# A pediatric case of single-level idiopathic cervical intervertebral disk calcification with symptom relapse 1 year after initial onset


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## Title

A pediatric case of single level idiopathic cervical intervertebral disc calcification with symptom relapse one year after initial onset


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## Title

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## Abstract

BACKGROUND: To date there has been only one reported case of the symptom relapse of pediatric idiopathic intervertebral disc calcification (PIIDC), as described by Yoon et al. in 1987, who reported symptom relapse in a patient with multi-level PIIDC. Thus, symptom relapse in patients with single level PIIDC have not been reported.

METHODS: We report here a case of single level PIIDC with symptom relapse 1 year after the initial onset.

RESULTS: The patient was a 7-year-old girl who developed cervical pain and fever to 38°C without an obvious cause. CT revealed calcification in the C 4/5 intervertebral disc space and in the epidural space at the C3-5 vertebral levels. The patient was diagnosed with PIIDC and began treatment with oral NSAIDs. Both

cervical pain and fever gradually improved and resolved in approximately 1 week. CT obtained 6 months after the initial onset showed calcifications localized in the posterior area of the C4/5 intervertebral disc space and reduced epidural calcifications which had nearly resolved. One year after the initial onset, the patient developed similar symptoms. CT revealed an enlarged calcified lesion in the epidural space. Thus, the patient was diagnosed with symptom relapse of PIIDC. Although there was enlargement of calcifications in the epidural space, there were no calcifications involving the intervertebral disc at the time of relapse. The patient was treated conservatively. Follow-up CT revealed that the lesion resolved with time.

CONCLUSIONS: This report described a patient with single level PIIDC and symptom relapse 1 year after the initial onset. In the case presented herein, calcifications of the intervertebral space had extruded into the epidural space, thus causing a symptom relapse. The patient was treated conservatively at the initial onset and at the time of relapse. The symptoms improved both times. Although patients with single level PIIDC usually have an uneventful clinical course, it is necessary to be mindful of potential symptom relapse.

## Introduction

Pediatric idiopathic intervertebral disc calcification (PIIDC) is a rare disease. PIIDC often involves in the cervical vertebrae, and clinical symptoms include cervical pain and fever. In a review of the literature, Tsutsumi et al.[1] identified 65 cases of PIIDC reported in the English-language literature since 1990. These cases had uneventful follow-ups and no symptom relapse. Based on our literature search,

symptom relapse was only observed in the 1987 report by Yoon et al[2]. Specifically, symptom relapse was reported in a patient with multi-level PIIDC. Thus, symptom relapse in patients with single level PIIDC has not been reported. We report here a case of single level PIIDC with symptom relapse 1 year after the initial onset.

## Case report

The patient was a 7-year-old girl with a non-contributory medical history.

## Initial onset

The patient developed cervical pain and fever to 38°C without an obvious cause. The patient had unremitting severe cervical pain and was unable to move her neck; she moved her body with difficulty. There was pain at rest and headaches. There were no neurologic symptoms involving the upper or lower limbs. A plain x-ray showed calcifications in the C4/5 intervertebral disc space (Fig. 1A). Computed tomography (CT) revealed calcifications in the C4/5 intervertebral disc space and in the epidural space at the C3-5 vertebral levels (Fig. 1B). T1- and T2-weighted magnetic resonance imaging (MRI) showed a low-density area in the C4/5 intervertebral disc and a low-density area around the lesion in the epidural space; however, the interior of the lesion had a low signal on T1 and a high signal on T2 (Figs. 1 C and D). There was mild spinal cord compression. Blood test results indicated a mild inflammatory response with a white blood cell (WBC) count of 8600/μl and a C-reactive protein (CRP) level of 1.43mg/l, but no other abnormalities.

The patient was diagnosed with PIIDC and began treatment with oral non-steroidal anti-inflammatory drugs (NSAIDs). Both cervical pain and fever gradually improved and resolved in approximately 1 week. There was no limitation to daily activities. No calcifications were observed at C4/5 on plain X-rays obtained

6 months after the initial onset (Fig. 2A). CT showed calcifications localized to the posterior area of the C4/5 intervertebral disc space and reduced epidural calcifications, which had nearly resolved (Fig. 2B).

### Symptom relapse

One year after the initial onset, the patient developed similar symptoms and had cervical pain and fever to 37.8°C without an obvious cause. Cervical pain was as severe as at the time of initial onset. A plain X-ray showed no calcifications of the intervertebral disc (Fig. 3A), but CT revealed an enlarged calcified lesion in the epidural space (Fig. 3B). The blood test results were consistent with an inflammatory response, with a WBC count of 10200/μl and a CRP level of 2.56 mg/l.

As at the time of the initial onset, there were no neurologic symptoms involving the upper or lower limbs, and treatment with oral NSAIDs was begun. Both cervical pain and fever improved again after 1 week, and the patient had no limitation of daily activities. Six months after symptom relapse, CT showed calcifications which had nearly completely resolved (Fig. 4). It has now been 2 years since the initial onset of PIIDC, and there are no limitations of daily activities or range of motion.

### Discussion

To date there has been only one reported case of the symptom relapse of PIIDC, as described by Yoon et al.[2] in 1987, who reported symptom relapse in a patient with multi-level PIIDC. Therefore, the case presented herein is the first report of symptom relapse in a patient with single level PIIDC. Thus, much is still unknown about the pathogenesis of such relapses. Yoon et al. reported on the changes in the lesions observed in plain X-rays; however, CT can generally depict

calcified lesions more clearly than plain radiography. In our case, CT was used to examine the changes in the lesions. There were calcifications involving the intervertebral disc and the epidural space at the time of initial onset. Six months later, the calcifications only remained in the posterior area of the intervertebral disc, and calcifications in the epidural space had nearly resolved. Upon symptom relapse, the calcifications in the epidural space were enlarged; however the lesions resolved with time.

In the case presented herein, imaging showed that the epidural lesion was enlarged at the time of symptom relapse, after the lesion had decreased in size 6 months after the initial onset. Thus, the patient was diagnosed with symptom relapse of PIIDC. Although there was enlargement of calcifications in the epidural space, there were no calcifications involving the intervertebral disc at the time of relapse. Therefore, it is possible that calcifications of the intervertebral space had extruded into the epidural space, suggesting that symptoms can recur if calcifications of the intervertebral disc persist during follow-up.

The cause and pathogenesis of PIIDC are not clearly known. There have been reports suggesting that mild trauma and infection are potential causes of PIIDC[3-6]. The patient presented herein had no episodes of trauma at the initial onset or at the time of symptom relapse. The pain, fever, and leukocytosis all suggest acute discitis. PIIDC differs from discitis in several ways[7]: 1) intervertebral disc calcification is exceedingly rare after discitis[8]; 2) radiographic changes are common in discitis, such as erosion of adjacent endplates and vertebral bodies and disc space collapse, which do not occur in PIIDC; and 3) in PIIDC, spontaneous improvement has been reported without antibiotic treatment[3]. For these reasons,

this case differed from discitis.

The etiology of PIIDC is still a matter of debate. Smith et al.[9] and Oga et al.[10] concluded that inflammation of the intervertebral disc is associated with the pathogenesis of PIIDC. However, Swick et al. [11] claimed that there is no association between PIIDC and inflammation or angiogenesis. In our case, CT showed slight calcifications in the epidural space at the initial onset, and MRI revealed the interior of the lesion with a high signal on T2. These findings suggest that there were calcifications, as well as other changes involving swelling and inflammatory cells.

The symptoms of PIIDC include cervical pain, neurologic symptoms, and fever[1]. Our patient presented with chief complaints of severe cervical pain and fever at the initial onset and at the time of relapse. The level of pain was similar both times. The patient had a lesion in the epidural space but there were no neurological symptoms at the initial onset or relapse.

Neurologic symptoms due to epidural calcifications are observed in 20% of PIIDC patients[1]. There have been few reports in which surgery was performed at the initial onset of PIIDC in patients with severe radiculopathies, muscle weakness, and myelopathic symptoms[12-16]; however, the majority of cases are treated conservatively and have good outcomes[1,17]. Therefore, conservative management is currently the mainstay of treatment for PIIDC, except for uncommon cases presenting with acutely progressive and severe neurologic impairment[1]. Our patient did not have neurologic symptoms. Therefore, the patient was treated conservatively at the initial onset and at the time of relapse, and the symptoms improved in 1 week both times. Both the clinical condition and imaging findings

were good at the time of follow-up. If a patient does not have a severe radiculopathy, muscle weakness, or myelopathic symptoms, our experience suggests that conservative treatment can be used with symptom relapse of PIIDC.

Patients with PIIDC usually have an uneventful clinical course[1], but it is necessary to be mindful of potential symptom relapse. Yoon et al.[2] reported that 1 patient had 2 relapses within 3 years of initial onset. Our patient had a 2-year follow-up since the initial onset and will need continued follow-up to monitor for relapse.

## Conclusion

This report described a patient with single level PIIDC and symptom relapse 1 year after the initial onset. In the case presented herein, calcifications of the intervertebral space had extruded into the epidural space, thus causing a symptom relapse. The patient was treated conservatively at the initial onset and at the time of relapse. The symptoms improved both times. Although patients with single level PIIDC usually have an uneventful clinical course, it is necessary to be mindful of potential symptom relapse.

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**Figure legends**

Figure 1.

Images at initial on set

A: PlainX-ray, B: CT, C: T1- weighted MRI, D: T2-weighted MRI

Figure 2.

Images obtained 6 months after the initial onset

A: Plain X-ray, B: CT

Figure 3.

Images at the time of symptom relapse

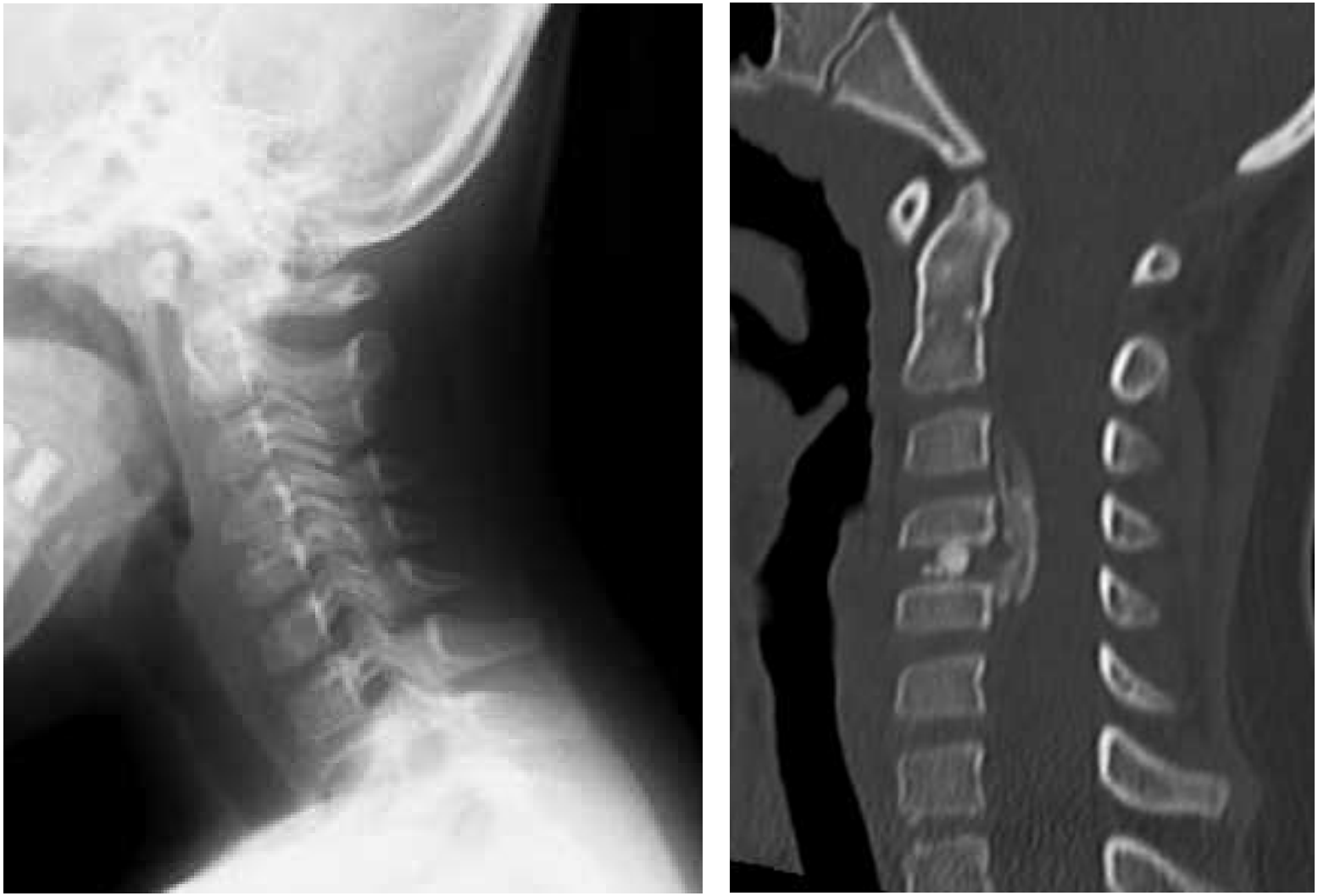
A: Plain X-ray, B: CT

Figure 4.

Images obtained 6 months after symptom relapse

A: Plain X-ray, B: CT

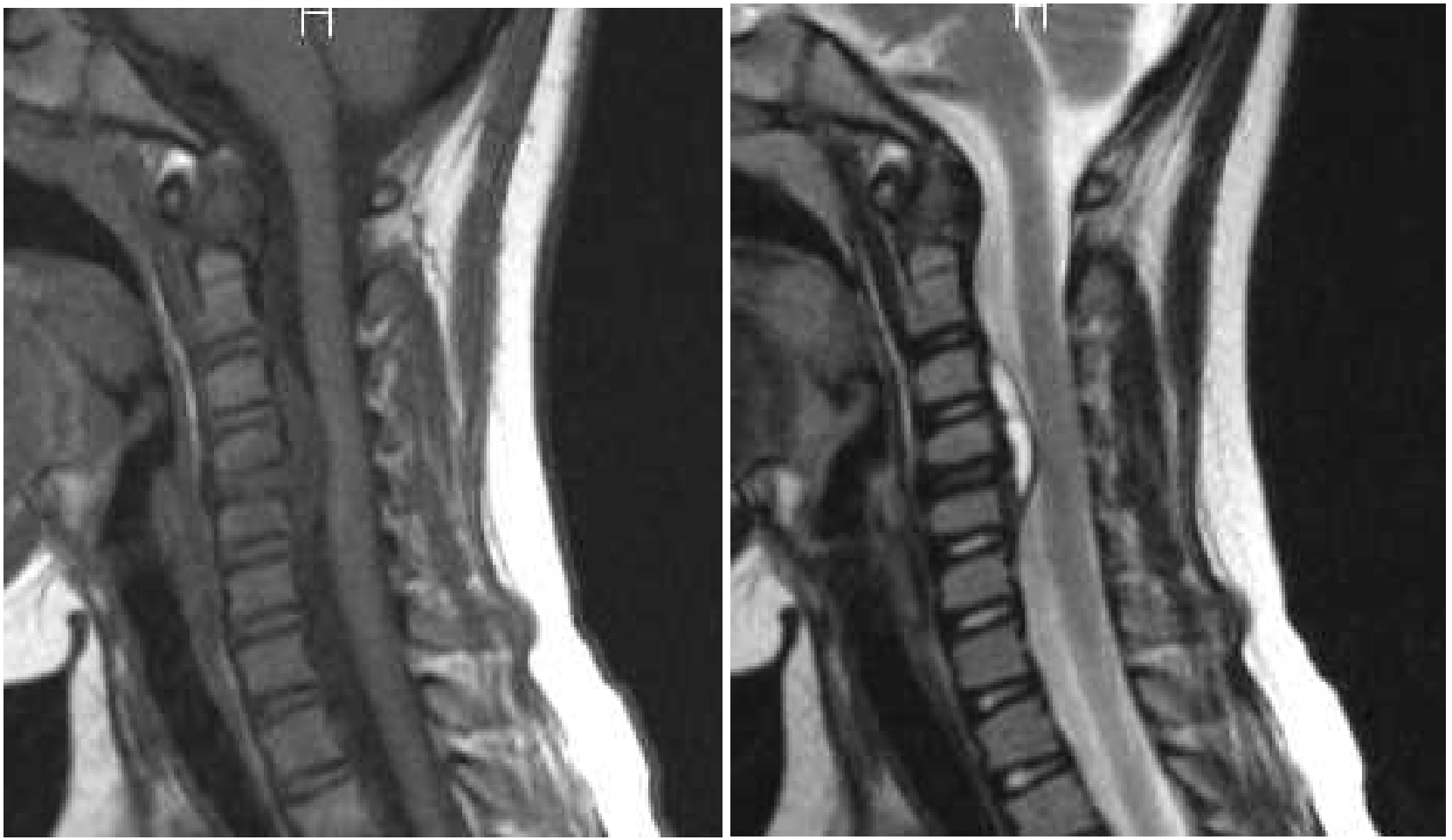
Fig 1.



A

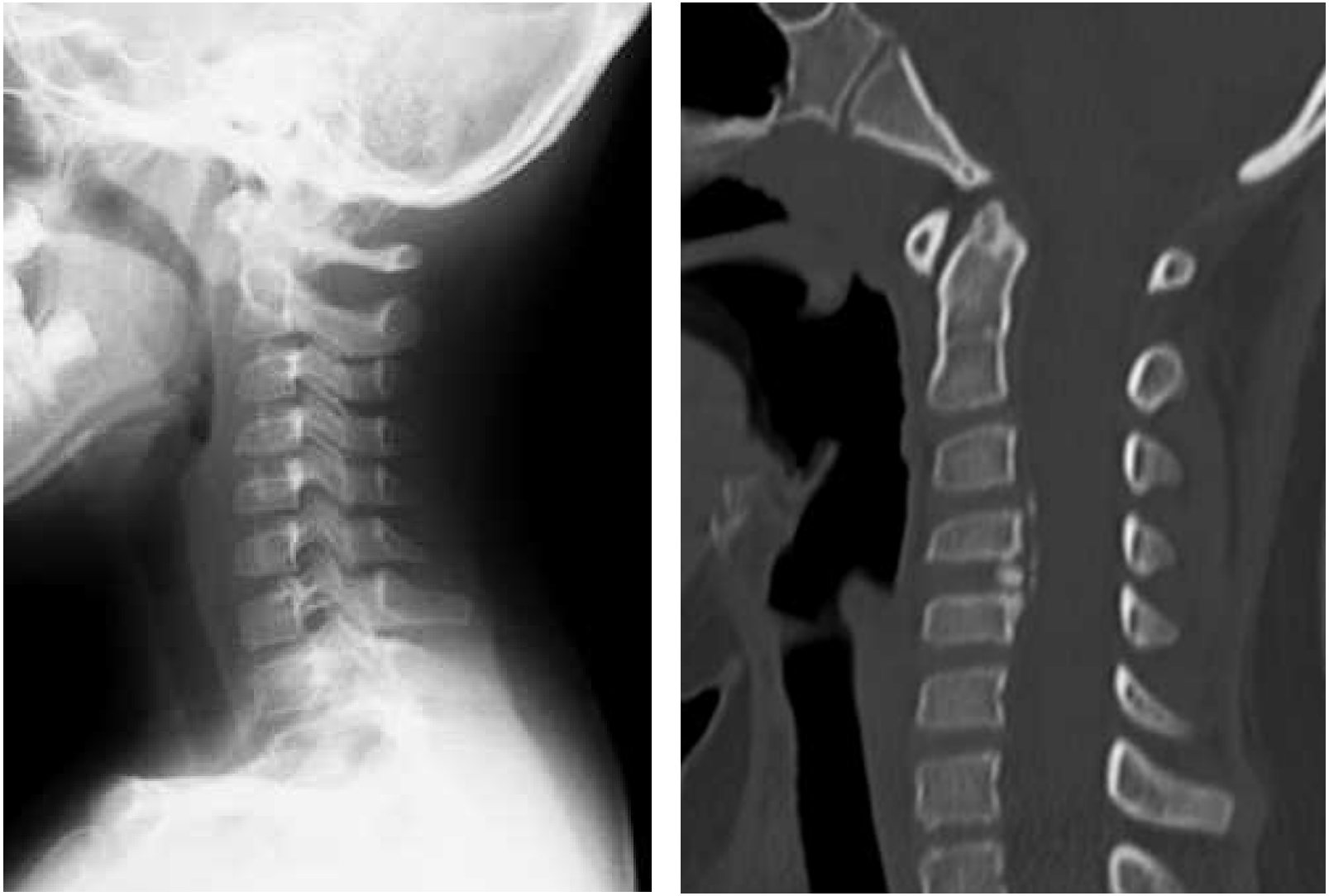
B

Fig 1.



C D

Fig 2.



A

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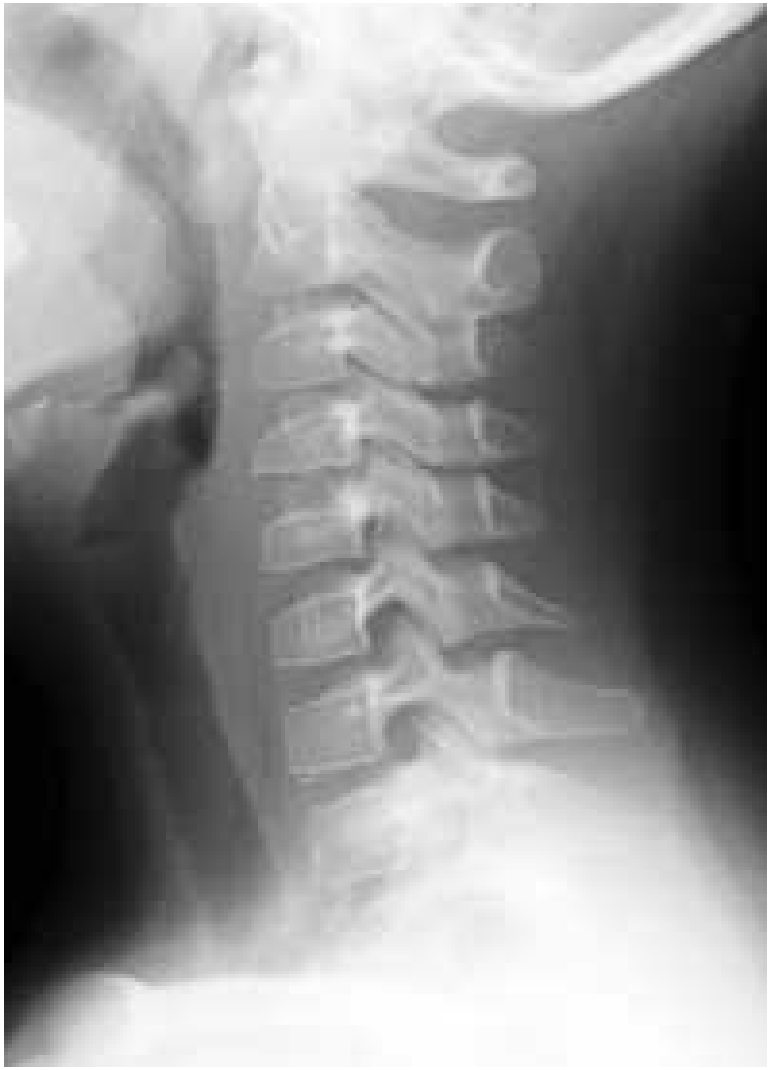
Fig 3.



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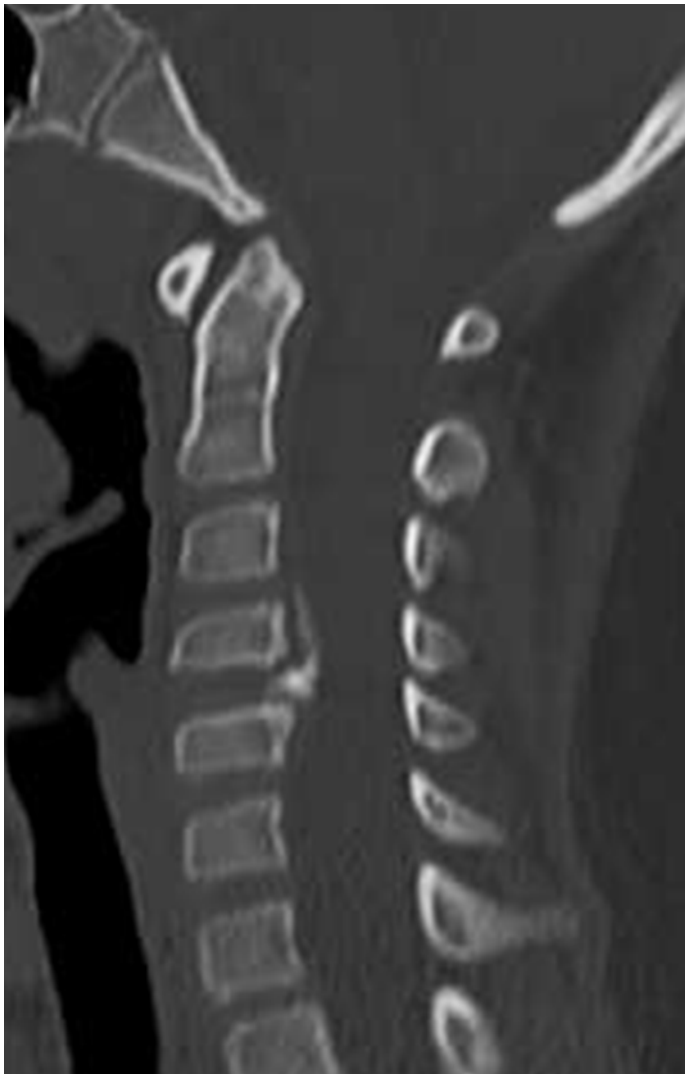
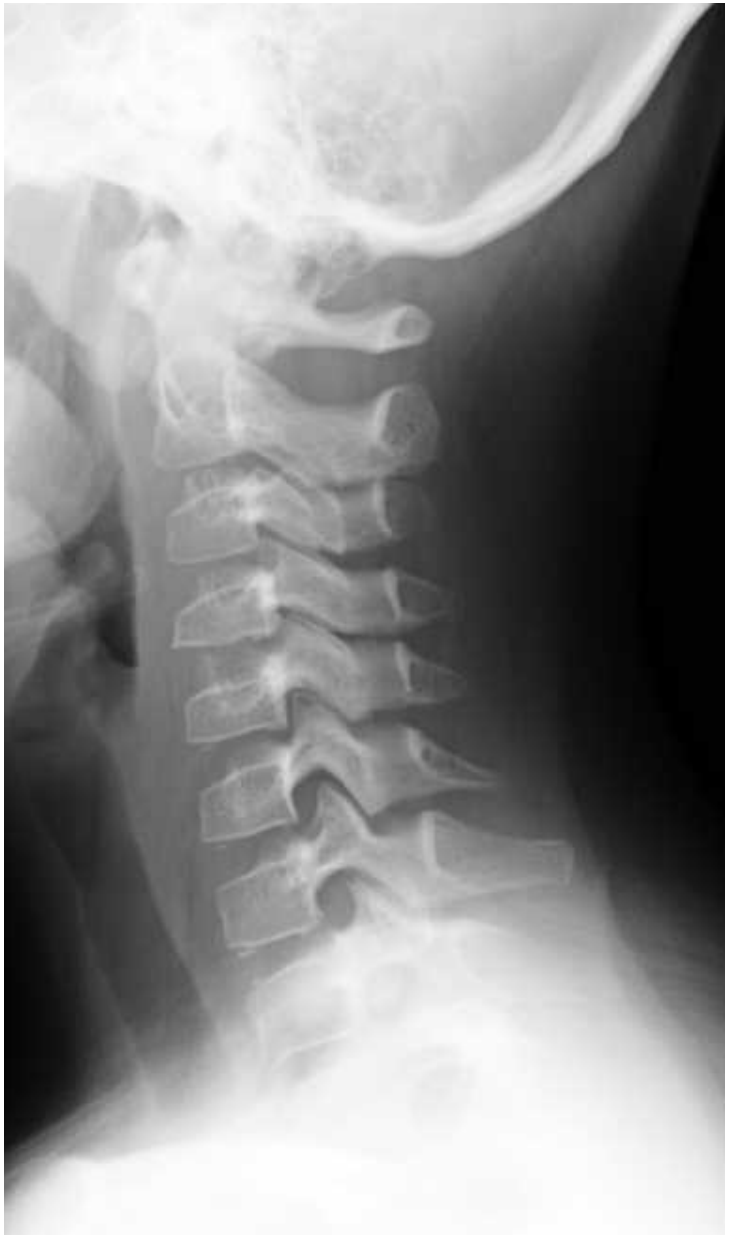
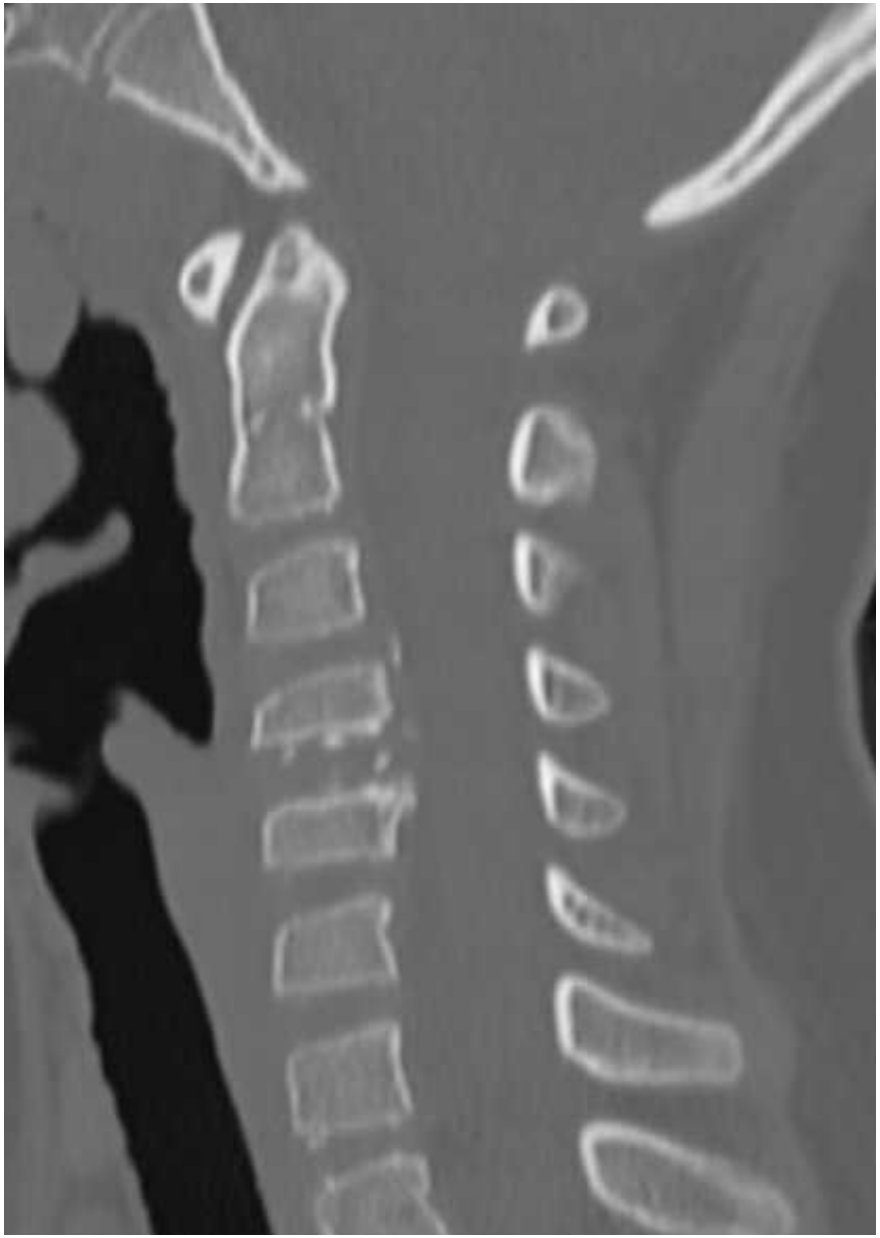


B

Fig 4.





A

B