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CASE REPORT

Regression of intervertebral disc calcification combined with ossification of the posterior longitudinal ligament: A case report

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Abstract

BACKGROUND

Intervertebral disc calcification (IDC) combined with ossification of the posterior longitudinal ligament (OPLL) in cervical discs is rarely reported. This case study presents a rare case of IDC combined with OPLL in the C2-C3 segment.

CASE SUMMARY

Here, we present a case of a 6-year-old Asian boy with severe neck pain and stiffness. Physical examination showed no neurological or other abnormalities. Digital radiography and computed tomography (CT) revealed a calcified intervertebral disc and OPLL at the C2-C3 vertebrae. The spinal canal compromise at C2-C3 was approximately 50% on magnetic resonance imaging. The final diagnosis was IDC combined with OPLL. We applied a neck brace for the patient to protect the neck. The patient's neck pain and stiffness recovered significantly within approximately 3 wk. At the 3 mo follow-up, the follow-up CT showed resolution of the ossified intervertebral disc herniation, and a small amount of calcification and slight OPLL remained at the involved segment.

CONCLUSION

IDC combined with OPLL is a relatively rare condition in children. However, the majority of patients could have a favorable outcome, and the ossified mass in the canal would be spontaneously resolved with conservative therapy.

Key Words: Intervertebral disc calcification; Ossification of the posterior longitudinal ligament; Diagnosis; Treatment; Case report

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Core Tip: Intervertebral disc calcification (IDC) with ossification of the posterior longitudinal ligament (OPLL) is rare. At present, there is no consensus on the diagnosis and treatment of the disease. If the disease is not recognized, it is easily misdiagnosed and leads to unnecessary treatment. In addition, there are few reports of upper cervical IDC with OPLL, which may limit doctors' awareness of the disease. This case study presents a rare case of IDC combined with OPLL in the C2-C3 segment to help diagnose and treat the disease.

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INTRODUCTION

Intervertebral disc calcification (IDC) with ossification of the posterior longitudinal ligament (OPLL) is rare. To date, only 8 cases of IDC with OPLL in children have been reported [1-7]. Of the 8 reported cases, 6 were in the cervical spine and 2 were in the thoracic spine. Finally, the clinical symptoms of all patients disappeared after conservative treatment. However, according to imaging evaluation, 4 patients recovered completely [2,4,6,7], 1 patient had persistent mild OPLL[3], 2 patients had persistent mild IDC[4,5], and 1 patient had progressive IDC1. All reported patients had neck pain or back pain to varying degrees due to the IDC and OPLL extending into the spinal canal, compressing the spinal cord or nerve root, and all went to the hospital with or without neurological defects. This does not exclude the possibility that some patients with IDC with OPLL have not been identified and reported due to their absence of symptoms. At present, there is no accepted treatment standard for this disease and conservative treatment is usually the first choice this kind of patient[3,6]. According to the results of a literature search, there are few reports of upper cervical IDC with OPLL, which may limit doctors' awareness of the disease.

This case study presents a rare case of IDC combined with OPLL in the C2-C3 segment to help diagnose and treat the disease.

CASE PRESENTATION

Chief complaints

A 6-year-old Asian boy complained of severe neck pain and limited neck movement for 3 d.

History of present illness

During a handstand performed while hip-hop dancing 3 d ago, the patient accidentally landed on the ground on his neck. He subsequently presented with severe neck pain and stiffness. The symptoms continued to worsen without improvement, so his parents took him to the spine department.

History of past illness

The patient had no significant medical history.

Personal and family history

The patient had no remarkable personal and family history.

Physical examination

Physical examination showed no neurological or other abnormalities.

Laboratory examinations

Laboratory examinations showed no significant abnormalities.



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Imaging examinations

Digital radiography (DR) showed a dense projection of the intervertebral disc between the second and third cervical vertebrae (Figure 1). Then computed tomography (CT) further demonstrated cervical disc calcification and partial OPLL in the C2-C3 segment (Figure 2). Magnetic resonance imaging (MRI) of the cervical spine revealed cervical disc herniation, resulting in a certain degree of spinal cord compression (Figure 3). The degree of spinal canal compromise was defined as a percentage that was calculated by dividing the smallest sagittal diameter of the spinal canal at the level of the disc herniation by the mean sagittal diameter at adjacent levels cephalad and caudad to the herniated disc as seen on CT or MRI[8]. According to the rough estimation by MRI, the spinal canal compromise at C2-C3 was approximately 50%.

FINAL DIAGNOSIS

According to the above findings, IDC combined with OPLL was diagnosed.

TREATMENT

Considering that his symptoms were not very serious and that this disease was benign and self-limited, the patient was treated with a neck brace for protection.

OUTCOME AND FOLLOW-UP

The patient reported significant relief in his neck pain after the cervical spine was immobilized. Then the patient was discharged from the hospital, and his parents were instructed to take good care of him to avoid new accidental falls and any other injuries. The patient's neck pain and stiffness recovered significantly within approximately 3 wk. At the 3 mo follow-up, the patient felt no discomfort. Follow-up CT showed resolution of the ossified intervertebral disc herniation, and only a small amount of calcification and slight OPLL remained (Figure 4). The patient and his parents expressed satisfaction with the treatment and expressed their sincere gratitude to the doctors.

DISCUSSION

IDC in children is a rare spinal disorder that was first reported in 1924[9]. OPLL is usually considered a degenerative cervical or thoracic pathology in adults and was first reported in 1838[10]. IDC combined with OPLL in pediatric patients is much less frequent, and to date only 8 cases have been published[1-7].

The patients in the previously reported cases were two men and six women, aged 6years-old to 11-years-old. In general, the injury occurs in a single segment of the disc, and a few cases have also involved two parts of the disc[1,4,6,7]. In the imaging examinations of the reported cases, abnormal signs of intervertebral discs were seen, and the masses penetrated into the spinal canal and compressed the spinal cord. However, masses in the spinal canal are not easily identified as a calcified disc herniation or OPLL. In addition, rarely herniated calcified discs protrude laterally into the transverse foramen[11]. Therefore, if the disease is not recognized, it is easily misdiagnosed and leads to unnecessary treatment.

The exact cause of IDC accompanied by OPLL is still unclear. However, trauma is often considered an inducer because some children's disc calcifications occur before trauma[12,13]. Beluffi et al[9] suggested that there are two types of IDC: Symptomatic and asymptomatic. Matsunaga et al[10] studied the natural course of OPLL and found that 90% of OPLL patients had clinical symptoms; 23% of OPLL patients had a history of neck trauma. Therefore, we presume that IDC may be in a stable period with no clinical symptoms, and due to the presence of asymptomatic IDC, OPLL can be induced by herniated IDC in an accidental cervical trauma. Disc calcification leads to a decrease in T2-weighted signal strength on MRI, which is considered an imaging manifestation of disc degeneration[14]. In children, calcification mainly affects the nucleus pulposus [15]. The presence of a calcified nucleus pulposus may affect the

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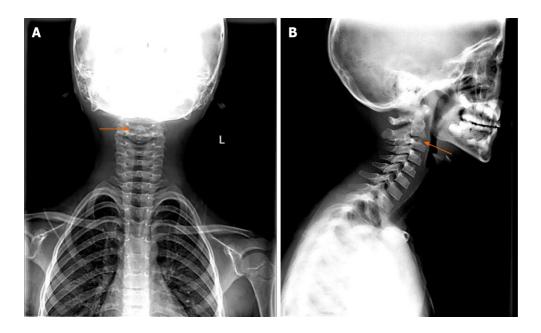


Figure 1 Digital radiography on admission. A and B: Digital radiography showing a dense projection of the intervertebral disc between the second and third cervical vertebrae.

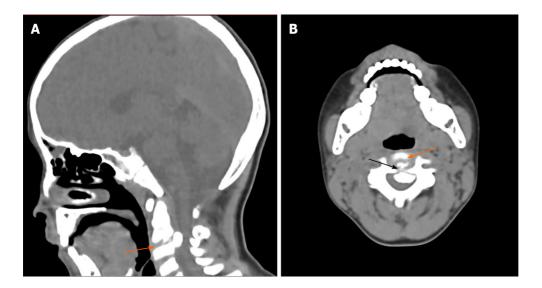


Figure 2 Computed tomography on admission. A: Sagittal computed tomography revealing cervical disc calcification (orange arrow); B: Axial computed tomography further demonstrating cervical disc calcification (orange arrow) and partial ossification of the posterior longitudinal ligament (black arrow).

stress balance of the affected intervertebral disc, which leads to the nucleus pulposus protruding and compressing, stimulating and causing an inflammatory reaction within the posterior longitudinal ligament. Chronic inflammatory stimulation leads to ossification and further aggravation of the compression. Any trauma to the area is an important inducer that leads to further backward penetration of the intervertebral disc into the spinal canal, compression of the spinal cord or nerve root, and corresponding clinical symptoms. The increase in erythrocyte sedimentation rate indicates that the disease is related to inflammation[16]. During conservative treatment, due to the inflammatory reaction, the blood supply to the lesion increases, which provides a basis for the spontaneous disappearance of the calcification in the herniated disc and the ossification in posterior longitudinal ligament. Simple IDC disappears in 3 to 60 mo, with an average of 15 mo[8]. However, after 9 years of follow-up, Du et al[4,12] found that residual IDC persisted and the height of the affected intervertebral space had become narrow, indicating that residual IDC may not subside spontaneously with time.



Figure 3 Magnetic resonance imaging on admission. Magnetic resonance imaging showing substantial spinal stenosis and spinal cord compression (orange arrow).

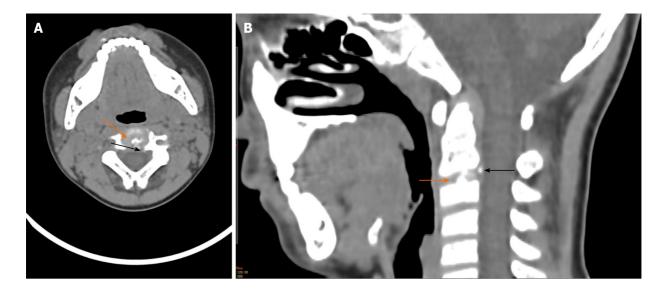


Figure 4 Computed tomography at the 3 mo follow-up. A and B: Follow-up computed tomography showing that only a small amount of calcification (orange arrow) and slight ossification of the posterior longitudinal ligament (black arrow) remain.

The main symptoms of IDC accompanied by OPLL are neck pain, stiffness, torticollis, and neck movement limitation. Physical examination can often reveal neck muscle spasms, tenderness at the lesion site, and limited neck movement. If there is nerve root or spinal cord compression, there may be corresponding signs of nervous system defects[12,15]. In the reported cases, the most frequently reported symptoms were neck pain. Similarly, neck pain was the main symptom in this case. Ninety-five percent of OPLL in adults is clinically symptomatic, with different degrees of myelopathy and radiculopathy[10]. However, child OPLL is different from adult OPLL. Although the imaging manifestations are very serious, the spinal canal compromise at C2-C3 could be approximately 50% but the clinical symptoms are not serious, and few children have neurological defects[2,4].

At present, there is no consensus on the diagnosis of the disease. We suggest that the following characteristics may be helpful for a preliminary diagnosis: local pain in the cervical or thoracic region, full or limited cervical motion, and high-density areas and ossified mass on DR and CT. MRI can be recommended to identify compression of the spinal cord. Generally, imaging findings are the main basis for diagnosis[15]. However, for this disease, there exists a possibility that the imaging performance and

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clinical symptoms are inconsistent[1,2,5-7,12].

At present, it is believed that conservative treatment including bed rest, brace braking, traction, and drugs should be prioritized[1-7]. However, traction has an uncertain outcome in patients with compressed cervical spinal cord because it has the possibility of aggravating the spinal cord compression[17]. If the calcification protrudes into the spinal canal, traction is contraindicated given the possibility of aggravating the spinal cord compression. For patients with no or mild neurological dysfunction, even without intervention, recovery is still possible within several weeks [2]. Patients receiving conservative treatment should focus on avoiding injury and must be closely followed up and return to the hospital immediately in case of aggravation of nerve compression symptoms. The symptoms could be totally relieved with conservative therapy, and the ossified mass can automatically regress in 3-6 mo [1-3,5,6], but in some cases, as much as 1-2 years may be needed [4,7]. When the patient has severe neurologic deficits and the disease continuously progresses, surgical intervention should be considered[18].

CONCLUSION

IDC combined with OPLL is rare, but we must be aware of its presence. The exact etiology is still unclear. Conservative treatment is usually effective. Clinical symptoms may subside in weeks to months without recurrence. The majority of patients may have a favorable outcome, and the ossified mass in the canal would be spontaneously resolved with conservative therapy in 3 mo to 2 years. However, slight IDC or OPLL may remain for a long time.

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