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Original Article

Co-existing odontoid subluxation syringomyelia and vertebral artery abnormalities: a case report

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Abstract: *Background:* Co-existing odontoid subluxation syringomyelia and vertebral artery abnormalities is a rare condition in the spinal cord, and the therapeutic approach of this disease has been also rarely reported. Hence, a case about co-existing odontoid subluxation syringomyelia and vertebral artery abnormalities is reported here. *Methods:* A male patient who suffered with limited head and neck functions is described in the current study. X-ray and MRI examinations for cervical vertebra were performed to examine the conduct physical examination. Furthermore, basic personnel characteristics were measured, such as speech, behavior, power, and state of mind. After these examinations, the patient received surgery treatment with a prone position, and the outcomes were checked using CT imaging. *Results:* According to patient, he had suffered from inability and pain in left lower limb accompanied with limited locomotive for more than 4 months. Further physical examinations showed that patient had a sober mind and powerful shrug with the obvious hoarse voice, blear speech, soft neck, and limited limb activity. Additionally, X-ray and MRI results showed obvious occipital-cervical deformity, odontoid subluxation and syringomyelia. During surgery, an obvious insertion and merge of lepospondylous arcus posterior was found in foramen magnum posterior, and no sever events had been observed. *Conclusion:* Limited neck and limbs activity were the main syndromes of the co-existing of odontoid subluxation syrignomyelia and vertebral artery abnormalities, and bleeding might be an important risk factor and should be significantly considered during surgery treatment.

Keywords: Odontoid subluxation, syringomyelia, vertebral artery abnormalities, spine disease, surgery

Introduction

Syrignomyelia is a common atlanto-occipital malformation disease in clinic [1], but co-existing odontoid subluxation syrignomyelia and vertebral artery abnormalities are still rarely reported. With the deformity of vertebral artery, the risks during surgery therapy were significantly increased. In the current study, a case of a patient that suffered with co-existing odontoid subluxation syrignomyelia and vertebral artery abnormalities is reported. Furthermore, the surgical method used for this disease was also provided.

Case report

Clinical primary information

A 33-year old male patient, who had a limited head and neck functions came to our clinical search therapy for inability and pain in left lower limb accompanied with limited locomotion for more than 4 months. The patient reported no fever, sensory disturbance, or gatism since these syndromes occurred. Before this attendance, he received X-ray and Magnetic Resonance Imaging (MRI) examinations for cervical vertebra in the Second Hospital of Shijiazhuang, Hebei, China, and occipito-cervical deformity, odontoid subluxation, and syringomyelia were found. The data were analyzed using Living Image 3.1 software (Caliper Life Sciences). The patient received further therapy with the additional examinations being performed in our hospital. The examination results showed that although he had a sober mind and powerful shrug, obvious hoarse voice, blear speech, and soft neck were identified. For the upper limbs, big and small thenar muscles of both hands were obviously atrophic, and musculus biceps and triceps brachii of two flanks showed marked hyperreflexia. For the lower

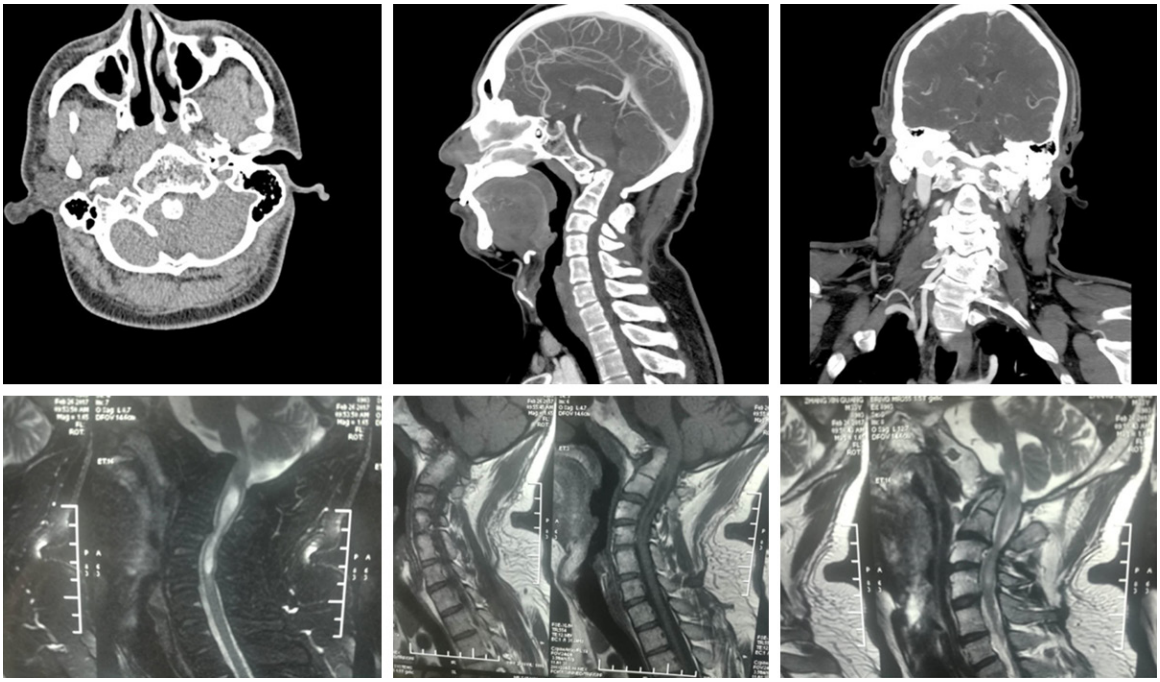

Figure 1. Images provided by CT before surgery.

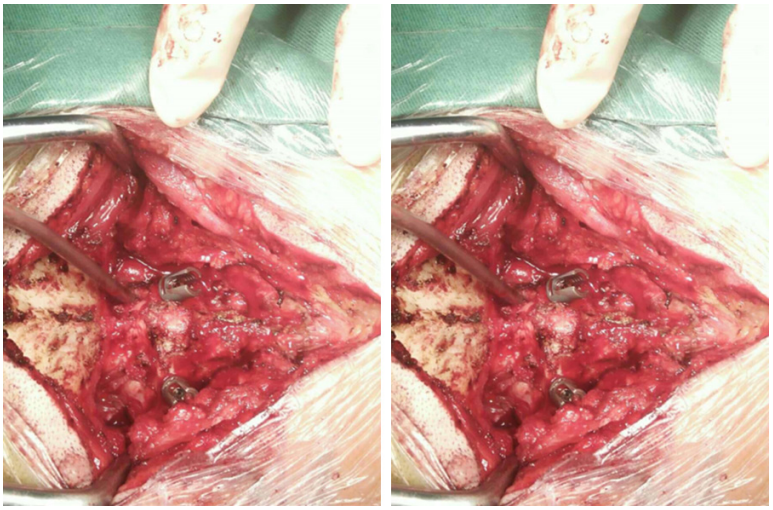

Figure 2. Photos during surgery.

limbs, knee jerks of both flanks were hyperreflexia, and ankles clonus, Hoffman and Babinski signs were positive. Furthermore, hypermyotonia and V stage muscle force were identified in all four limbs along with unstable tread. X-ray and MRI examinations revealed obvious developmental malformation in craniocervical, syncretic atlanto-occipital odontoid subluxation and syringomyelia in the head and neck (**Figure 1**).

Surgery treatment

For treatment, patients were general anesthesia and prepared for surgery treatment with a prone position. With alcohol disinfects, a 10-cm incision was performed with a posterior approach at neck, and a traction with 16 kg was made. For this incision, skin was cut apart, and spinous process was first uncovered. Followed by this, the muscle around spinous process was apart, and vertebra, inion and occipital bone were exposed. Then, an obvious insertion and merge of lepospondylous arcus posterior in foramen magnum posterior was found, and after open lepospondylous arcus posterior about 3 cm, remarkable merge was identified in 2nd and 3rd neck spinous process. Both flank zygopophyses were carefully dissected to expose the 1st-2nd joints. For the right side, an obvious abnormality was identified in the zygopophysis between 1st-2nd joints, and a malformation was identified in vertebral artery in the right side. For the left side, the

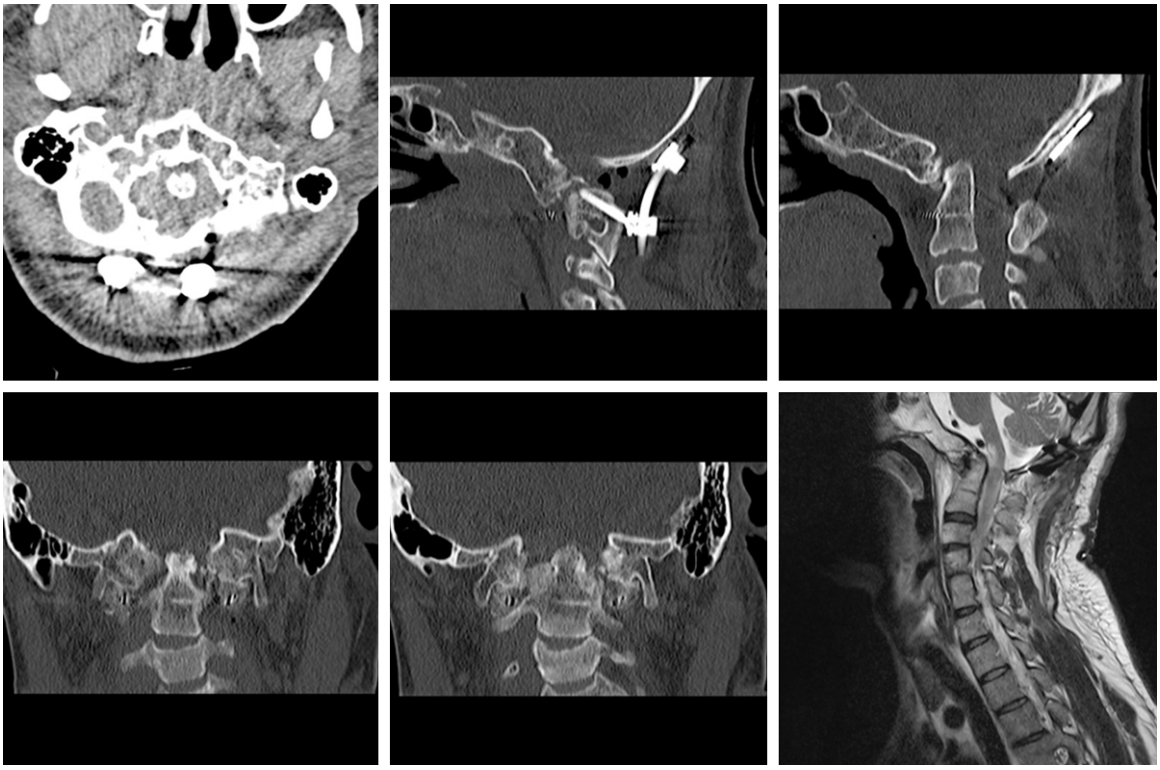

Figure 3. Images provided by CT and MRI after surgery.

other was not revealed. The 1st and 2nd joints were fully debonding with rimer through foramen magnum pathway. Based on these results, a suitable height Cage was selected. After filled up with right ilium, cages were placed between 1st and 2nd of both flank joints (**Figure 2**). Moreover, at the bone ridge under inion, 2 occipital nails were used to fix the occipital plate. In addition, a pedicle screw was inserted at the both flank vertebral arch, and pitman was bent and fixed. Finally, hemostasis and sutured layer by layer were performed. The surgery outcomes were showed by CT images (**Figure 3**).

Discussion

Syringomyelia is a common neurological disease, and usually associated with conditions of spinal cord injury, spinal tumor and Chiari I malformation [2]. Specially, there are about 50-76% of syringomyelia occurred along with Chiari I malformation [3]. However, co-existing odontoid subluxation syrignomyelia and vertebral artery abnormalities is still rarely reported. Although surgery is still the first choice of syringomyelia treatment, the surgery outcomes are still not satisfied. For the co-existance of vertebral artery malformation, the bleeding during surgery also performs a more difficult in the treatment.

Atlanto-occipital odontoid subluxation is an infrequent atlantoaxial subluxation in clinic, which is a serious condition that can result in sudden death owing to cervical cord compression [4]. Parke et al. have demonstrated that the upper cervical spine might be affected by the congestion of effusion from the lymph and veins in the posterior pharynges [5]. For the odontoid subluxation, the odontoid will be significant retro-position and result in an increased suppression in the veutro of brainstem [6, 7]. Therefore, this injury in spine will also increase the ataxia and muscular tension, and impair the progressive of muscles [8, 9].

In the current study, a case of co-existing of odontoid subluxation syrignomyelia and vertebral artery abnormalities was reported. According to radiography images, remarkable odontoid subluxation syrignomyelia was identified and confirmed by surgery findings. A vertebral artery abnormity was uncovered. Considering

these conditions, single posterior fossa decompression was not suitable owing to the increasing unstable of atlanto-occipital structure, which would not improve symptoms, but also could aggregate them. Furthermore, even pedicle screws were utilized to reinforce the stability of atlanto-occipital structure after posterior fossa decompression, the odontoid was not able to be restored, and the brainstem compression could not be alleviated. Therefore, combined with the posterior fossa decompression, the 1st and 2nd joints were fully debonding, and cages were inserted to promote the restoration of odontoid and alleviate compression of brainstem. Rahul Kadam et al. have reported a case of atlanto-axial subluxation with occipotal C2 fusion, and found that the fusion performed at occiput C2 level was enough to release the pressure of the cord [10]. However, whether this method appropriate for patients suffered with vertebral artery abnormalities is still needed further investigation.

In conclusion, co-existence of odontoid subluxation syrignomyelia and vertebral artery abnormalities is rarely reported in spinal related disease. The therapies for this disease are still needed further exploration, and bleeding may be an important risk factor and should be significantly considered.

Disclosure of conflict of interest

None.

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References

[1] Samdani AF, Hwang SW, Singla A, Bennett JT, Ames RJ, Kimball JS. Outcomes of patients with syringomyelia undergoing spine deformity surgery: do large syrinxes behave differently than small? Spine J 2017; 17: 1406-1411.

[2] Shields CB, Zhang YP, Shields LB. Post-traumatic syringomyelia: CSF hydrodynamic changes following spinal cord injury are the driving force in the development of PTSM. Handb Clin Neurol 2012; 109: 355-67.

[3] Suchomel P. Expert's comment concerning grand rounds case entitled "syringomyelia with irreducible atlantoaxial dislocation, basilar invagination and Chiari I malformation" (by Shenglin Wang, Chao Wang, Ming Yan, Haitao Zhou, Liang Jiang). Eur Spine J 2010; 19: 367-9.

[4] Hasegawa A, Yagi M, Takemitsu M, Machida M, Asazuma T, Ichimura S. Atlantoaxial subluxation after pyogenic spondylitis around the odontoid process. Case Rep Orthop 2015; 2015: 861403.

[5] Parke WW, Rothman RH, Brown MD. The pharyngovertebral veins: an anatomical rationale for Grisel's syndrome. J Bone Joint Surg Am 1984; 66: 568-74.

[6] Ogata T, Morino T, Hino M, Miura H. Cervical myelopathy caused by atlantoaxial instability in a patient with an os odontoideum and total aplasia of the posterior arch of the atlas: a case report. J Med Case Rep 2012; 6: 171.

[7] Wu X, Ming L, Chang Y, Sun Z. Accidental or linked: separated odontoid process fused to the enlarged anterior arch of the atlas associated with atlantoaxial subluxation in a Kashin-Beck disease patient. Eur Spine J 2017; 26 Suppl 1: 85-89.

[8] Hatzantonis C, Muquit S, Nasto LA, Mehdian H. Congenital defects of C1 arches and odontoid process in a child with Down's syndrome: a case presentation. J Craniovertebr Junction Spine 2016; 7: 115-7.

[9] Dohzono S, Suzuki A, Koike T, Takahashi S, Yamada K, Yasuda H, Nakamura H. Factors associated with retro-odontoid soft-tissue thickness in rheumatoid arthritis. J Neurosurg Spine 2016; 25: 580-585.

[10] Kadam R, Bauva V, Shah K, Yadav S. Atlantoaxial subluxation with cervical myelopathy operated with occipital C2 fusion: a case report. J Med Res Innov 2017; 1: 4-7.