

World Journal of *Clinical Cases*

World J Clin Cases 2023 May 16; 11(14): 3114-3368



OPINION REVIEW

- 3114 Modernising autism spectrum disorder model engineering and treatment *via* CRISPR-Cas9: A gene reprogramming approach
Sandhu A, Kumar A, Rawat K, Gautam V, Sharma A, Saha L

REVIEW

- 3128 Burden of disability in type 2 diabetes mellitus and the moderating effects of physical activity
Oyewole OO, Ale AO, Ogunlana MO, Gurayah T

MINIREVIEWS

- 3140 Postoperative hypoxemia for patients undergoing Stanford type A aortic dissection
Liu HY, Zhang SP, Zhang CX, Gao QY, Liu YY, Ge SL

ORIGINAL ARTICLE**Case Control Study**

- 3148 Impact of extended nursing model after multi-disciplinary treatment on young patient with post-stroke
Xu XY, Pang ZJ, Li MH, Wang K, Song J, Cao Y, Fang M
- 3158 Changes and significance of serum ubiquitin carboxyl-terminal hydrolase L1 and glial fibrillary acidic protein in patients with glioma
Zhu QH, Wu JK, Hou GL

Retrospective Study

- 3167 Multitrack and multianchor point screw technique combined with the Wiltse approach for lesion debridement for lumbar tuberculosis
Yuan YF, Ren ZX, Zhang C, Li GJ, Liu BZ, Li XD, Miao J, Li JF
- 3176 Clinical features and prognostic factors in 49 patients with follicular lymphoma at a single center: A retrospective analysis
Wu H, Sun HC, Ouyang GF
- 3187 Value of optical coherence tomography measurement of macular thickness and optic disc parameters for glaucoma screening in patients with high myopia
Mu H, Li RS, Yin Z, Feng ZL

Observational Study

- 3195 Comparative study of the clinical efficacy of all-inside and traditional techniques in anterior cruciate ligament reconstruction
An BJ, Wang YT, Zhao Z, Wang MX, Xing GY

- 3204** Positioning and design by computed tomography imaging in neuroendoscopic surgery of patients with chronic subdural hematoma

Wang XJ, Yin YH, Zhang LY, Wang ZF, Sun C, Cui ZM

- 3211** Evaluation of chronic idiopathic tinnitus and its psychosocial triggers

Hamed SA, Attiah FA, Fawzy M, Azzam M

- 3224** Intestinal complications in patients with Crohn's disease in the Brazilian public healthcare system between 2011 and 2020

Sasaki LY, Martins AL, Galhardi-Gasparini R, Saad-Hossne R, Ritter AMV, Barreto TB, Marcolino T, Balula B, Yang-Santos C

Randomized Controlled Trial

- 3238** Effect of non-pharmacological treatment on the full recovery of social functioning in patients with attention deficit hyperactivity disorder

Lv YB, Cheng W, Wang MH, Wang XM, Hu YL, Lv LQ

CASE REPORT

- 3248** Diagnosis of tuberculous uveitis by the macrogenome of intraocular fluid: A case report and review of the literature

Zhang YK, Guan Y, Zhao J, Wang LF

- 3256** Intra-gastric fish bones migrate into the liver: A case report

Dai MG, Zheng JJ, Yang J, Ye B

- 3261** Primary seminal vesicle adenocarcinoma with a history of seminal vesicle cyst: A case report and review of literature

Yao Y, Liu S, He YL, Luo L, Zhang GM

- 3267** Immune checkpoint inhibitor therapy-induced autoimmune polyendocrine syndrome type II and Crohn's disease: A case report

Gao MJ, Xu Y, Wang WB

- 3275** Late-onset mitochondrial encephalomyopathy with lactic acidosis and stroke-like episodes syndrome with mitochondrial DNA 3243A>G mutation masquerading as autoimmune encephalitis: A case report

Wang JW, Yuan XB, Chen HF

- 3282** Metastatic gastric cancer from breast carcinoma presenting with paraneoplastic rheumatic syndrome: A case report

Rech MB, da-Cruz ER, Salgado K, Balbinot RA, Balbinot SS, Soldera J

- 3288** Novel mutation of SPG4 gene in a Chinese family with hereditary spastic paraplegia: A case report

Wang J, Bu WT, Zhu MJ, Tang JY, Liu XM

- 3295** Chronic pulmonary mucormycosis caused by rhizopus microsporus mimics lung carcinoma in an immunocompetent adult: A case report

Guo XZ, Gong LH, Wang WX, Yang DS, Zhang BH, Zhou ZT, Yu XH

- 3304** Idiopathic sclerosing mesenteritis presenting with small bowel volvulus in a patient with antiphospholipid syndrome: A case report
Chennavasin P, Gururatsakul M
- 3311** *Neisseria mucosa* - A rare cause of peritoneal dialysis-related peritonitis: A case report
Ren JM, Zhang XY, Liu SY
- 3317** Rectal prolapse in a 30-year-old bladder stone male patient: A case report
Ding HX, Huang JG, Feng C, Tai SC
- 3323** Successful treatment of veno-arterial extracorporeal membrane oxygenation complicated with left ventricular thrombus by intravenous thrombolysis: A case report
Wang YD, Lin JF, Huang XY, Han XD
- 3330** Successful remimazolam sedation-epidural block in an older patient with severe chronic obstructive pulmonary disease: A case report
Yu JJ, Pei HS, Meng Y
- 3340** *De novo* mutation of NAXE (APOA1BP)-related early-onset progressive encephalopathy with brain edema and/or leukoencephalopathy-1: A case report
Ding L, Huang TT, Ying GH, Wang SY, Xu HF, Qian H, Rahman F, Lu XP, Guo H, Zheng G, Zhang G
- 3351** Iatrogenic atlantoaxial rotatory subluxation after thyroidectomy in a pediatric patient: A case report
Hong WJ, Lee JK, Hong JH, Han MS, Lee SS
- 3356** Bladder metastasis from epidermal growth factor receptor mutant lung cancer: A case report
Jin CB, Yang L
- 3362** Primary rectal mucosa-associated lymphoid tissue lymphoma treated with only endoscopic submucosal dissection: A case report
Lee WS, Noh MG, Joo YE

ABOUT COVER

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Iatrogenic atlantoaxial rotatory subluxation after thyroidectomy in a pediatric patient: A case report

Woo-Joon Hong, Jung-Kil Lee, Jong-Hwan Hong, Moon-Soo Han, Shin-Seok Lee

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Abstract

BACKGROUND

Atlantoaxial rotatory subluxation (AARS) is an uncommon disease with a greater prevalence among children than adults, and it is mostly associated with trauma. Iatrogenic spinal injury accounts for a low percentage of injuries. However, in AARS, 20%-40% of cases are associated with surgery, and 48% are caused by infection. Here, we describe our experience with a case of iatrogenic AARS after general anesthesia.

CASE SUMMARY

A 12-year-old girl presented with right-sided torticollis and cervical motion limit. The patient had undergone thyroidectomy 2 mo ago. Computed tomography revealed AARS with bilateral locked facets. Following the failure of repeated external reduction under general anesthesia, the patient underwent an open surgical reduction. The patient gained atlantoaxial alignment without any complications. Follow-up radiographs showed a normal appearance without instability. The cervical spine of children is more predisposed to injury due to anatomical and biomechanical differences. AARS secondary to infection and surgery is known as Grisel's syndrome, which involves non-traumatic AARS. Several cases of AARS after surgery and other procedures with no evidence of inflammation have been reported. Our experience shows that surgery requiring hyperextension of the neck after general anesthesia should also be included as a risk factor.

CONCLUSION

Surgeons and anesthesiologists should be careful not to excessively extend the neck during pediatric surgery. Moreover, clinicians caring for pediatric patients with recent head and neck procedures must be aware of common AARS presentations.

Key Words: Atlantoaxial joint; Joint subluxation; Adolescent; Grisel's syndrome; Case report

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Core Tip: Atlantoaxial rotatory subluxation (AARS) is a rare condition with a higher prevalence in children, often associated with trauma or infection, and occasionally surgery. This case highlights iatrogenic AARS after general anesthesia and the importance of caution during surgery for AARS in pediatric patients.

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INTRODUCTION

Atlantoaxial rotatory subluxation (AARS) is an uncommon disease with a greater prevalence among children than adults, and it is mostly associated with trauma. Traumatic spinal injuries in children have been reported to be relatively rare, accounting for 1%-10% of all spinal injuries. The incidence of traumatic spinal injuries increases with age, with most injuries being associated with car accidents, followed by falls and sports. Other causes comprise only 3% of spinal injuries, including iatrogenic causes[1,2]. Therefore, iatrogenic spinal injury accounts for only a low percentage of injuries. However, in AARS, 20%-40% of cases are associated with surgery, and 48% are caused by infection (Grisel's syndrome)[3,4]. As mentioned above, AARS has a high prevalence in the pediatric population. The cervical spine is the most vulnerable and commonly injured part of the pediatric spine. Moreover, pediatric spine kinematics is significantly different from that of adults. The facet joints are more horizontally oriented and provide less resistance to rocking and translation between the vertebrae, and the uncinate processes are not developed. Therefore, cervical dislocation after a surgical procedure in pediatric patients is a complication that may occur more often than expected. Nevertheless, only few cases of cervical dislocation due to the surgical position of the patient have been reported. Here, we describe our experience with a case of AARS after thyroid surgery.

CASE PRESENTATION

Chief complaints

The patient complained of right-sided torticollis and cervical motion limit.

History of present illness

A 12-year-old female patient visited our department with right torticollis and cervical motion limit 2 mo ago.

History of past illness

The patient underwent thyroidectomy 2 mo ago. The symptoms appeared after thyroidectomy.

Personal and family history

There was no significant past or family history.

Physical examination

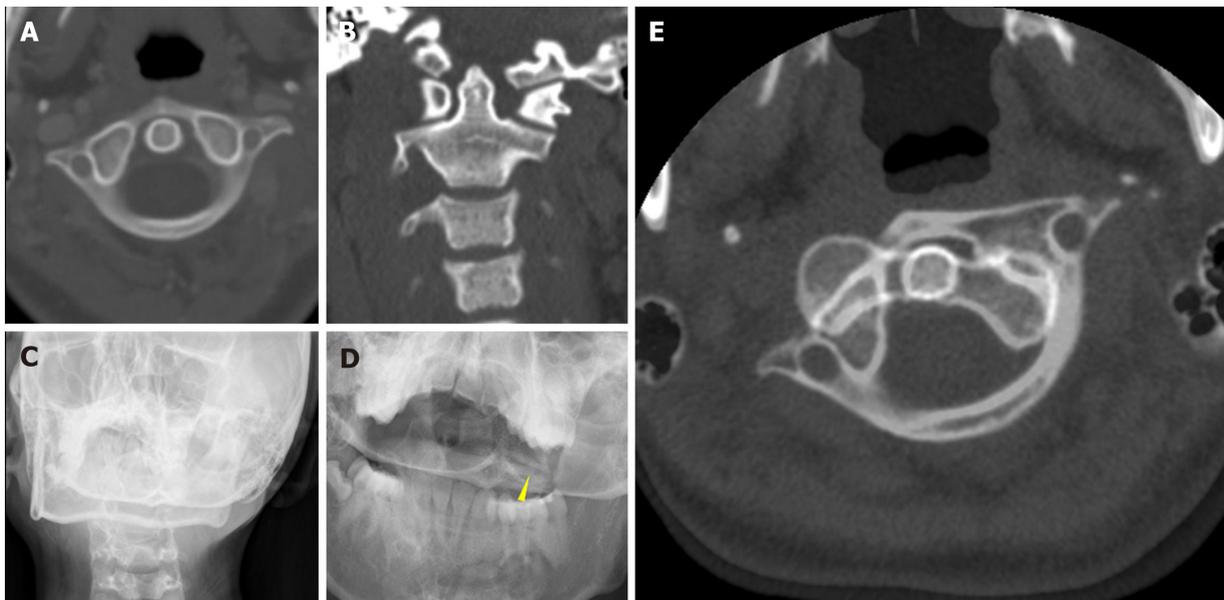
There was no motor weakness or sensory change.

Laboratory examinations

There were no significant laboratory findings.

Imaging examinations

Plain radiographs demonstrated torticollis and the "cock robin" position of the head (Figure 1A and B). Cervical computed tomography (CT) revealed AARS with bilateral locked facets (Figure 1C and D). Neck CT before thyroidectomy showed the normal alignment of the atlantoaxial joint.



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Figure 1 Preoperative imaging. A: Computed tomography of the neck before thyroidectomy showing no subluxation of C1–2; B: Computed tomography coronal view after thyroidectomy showing the asymmetry of the lateral atlantodental interval; C: Plain radiograph showing the “cock robin” position of the head; D: Open mouth view showing the asymmetrical atlantodental interval and narrowing atlantoaxial joints (yellow arrow); E: Computed tomography showing the anterior subluxation of C1 on C2 at the left facet joint and the posterior subluxation of C1 on C2 at the right facet joint.

FINAL DIAGNOSIS

The patient was diagnosed with iatrogenic AARS with bilateral locked facets based on imaging and clinical findings.

TREATMENT

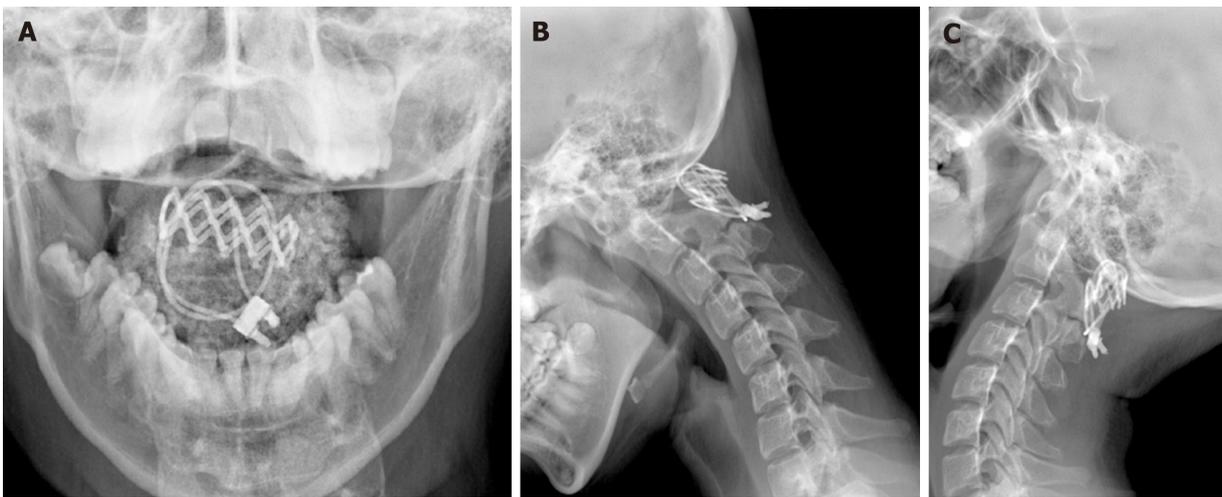
Halter neck traction with 5 pound weight was applied with sedatives and analgesia. Follow-up CT was performed after a week of continuous halter traction; however, AARS remained. Following the failure of repeated external reduction under general anesthesia, we decided on an open surgical reduction. First, through a posterior cervical midline approach, we exposed the C1 and C2 Lamina. Manual reduction was performed by wiring C1, which failed. As there was no other alternative, we sacrificed the C2 root, and the facet subluxation was reduced by manual traction. After confirming the reduction, iliac bone graft and interspinous wiring were performed. The patient gained atlantoaxial alignment without any complications.

OUTCOME AND FOLLOW-UP

After surgery, the patient's torticollis was corrected, and she was discharged without any complications. One-year follow-up dynamic radiographs showed a normal appearance without instability (Figure 2). The patient remained asymptomatic without recurrence until 8 years after surgery.

DISCUSSION

Pediatric AARS can be divided into three main categories depending on their cause: Traumatic, congenital, and inflammatory. Traumatic and congenital causes are related to instability. The cervical spine of children is more predisposed to injury than that of adults due to anatomical and biomechanical differences. In children, the head mass is relatively high, and neck muscles are underdeveloped. The vertebral bodies are anterior wedge-shaped, the facets are angled horizontally, the unciniate process is absent, and the ligaments and joint capsules are highly elastic. Various congenital abnormalities contribute to vertebral dysplasia, leading to instability of the cervical spine (*e.g.*, Klippel-Feil syndrome, Chiari malformation, and Down syndrome). Due to this instability, the pediatric spine is relatively



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Figure 2 Postoperative cervical dynamic radiographs of a stable atlantoaxial joint. A: Open mouth radiograph showing the reduction of the “cock robin” position of the head; B and C: Lateral flexion dynamic radiograph showing C1–2 fusion and no instability.

vulnerable to external forces and is easily damaged by minor trauma.

AARS secondary to infection is known as Grisel’s syndrome. It can be caused by infections, such as in otitis media, viral syndromes, or pharyngitis. Additionally, AARS after head and neck surgery has been occasionally reported in the literature. Symptoms associated with inflammatory reactions after surgery have been observed[3]. The specific mechanism of Grisel’s syndrome is unknown; however, the hematogenous spread of infection from the pharynx to the cervical spine with hyperemia and abnormal relaxation of the atlantoaxial ligaments is accepted as a reasonable explanation[5]. Therefore, AARS rarely appears immediately and is usually detected during postoperative recovery, and laboratory tests show increased inflammatory markers. The most common inflammatory markers measured in clinical practice are C-reactive protein (CRP) and erythrocyte sedimentation rate (ESR). In our case, both CRP and ESR were in a normal range 2 wk after thyroidectomy. In addition, the patient had no specific neurological symptoms other than mild neck discomfort, which could be considered a postoperative change.

Several cases of AARS after surgery and other procedures, such as ventricular abdominal shunt surgery, otitis media surgery, and central venous catheter insertion, have been reported with no evidence of inflammation in pediatric patients[6-8]. The common features of these cases include a pediatric population and a head rotation procedure under general anesthesia. The use of muscle relaxants and general anesthesia loosens the neck muscles, which increases instability in the cervical joint. In addition, pediatric spine kinematics may contribute to the development of AARS. According to Kim *et al*[9], the risk factors for iatrogenic AARS could include pediatric surgery, oropharyngeal inflammation, general anesthesia, and extreme rotation of the head. Nevertheless, we believe that surgery requiring hyperextension of the neck after general anesthesia should also be included as a risk factor.

Early diagnosis is crucial for patients with cervical dislocation, including AARS. Late diagnosis may lead to late management, thereby resulting in invasive treatment. This case required surgical treatment with posterior C1-C2 fusion, resulting in extended hospital stay, additional expenses, and complications. We hope that our experience may help other clinicians prevent such rare and unfortunate incidents from occurring among pediatric patients.

CONCLUSION

Surgeons and anesthesiologists should be careful not to excessively extend the neck during surgery for pediatric patients and ensure that it is in the normal axis even after the operation is complete. Moreover, clinicians caring for pediatric patients with recent head and neck procedures must be aware of the common presentations of AARS.

FOOTNOTES

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