CASE REPORT

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Grisel Syndrome as a Complication After Adenoidectomy, Case Report

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Summary

Torticollis in paediatric patients is relatively common symptom. It can be a result of a rare condition called Grisel's syndrome which is characterised by a non-traumatic atlanto-axial subluxation. This syndrome developed either post nasopharyngeal inflammation or ENT surgeries. According to Fielding and Hawkins classification, Grisel's syndrome can be divided to 4 types based on the degree of subluxation between atlas and axis. stage III-IV may require surgical intervention. Manual repositioning showed a good result in type I-III. As a consequence of joint subluxation there are changes on the myofascial structures of the neck. Decrease ROM in the joint area, Muscular spasm, Trigger points and muscles shorting all these can be noticed on physical examination. Our case study focused on conservative treatment for this syndrome. Using the Goniometer, we were able to monitor improvement in joint ROM, manual muscle length test to control involved muscles stretching. Physical medicine and rehabilitation can be considered as an essential part of the treatment protocol.

Keywords Grisel's syndrome, ROM, physical therapy, Atlanto-axial subluxation, Torticollis, spontaneous or Non-traumatic atlantoaxial subluxation (NAAS). ENT surgical complication.

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Introduction

Grisel's syndrome is a non-traumatic spontaneous subluxation of the atlanto-axial joint consequent to pharyngeal inflammation 1. In 1830, Sir Charles Bell was the first to describe it in a patient with syphilitic pharyngitis 2. The name was derived from the French otorhinolaryngologist Pierre Grisel who reported 2 cases of nasopharyngeal inflammation leading to the development of torticollis. 3

The pathogenesis of this condition is still not clearly understood. The inflammation process may lead to increased flexibility of the ligaments which causes subluxation between axis and atlas. 4

Grisel syndrome has been reported in association with inflammatory processes of the pharynx and following ENT surgical procedures as adenoidectomy and tonsillectomy. 5

In this paper, we report a case of persistent torticollis following adenoidectomy which successfully recovered with conservative treatment using physical medicine and rehabilitation methods.

Case report

An 8-year-old boy underwent adenoidectomy under general anaesthesia. The child was placed in the Rose position, adenoidectomy was done endoscopically with a Thomson's adenotome. The procedure was completed without any complications. On the 4th postoperative day, he presented with painful torticollis. X-ray of the cervical spine as well as Computed Tomography (CT) scan were performed for diagnosis. Cock-robin position was detected on anterior-posterior cervical spine projection. CT scan of the cervical spine showed atlanto-axial rotary subluxation, which was confirmed by three-dimensional (3D) reconstruction and Classified as Fielding typ 1. MRI of the cervical spine was performed and showed no spinal cord pathological changes, there was a clear rotation in C1-C2 segment of 25-degree, face

rotated to the right with a subluxation posture in the left intervertebral ligaments. Accumulation of fluid in the area of Atlanto-axial joint and in the region of left C1-C2 INTERVERTABRAL DISC. The soft tissue of the neck showed no access. Lymphatic nodes enlargement along the main vessels of neck. The patient was given antibiotics, spasmolytics and anti-inflammatory treatment, in addition to bed rest and immobilisation with a cervical collar for one month. In rehabilitation clinic patient presented with neck pain, a clinical picture of left torticollis, left lateral flexion of the head with rotation to the right side. ROM was limited due to fixed torticollis. Lateral flexion to right was impossible, left head rotation limited, palpation of m. sternocleidomastoids painful, shortening of left upper and middle trapezius, left m. sternocleidomastoids left scalene and right levator scapulae, shoulder asymmetry. The rehabilitation protocol was planned for 6 months, starting with 3 session per week of physical and manual therapy including myofascial release technic, manual traction of the Cervical spine. Followed by Post isometric relaxation (PIR) of shortened muscles, mainly upper and middle Trapezius, sternocleidomastoids and scalene. Paraffin administered as a form of thermotherapy 2 sessions per week. Monthly the patient status was re-evaluated with gradually decrease of the symptoms and increase in ROM. After first month BS 1 cm, lateralflexion to right better in 2 cm , rotation of head better 3 cm to left, by end of rehabilitation plan, symptoms disappeared and the range of motion was fully restored.

Figure 1 Patient before and after therapy



Figure 2 Cock-robin position on anterior-posterior cervical spine projection



Figure 3 Three-dimensional reconstruction of CT scan, showing rotatory atlanto-axial subluxation.

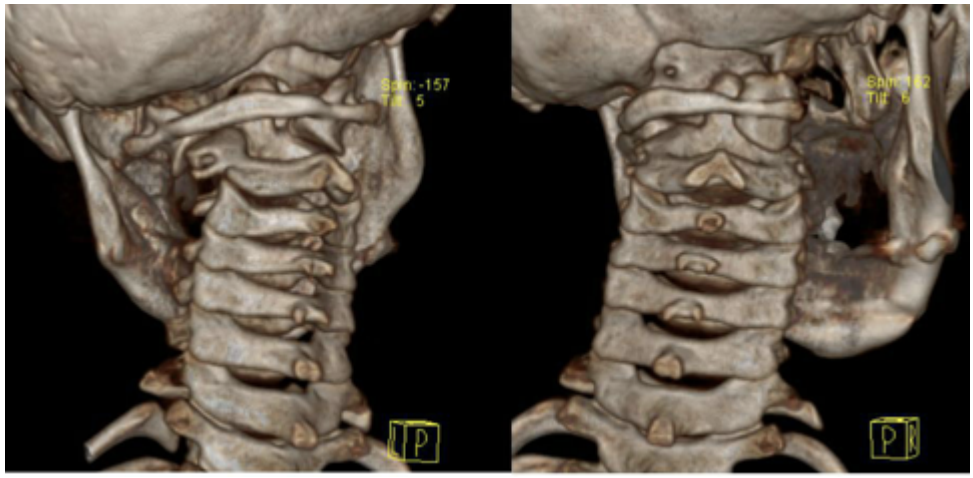


Figure 4 Axial view of CT-scan shows atlanto-axial dislocation (Fielding Hawkin I)

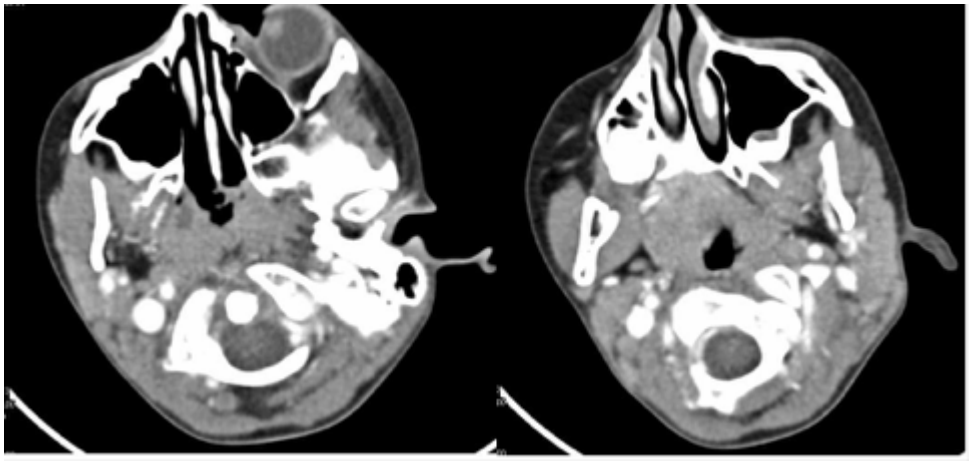


Figure 5 MRI

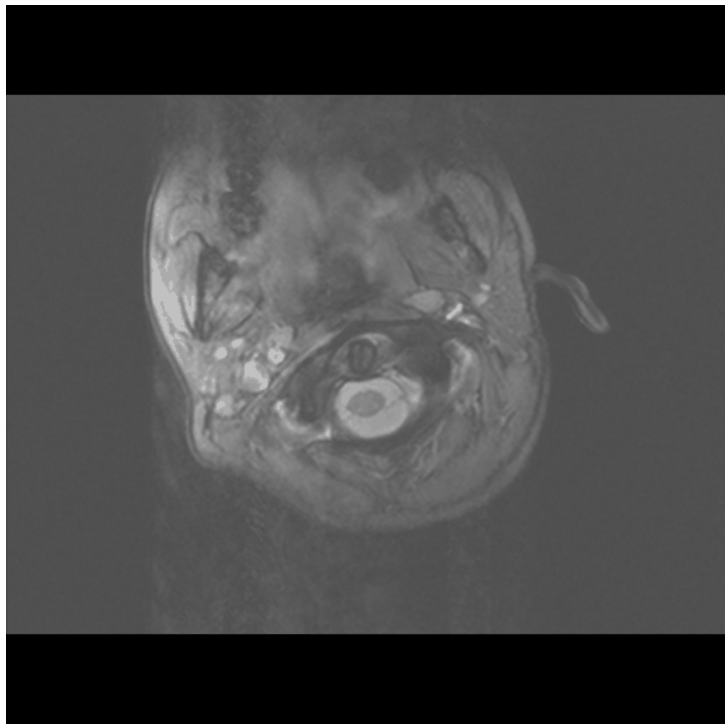
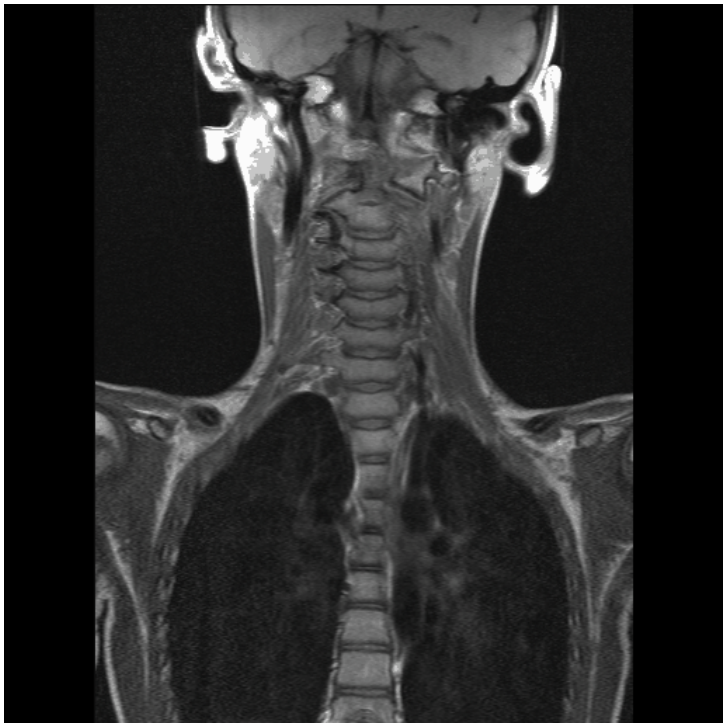


Figure 6 MRI



Department of radiology Uniklinik

(Rotation in the C1/2 segment • 25° (face turned to the right) - with subluxation - in the left intervertebral flexion. Increased fluid in the atlantoaxial joint and in the area of the left C1/2 intervertebral flexion. Slightly increased signal of the C2/3 disc in the T2 sequence. Slightly increased fluid in the C0/C1 joint on the right. No abscess found in the soft tissues of the neck. Bilateral enlargement of the left lymph nodes along the major vessels of the neck and under the m. sternocleidomastoids. Dorsally to the submandibular salivary gland, there is an enlarged lymph node measuring 10x15x35mm with higher signal in T2 compared to other lymph nodes (cannot rule out lymphadenitis/coliculation). Recommend follow-up ultrasound examination.

C4-7: Mild dorsal disc protrusions (up to 1mm) without contact with the spinal cord.)

MRI report after 1 month

MRI of the spinal cord + CC transition

(Rotation in the C1/2 segment by 25° (C1 is rotated compared toC2 counter clockwise when viewed from the front;

subluxated position in the left intervertebral flexion.

Increased fluid in atlantoaxial junction and in the area of the left intervertebral flexion of C1/2.

finding without significant change compared to earlier report one month ago

C4-7: Mild dorsal disc protrusions (up to 1mm) without contact with the spinal cord.)

Discussion

Grisels syndrome is a rare condition with only few number of cases reported in the literature. Generally it occurs in the paediatric population due to infections of the upper aerodigestive system (i.e., pharyngitis, tonsillitis) or as surgical complication of ENT procedures (i.e, adenoidectomy, tonsillectomy, ventilation tube insertion.)6 According to Al-driweesh et al., the most common post-surgical cause was adenotonsillectomy (55%), followed by pharyngoplasty (11%)7. Karkos, P D et al. in his systemic review included 103 cases between 1950-2006. Of which 48% were due to infection and 31% post-adenotonsillectomy. According to his finding most of the cases were treated conservatively; arthrodesis was necessary in a few cases. Al-Hussain, Omair H, and Ghadah Al-Hussain

came with a conclusion that most patients are treated conservatively with antibiotic, NSAID to alleviate clinical symptoms which include neck pain, torticollis and decrease neck ROM. Other cases which were not related to post-surgical GS were reported as a complication of Kawasaki disease or Mycoplasma pneumoniae it was accompanied by increase in inflammatory markers e.g., ESR, neutrophilia. Other viral or bacterial specious which was found to cause GS including Epstein-Barr virus, Kawasaki disease, *Streptococcus pyogenes*, *Staphylococcus aureus*, and other infectious agents. Liu . falsaperla

Etiopathogenesis of this condition is not fully understood. Some authors suggested a two hits hypothesis. One theory proposed the pre-existing higher flexibility of ligaments in children. The second is that the inflammatory process in the cervical muscles leading to spasm which results in the atlantoaxil joint subluxation. 8 Symptoms usually occurs few days after the procedure, where most the patients presented within 5days 9, which was the case in our patient who presented with symptoms on the 4th postoperative day.

Diagnosis of the Grisels syndrome can be made based on clinical examinations, while history of previous ENT surgery or pharyngeal infections should rise suspicion of Grisels syndrome. Early diagnosis can prevent from the devastating neurological complications. 6 this should be followed by immediate start of the treatment Deichmueller

Typical symptoms of GS include cervical pain and fixed rotation to one side with a slight fixation resulting from the limited rotation of the C1-C2 joint. 9 Laboratory investigations are considered to be not very helpful for the diagnosis of GS although inflammatory markers can be noticed in post infectious cases(7). Paolo spinato noticed that the presence of concomitant infection or inflammation in the head and neck region can negatively impact the rate of recovery. Imaging studies play an important role in the diagnosis of GS, this include X-ray, CT scan and MRI of the cervical spine. CT scan is considered to be the most important imaging method for diagnosis of GS (8) 19.

Fielding and Hawkins classification of GS was made to describe the degree of atlantoaxial displacement. The classification is divided into 4 subtypes. 10 Type 1 is the most common subtype. 11 Type 1 is characterised by fixed rotation of C1 - C2 , disluxation less than 3mm without subluxation.

Type 2 presence of subluxation in one side of the atalno-axial joint. Dislocation from 3-5 mm.

Type 3 Bilateral ventral joint subluxation. Dislocation more the than 5mm.

Type 4 posterior C1 subluxation with combination of dnes fructure or dnes aplasia. 12.

The need for surgical treatment is increased with higher stages. Stage I-II generally are treated with non invasive approach. While stage III-IV are treated surgically with C1-C2 fusion especially when it is combined with dens fracture or dens aplasia. Some authors treated stage III with 3 months immobilisation and a halo brace. Pilge came with a new approach of treatment by manual repositioning under general anaesthesia followed by immobilisation of cervical spine with Minerva cast for 2 months.12.

In our case we were able to prove that early diagnosis with appropriate physical therapy can resolve the problem. Even after the normalisation of the Atlanto axial joint on CT scan we noticed that, there can be still changes in the soft tissue which cause persistence of the symptoms. Physiotherapy and rehabilitation can be used for resolving such a functional changes without the need for long term general myorelaxant and NSAID.

Conclusion

In children with a non-traumatic sudden rigid neck fixation and torticollis a close collaboration between paediatrician, physiatrist and surgeon is crucial. early diagnosis and treatment plan can prevent the need of surgical intervention. Physical medicine and rehabilitation treatment should be included always in the treatment for avoiding long term neurological complications. Benefits of rehabilitation was remarkable in our case study.

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