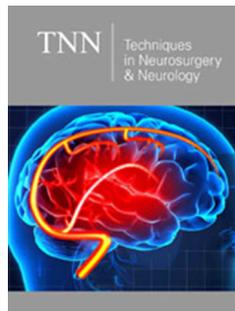


Post COVID Grisel's Syndrome, is there any Association?

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Abstract

Grisel's Syndrome (GS) is a rare, non-traumatic and rotatory atlantoaxial subluxation. Upper airways, head and neck infections and surgical procedures are the main causes of this syndrome. Post COVID GS is not studied in the literature. Although we believe that COVID-19 spreading in upper airways, related immune response and over-relaxation of ligaments can lead to C1-C2 rotatory subluxation and GS, the exact pathophysiology of this process is unknown. We conclude that the virus may present with an extensive range of manifestations such as GS and should be considered in children with COVID-19 infection. As a result, we report 3 GS cases associated with COVID-19 infection for the first time.

Introduction

Grisel's Syndrome (GS) is a rare, non-traumatic and rotatory atlantoaxial subluxation. It is usually detected in children. Upper airways, head and neck infections and surgical procedures are the main causes of this syndrome [1-12]. History of the mentioned items and patients presenting with painful torticollis, fever and reduced range of motion should be notified. Anterior-posterior Direct Cervicography (DCG), Cervical Computerized Tomography (CT) and Magnetic Resonance Imaging (MRI) are usually recommended to confirm diagnosis and classify the patients. Fielding-Hawkins classification can be used to evaluate these cases (Table 1). Based on type and time of syndrome, supportive and surgical therapy is recommended [13,14]. Clinical symptoms and signs of CNS (Central Nervous System) and PNS (Peripheral Nervous System) involvement can be seen in up to 25% of SARS-CoV-2 (severe acute respiratory syndrome coronavirus-2) infected patients. Some researchers propose the term "euro-COVID syndrome" for presentations with pure CNS and PNS presentations at onset. Post COVID GS is not studied in the literature. As a result, we report 3 GS cases associated with COVID-19 infection for the first time.

Table 1: Fielding hawkins classification.

Fielding Hawkins Classification	
Type 1	Rotation of the atlas on the axis without displacement, or with anterior displacement ≤ 3 mm
Type 2	Rotatory fixation with anterior displacement of the atlas 3-5mm
Type 3	Rotatory fixation with anterior displacement of the atlas ≥ 5 mm
Type 4	Rotatory fixation with posterior displacement (extremely rare condition)

Description of Cases

Case 1

A 9-year-old female patient was admitted with painful torticollis and reduced cervical range of motion. She had a history of hospitalization for 4 days due to COVID-19 infection one

week before. Her past chief complaint of first admission was fever and cough and positive subjective or objective clinical findings were malaise, dyspnea, and anosmia. Moreover, the standard diagnostic method by real-time reverse transcription polymerase chain reaction (rRT-PCR), from a nasopharyngeal swab confirmed the detection of the virus' nucleic acid. At this time, there was not any complication related to COVID-19 on physical examination. Neurological assessment was also normal. Anterior-posterior DCG showed Cock-robin position and cervical CT detected asymmetric thickening of the left parapharyngeal soft tissue and rotator atlantoaxial subluxation (Type 2 F-H). She was assessed by neurosurgery department and recommendations of bed rest, immobilization, physical therapy, reduction, antibiotics, and anti-inflammatory treatment. Clinical manifestations slowly decreased, and the patient was symptom free after one month.

Case 2

A 7-year-old male patient was hospitalized with clinical manifestations including fatigue and breathing difficulties. rRT-PCR detected COVID-19 infection. 5 days after admission, her neck was painful and deviated severely. Physical examination showed fever and torticollis and cervical imaging's (DCG and CT) confirmed the similar positive findings of Case 1 (Type 2 F-H). Physical limitation, manipulation, traction, reduction, antibiotics, and anti-inflammatory medications lead to full recovery after 3 weeks.

Case 3

An 8-year-old female with a 2-week history of recent COVID-19 infection manifestations such as fever, cough, nausea, and fatigue presented to the neurosurgery clinic with painful torticollis, fever and reduced range of motion. She had a documented positive rRT-PCR. Similar diagnostic (DCG and CT, Type 3 F-H) and therapeutic (conservative therapy) approaches could not reach to recovery after 3 months. MRI showed C1-C2 rotatory subluxation with narrowing of cervicomedullary junction. Unfortunately, Halo application was not successful. Consequently, we decided to have C1-C2 cervical fusion. This led to complete resolution of subluxation without adverse outcomes.

Discussion

Based on literature, there are different symptoms and signs related to SARS-CoV-2 including dizziness, headache, altered mental state, meningitis, encephalitis, ischemic stroke, hemorrhagic stroke, venous sinus thrombosis, seizure, subarachnoid hemorrhage, neuroimmunology disorders, movement disorders, smell and/or taste impairment, Guillain-Barre syndrome, myasthenia gravis, myositis, rhabdomyolysis, myopathy, neuropathy, and hydrocephalus. As mentioned earlier, there is not any association between this infection and GS in the literature, but we can focus on this possibility [15-18]. Moreover, there is not any history of even mild trauma in these patients. Clinical manifestations of GS were initially described by Charles Bell in 1830. After one century, the French physician Grisel reported 2 cases of C1-C2 subluxation after nasopharyngeal inflammation [19-25]. Probable causes of

higher rate of GS in children are immature bone structure, more horizontal facets, larger synovial folds, adenotonsillar hypertrophy, further hypermobile C1-C2, more laxity of ligaments and elevated rate of infections. Totally, there are 183 reported cases of GS in the literature. Range of patients is between 4.5-14 years. 61.5% of cases were involved due to upper respiratory tract and head/neck infections and 36% were postsurgical ones. Type 1 was the most common F-H grade and most of patients (96%) were treated conservatively [26,27]. As you can see, our cases are matched with these data. Although we believe that COVID-19 spreading in upper airways, related immune response and over-relaxation of ligaments can lead to C1-C2 rotatory subluxation and GS, the exact pathophysiology of this process is unknown.

Conclusion

We conclude that the virus may present with an extensive range of manifestations such as GS and should be considered in children with COVID-19 infection.

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