerequisite is probably elastic or lax ligaments, explaining why Grisel's syndrome is almost exclusively seen in children and patients with Down's syndrome, although cases occurring in adults have been described.[20–22]

Non-traumatic subluxation of the atlantoaxial joint has been classified by Fielding and Hawkins into four types (Figure 5).[23] Fielding Type I and II subluxations are the most common found clinically with no neurologic impairment. Case 1 of our series was consistent with a Type I subluxation. Fielding Type III and IV subluxations, although rare, are invariably associated with spinal cord compression and possible fatal consequences.[21] The Fielding classification describes different degrees of atlantoaxial subluxation and is therefore a prognostic indicator of the potential for complications.[23] As its development seems to be a step-by-step process rather than a sudden onset, we propose to add the Type 0 subluxation to this classification. Those patients with persistent torticollis in beginning Grisel's syndrome demonstrate no radiologic displacement or rotatory fixation of the atlantoaxial joint yet.

One reason for a higher incidence of post-operative neck complaints might be the use of monopolar suction electrocautery. Although a very helpful instrument for meticulous haemostasis and removal of small tissue rests within the choanae and around the eustachian tube during adenoidectomy, the disadvantage of excessive use is augmented post-operative pain and predisposition for local wound infection. Therefore prophylactic antibiotic therapy should be considered, if extensive haemostasis is necessary during the procedure.

The clinical management begins pre-operatively in advising the parents to present their child at the surgeon's institution in any case of persistent torticollis. If possible, physical examination of a child with neck complaints should include mouth inspection and flexible nasopharyngoscopy.[3] Antibiotic therapy is initiated to control the wound infection.[13] If compliance and a close follow-up are guaranteed, this can be done on an outpatient basis. In all other patients, hospitalization and intravenous antibiotic therapy is recommended. In case of persistent torticollis without improvement to conservative treatment within two weeks, computed tomography (CT) with three-dimensional reconstruction should be achieved.[24,25] This investigation proved to be more conclusive than plain anteroposterior and lateral films, while dynamic roentgenograms, cineroentgenography and dynamic CT with maximal head rotation to each side provide no additional information and have to be performed under general anaesthesia.[26,27] Depending on the result, orthopaedic or neurosurgery consultation is obtained. A Type 0 subluxation generally needs no further treatment. However, a close follow-up is recommended, as atlantoaxial rotatory fixation can still develop in the course of the disease.

Fielding Type I and II subluxations may be treated with antibiotics and a cervical collar, but in some patients cervical traction is necessary for reduction. Fielding Type III and IV subluxations generally need bed rest in cervical traction followed by a period of neck immobilization in a cervical collar to prevent recurrent subluxation. If conservative treatment fails to achieve reduction or is followed by neurological symptoms, arthrodesis of the first and second cervical vertebrae is indicated.[8,27]

Early recognition and adequate treatment within one month after the onset of symptoms has shown the best outcome as the articulations are still mobile. In most of these cases, subluxation reduces spontaneously or after a short period of cervical traction. However, late diagnosis more than one month after the initial symptoms often requires some form of operative arthrodesis as the deformity is usually immobile.[26] Therefore all physicians involved in the post-operative care of children following otolaryngological procedures should be aware of this rare diagnosis.[3]

Conclusions

Persistent torticollis following adenoidectomy should alert the surgeon to a beginning subluxation of the atlantoaxial joint (Grisel's syndrome). As pathogenetically a step-by-step development beginning with metastatic spread of infection via the pharyngovertebral veins is assumed, early antibiotic therapy and a close follow-up of these patients is recommended to prevent potentially serious complications. Computed tomography with three-dimensional reconstruction has proved to be an excellent method of documenting the presence and degree of atlantoaxial subluxation. A modest use of electrocautery and a perioperative antibiotic prophylaxis might eventually prevent this rare condition after otolaryngological procedures.

References

1 Carithers JS, Gebhart DE, Williams JA. Postoperative risks of pediatric tonsilloadenoidectomy. *Laryngoscope* 1987;**97**:422–9
2 Rundle FW. Post-tonsillectomy morbidity: a clinical trial of a local penicillin-steroid-anesthetic mixture. *Ann Otol Rhinol Laryngol* 1967;**76**:1060–6
3 Singer JI. Evaluation of the patient with neck complaints following tonsillectomy or adenoidectomy. *Pediatr Emerg Care* 1992;**8**:276–9
4 Baker LL, Bower CM, Glasier CM. Atlanto-axial subluxation and cervical osteomyelitis: two unusual complications of adenoidectomy. *Ann Otol Rhinol Laryngol* 1996;**105**:295–9
5 Bartrakov N. Meningitis following tonsillectomy. *Vestn Otorinolaringol* 1964;**26**:92–5
6 Bell C. *The nervous system of the human body: embracing papers delivered to the Royal Society on the subject of nerves*. London: Longman, Rees and Orme, 1830:118,403
7 Grisel P. Enucleation de l'atlas et torticollis nasopharyngien. *Presse Med* 1930;**38**:50–3
8 Wetzel FT, La Rocca H. Grisel's syndrome: a review. *Clin Orthop* 1989;**240**:141–52
9 Rasmussen N. Complications of tonsillectomy and adenoidectomy. *Otolaryngol Clin North Am* 1987;**20**:383–90
10 Davidson TM, Calloway CA. Tonsillectomy and adenoidectomy. *West J Med* 1980;**133**:451–4
11 Gibb AG. Unusual complications of tonsil and adenoid removal. *J Laryngol Otol* 1969;**83**:1159–74
12 Tolczynski B. Tonsillectomy, its hazards and their prevention. *Eye Ear Nose Throat Monthly* 1969;**48**:71–80
13 Mathern GW, Batzdorf U. Grisel's syndrome: cervical spine clinical, pathologic and neurologic manifestations. *Clin Orthop* 1989;**244**:131–46
14 Wittek A. Ein Fall von Distensionsluxation im Atlanto-epistropheal-Gelenke. *Münch Med Wochenschr* 1908;**55**:1836–7
15 Parke WW, Rothman RH, Brown MD. The pharyngovertebral veins: an anatomical rationale for Grisel's syndrome. *J Bone Joint Surg* 1984;**66**:568–74
16 Tami TA, Burkus JK, Strom CG. Cervical osteomyelitis: an unusual complication of tonsillectomy. *Arch Otolaryngol Head Neck Surg* 1987;**113**:992–4
17 Werne S. Studies in spontaneous atlas dislocation. *Acta Orthop Scand Suppl* 1957;**23**:1–150
18 Hecker P. Appareil ligamenteux occipito-atloido-axoidien: étude d'anatomie comparée. *Arch Anat Histol Embryol* 1923;**2**:61–95
19 Welinder NR, Hoffmann P, Håkansson S. Pathogenesis of non-traumatic atlanto-axial subluxation (Grisel's syndrome). *Eur Arch Otorhinolaryngol* 1997;**254**:251–4
20 Harley EH, Collins MD. Neurologic sequelae secondary to atlantoaxial instability in Down syndrome. *Arch Otolaryngol Head Neck Surg* 1994;**120**:159–65
21 Wilson MJ, Michele AA, Jacobson EW. Spontaneous dislocation of the atlanto-axial articulation, including a report of a case with quadriplegia. *J Bone Joint Surg* 1940;**22**:698–707
22 Sullivan AW. Subluxation of the atlanto-axial joint: sequel to inflammatory processes of the neck. *J Pediatr* 1949;**35**:451–64
23 Fielding JW, Hawkins RJ, Hensinger RN, Francis WR. Atlantoaxial rotary deformities. *Orthop Clin North Am* 1978;**9**:955–67
24 Fielding JW, Stillwell WT, Chynn KY, Spyropoulos EC. Use of computed tomography for the diagnosis of atlanto-axial rotatory fixation. *J Bone Joint Surg* 1978;**60**:1102–4
25 Samuel D, Thomas DM, Tierney PA, Patel KS. Atlanto-axial subluxation (Grisel's syndrome) following otolaryngological diseases and procedures. *J Laryngol Otol* 1995;**109**:1005–9
26 Phillips WA, Hensinger RN. The management of rotatory atlanto-axial subluxation in children. *J Bone Joint Surg* 1989;**71**:664–8
27 Fielding JW, Hawkins RJ. Atlanto-axial rotatory fixation. *J Bone Joint Surg* 1977;**59**:37–44

Address for correspondence:
Marcel Kraft, M.D.,
Department of Otorhinolaryngology,
Kantonsspital Liestal,
Rheinstrasse 41,
CH-4410 Liestal,
Switzerland

Fax: +41 61 925 28 11.

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