

Conservative management of crowned dens syndrome and atlas posterior arch resection: Two successful experiences of individualized treatment

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Received June 17, 2026; Accepted September 9, 2026

DOI: 10.3892/etm.2026.13333

Abstract. Crowned dens syndrome (CDS) is a rare crystal deposition disorder characterized by distinctive halo-like or coronal calcification surrounding the odontoid process, primarily manifesting as neck pain and restricted mobility, occasionally accompanied by systemic inflammatory signs. The present report describes two cases of patients with this condition with the aim of elucidating the clinical spectrum and management of CDS. The first case involved a 72-year-old male patient who exhibited marked symptom relief following treatment with non-steroidal anti-inflammatory drugs and a brief corticosteroid regimen, with no recurrence noted over a 2-year follow-up period. The second case involved an

81-year-old male patient who developed severe cervical spinal cord compression due to a calcified pseudotumor posterior to the odontoid process, leading to debilitating neurological deficits. This patient underwent a posterior atlas arch resection with microscopy assistance, resulting in the marked alleviation of spinal cord compression symptoms and sustained improvement over 3 years of follow-up. On the whole, the present report underscores the importance of recognizing CDS and highlights that isolated posterior arch resection may serve as an effective intervention for patients with associated high cervical myelopathy. Given the rarity of this condition, further investigations through additional case reports are warranted to confirm these findings.

Introduction

Crowned dens syndrome (CDS) was first defined in 1985 as a clinico-radiological syndrome resulting from the deposition of periodontoid calcium pyrophosphate dihydrate (CPPD) or hydroxyapatite (HA) crystals (1). The characteristic imaging features include a halo-like or coronal calcification located posterior to the odontoid process (2). Clinical manifestations can be categorized into two types: The acute type, which is more prevalent, often presenting with fever and elevated levels of inflammatory markers, such as erythrocyte sedimentation rate (ESR), C-reactive protein (CRP) and leukocyte count (white blood cells) (3). Conversely, the chronic type is rare and is characterized by substantial calcified lesions that form retroodontoid pseudotumors, leading to the compression of the cervical spinal cord. This may result in cervical spondylotic myelopathy, limb muscle weakness, paresthesia and positive pathological signs (2,4,5).

The management of CDS typically depends on the symptoms of the patient. The majority of cases respond favorably to conservative treatment approaches. Currently, primary medical interventions involve non-steroidal anti-inflammatory drugs (NSAIDs) or short courses of corticosteroids. Patients

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Abbreviations: CDS, crowned dens syndrome; CPPD, calcium pyrophosphate dehydrate; CRP, C-reactive protein; CT, computed tomography; ESR, erythrocyte sedimentation rate; HA, hydroxyapatite; JOA, Japanese Orthopedic Association; MRI, magnetic resonance imaging; NSAID, non-steroidal anti-inflammatory drug; VAS, visual analogue scale

Key words: CDS, cervical spondylotic myelopathy, NSAID, atlas posterior arch resection

frequently experience significant symptom relief within a few days of administration (6,7).

However, for those suffering from cervical spondylotic myelopathy due to prolonged pain associated with spinal cord compression, accompanied by limb muscle weakness, regional paresthesia and positive pathological indicators of spinal nerve injury, surgical intervention becomes imperative before irreversible damage occurs to the spinal cord (8). Various surgical techniques are employed based on individual patient conditions, hospital resources available for surgery and the clinical expertise of the attending physicians. Presently utilized surgical methods primarily include the following: Anterior approaches via microsurgery (transoral) or endoscopy (transnasal), as well as posterior approaches through epidural access involving the direct resection of lesions, while simultaneously performing posterior internal fixation (2,5,9,10). Therefore, the present study reports two cases of CDS with distinct clinical presentations and therapeutic strategies, one successfully managed conservatively and the other with CDS-related high cervical myelopathy treated by isolated resection of the posterior arch of the atlas under microscopic assistance, a surgical approach rarely documented in the literature. The study aims to elucidate the clinical spectrum of this rare disorder and to highlight that posterior atlas arch resection alone may serve as an effective and less invasive option for patients with CDS complicated by cervical cord compression.

Case reports

Case 1. A 72-year-old man was admitted to the Department of orthopedics, Guangzhou Red Cross Hospital of Jinan University (Guangzhou, China) in April 2022, presenting with occipital and cervical pain that had persisted for >1 month prior to admission, accompanied by restricted mobility of the cervical spine. The pain was exacerbated in the morning and was associated with episodes of dizziness and occasional numbness in both hands. The patient had no notable previous medical history, including rheumatoid arthritis or pseudoarthritis, or any other predisposing factors.

A physical examination revealed tenderness upon palpation of the spinous processes and interspinous spaces of the cervical spine, as well as limited extension and rotation of the neck. All other aspects of the physical examination did not reveal any notable findings. A computed tomography (CT) scan of the cervical spine demonstrated thickening of the posterior ligament at the odontoid process along with coronal calcification, which resulted in the narrowing of the adjacent spinal canal (Fig. 1). Magnetic resonance imaging (MRI) showed thickening of the soft tissue posterior to the odontoid process, with compression and deformation of the anterior margin of the spinal cord (Fig. 1). Laboratory tests did not show an increase in inflammatory markers such as C-reactive protein, interleukin-6, erythrocyte sedimentation and rheumatoid factor (data not shown).

Following a thorough assessment to exclude contraindications for medication use, treatment was initiated with diclofenac sodium sustained-release tablets (75 mg once daily) and prednisone tablets (5 mg three times daily) for a duration of 1 week. This regimen led to symptomatic relief. Upon discharge, medication was continued for an additional

week to reinforce therapeutic outcomes. The relevant clinical information for case 1 is presented in Table I.

Prior to treatment initiation, the visual analog scale (VAS) score (11) of the patient was recorded at 5 points; however, a post-treatment evaluation indicated a reduction in the VAS score to 1 point. During 2 years of subsequent follow-up visits, as the patient reported no recurrence of neck pain and was unwilling to undergo further cervical spine imaging, no imaging analyses were performed.

Case 2. An 81-year-old man was admitted to the Department of Orthopedics, Guangzhou Red Cross Hospital of Jinan University in December 2021, presenting with progressive neck and shoulder pain for 3 months, accompanied by restricted neck mobility, radiating pain in the upper limbs, numbness, gait instability, dysuria and impaired fine finger movements indicative of nerve compression. The patient had no prior history of rheumatoid arthritis, pseudoarthritis or cervical spine trauma, but had a 6-year history of hypertension and a 20-year history of chronic bronchitis. The patient had previously undergone knee joint replacement surgery and percutaneous coronary drug-coated balloon angioplasty.

A physical examination revealed limited neck movement, a spastic gait, diminished fine motor function in both hands, reduced muscle strength and superficial sensation in the left upper limb, as well as a positive left Hoffman sign. Laboratory tests indicated no elevated levels of inflammatory markers. Dynamic X-rays of the cervical spine revealed no signs of instability. A CT scan revealed multiple bone erosions with irregular density surrounding the odontoid process and the left atlantooccipital joint surface (Fig. 2). MRI demonstrated spinal canal stenosis posterior to the odontoid process along with cervical spinal cord compression at the C1-C2 level (Fig. 3). The relevant clinical information for case 2 is presented in Table I.

The patient presented with obvious symptoms of spinal cord compression, and exhibited a poor response to conservative treatment (200 mg oral celecoxib every 12 h for 2 weeks), with neurological function gradually deteriorating. Surgery had to be performed prior to the occurrence of irreversible spinal cord damage. However, considering the advanced age of the patient and the potential complex complications associated with surgery, a microscope-assisted posterior arch resection of the atlas via the posterior approach was performed. The surgery was performed under general anesthesia with endotracheal intubation. Following successful anesthesia, the patient was placed in the prone position, and cranial traction was applied to the cervical vertebrae using a cranial traction device. Following satisfactory positioning, routine disinfection and draping of the neck were performed. After locating the C1 vertebral segment on the body surface, a midline posterior incision of ~4 cm was made. The skin, subcutaneous tissue and supraspinous ligament were incised longitudinally. The erector spinae muscles on both sides were stripped using a periosteal elevator to expose the posterior arch of C1. With the assistance of a neurosurgical microscope, the posterior arch of the atlas was precisely resected using a rongeur and the width of the space created by the arch removal was ~3 cm. The dural sac was gradually exposed, and adequate decompression was achieved

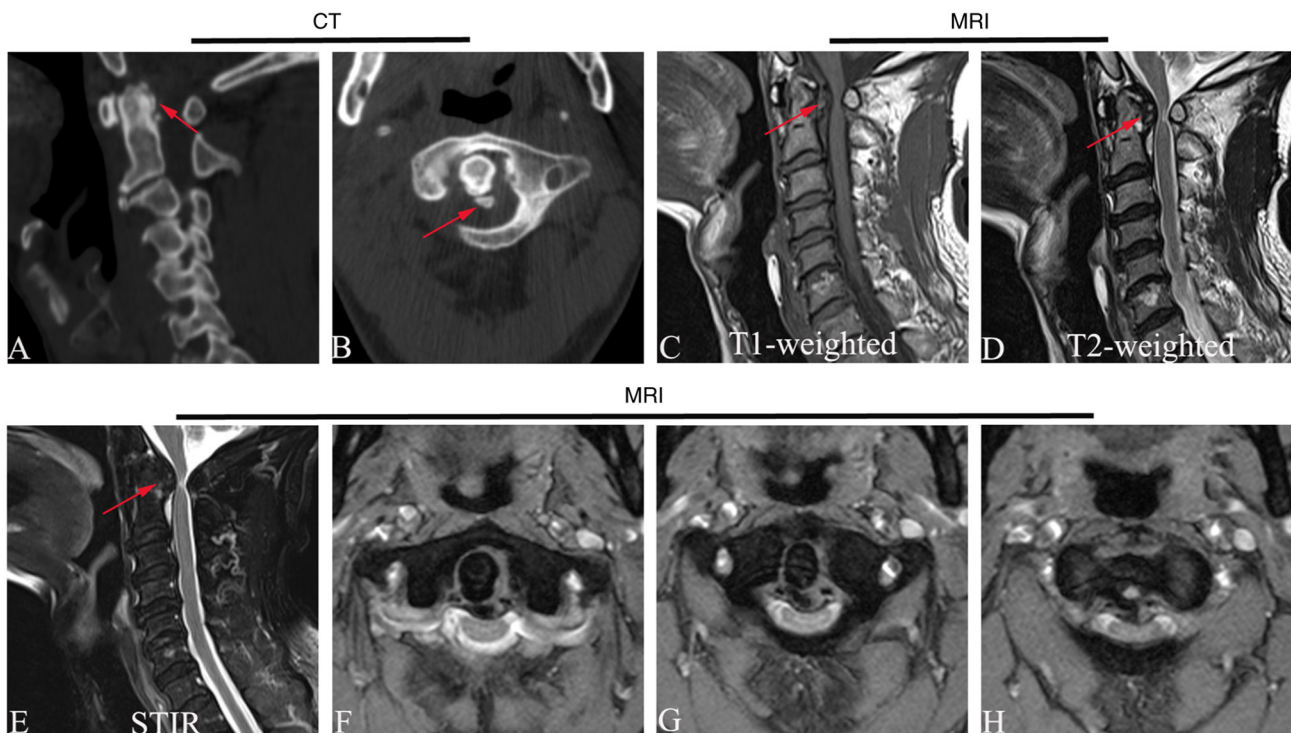


Figure 1. CT and MRI data of case 1 with crowned dens syndrome. (A and B) CT of the cervical spine: Calcifications posterior to the odontoid process with secondary narrowing of the adjacent spinal canal (arrows). (C-H) MRI of the cervical spine: The soft tissue posterior to the odontoid process was markedly thickened, the adjacent spinal canal was narrowed, and the corresponding anterior margin of the spinal cord was compressed and deformed (arrows). CT, computed tomography; MRI, magnetic resonance imaging; STIR, short-T1 inversion recovery.

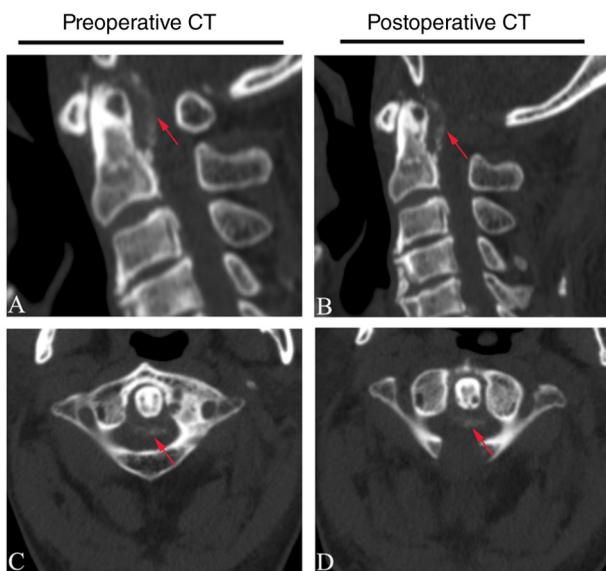


Figure 2. CT imaging data of case 2 with crowned dens syndrome. (A and C) Preoperative CT of the cervical spine suggested that multiple bone erosions and surrounding flocculent high-density shadows compressed the cervical spinal cord and led to spinal canal stenosis (arrows). (B and D) Postoperative cervical CT: Partial resection of the posterior arch of the atlas and improvement of the spinal stenosis (arrows). CT, computed tomography.

to provide sufficient space for the compressed spinal cord to shift. During the surgery, two anti-adhesion membranes were placed, and hemostatic suturing was performed. Intraoperative

blood loss was ~30 ml. The total duration of the surgery was 2 h and 10 min.

Postoperatively, NSAIDs were administered for 2 weeks. The preoperative Japanese Orthopedic Association (JOA) score (12) was 8, and the VAS score was 5. Post-operatively, the JOA score improved to 14, and the VAS score decreased to 1. At 3 years post-surgery, the JOA score had reached 16, and the VAS score was 0. The post-operative neurological function improvement rate was 66.7%, which increased to 88.9% at 3 years post-surgery. During the 3-year follow-up, the symptoms of the patient did not recur, and digital radiography revealed no increase or absorption of calcified foci around the odontoid process, no stress fracture of the anterior arch, no atlantoaxial instability and no other complications (Fig. 4).

Discussion

The prevalence of CPPD crystal disease in individuals presenting with acute neck pain is ~2% (13). Due to the limited understanding of CDS, a misdiagnosis or a missed diagnosis frequently occurs in routine clinical practice (14). In the two cases presented in the current study, both patients were elderly men at >70 years of age who sought medical attention for acute neck pain accompanied by restricted neck movement; both were subsequently diagnosed with CDS following admission. Consequently, it is imperative for clinicians, particularly those working in Emergency Departments, to enhance their awareness of CDS. This condition should be considered in patients experiencing acute neck pain alongside elevated inflammatory

Table I. Summary of clinical data of case 1 and case 2.

Clinical information	Case 1 (conservative treatment)	Case 2 (surgical treatment)
Age, years	72	81
Sex	Male	Male
Main symptoms	Isolated neck pain and restricted mobility	Neck pain accompanied by spinal cord compression symptoms such as neck stiffness, radiating pain and numbness in both upper extremities, unsteady gait and dysuria
Neurological examination	Tenderness and percussion pain over each cervical spinous process and interspinous space, with limited neck extension and rotation.	Limited neck mobility, spastic gait and impaired fine motor skills of both hands; decreased muscle strength and palmar sensation in the left upper extremity, positive Hoffmann's sign on the left
Laboratory examination	No elevation of inflammatory markers	No elevation of inflammatory markers
CT/MRI findings	CT: Thickening and calcification of the posterior ligament of the odontoid process, secondary spinal canal stenosis. MRI: Marked thickening of the soft tissue posterior to the odontoid process, with compression and deformation of the anterior margin of the spinal cord	CT: Multiple bone erosions of the odontoid process and the left atlanto-occipital articular surface, with surrounding irregular density shadows. MRI: Spinal canal stenosis posterior to the odontoid process, degenerative compression of the cervical spinal cord at the C1-C2 segment
Treatment	Diclofenac sodium sustained-release tablets (75 mg orally once daily) combined with prednisone tablets (5 mg orally three times daily) for conservative treatment for 2 weeks	Microscope-assisted posterior atlas posterior arch resection; postoperative maintenance treatment with NSAIDs for 2 weeks
Follow-up duration	2 years	3 years
Recurrence	Neck pain has not recurred	Symptoms of spinal cord compression were markedly relieved, the patient regained self-care ability and the symptoms did not recur
Complications	None	No complications such as anterior arch stress fracture, atlantoaxial instability or enlargement of calcified foci

CDS, crowned dens syndrome; NSAID, non-steroidal anti-inflammatory drug; JOA, Japanese Orthopedic Association score; VAS, visual analog scale.

markers. Timely cervical spine CT scans are essential to improve the diagnostic rate of CDS and to prevent unnecessary examinations or treatments that could delay appropriate diagnosis and management.

The pathological mechanism underlying CDS is closely associated with the deposition of CPPD or HA crystals. While CPPD deposition typically affects peripheral joints, such as the knees and wrists, spinal involvement remains rare (15,16). Conversely, HA crystal deposition predominantly occurs at tendon attachment sites. Existing literature indicates that CPPD-related CDS is more commonly reported than HA crystal deposition-induced CDS, which is consistently recognized as a rare subtype predominantly affecting middle-aged women. Although precise incidence rates are unavailable due to the rarity of these conditions and the absence of large-scale epidemiological data, the number of published cases and

case series for CPPD-related CDS substantially exceeds that for HA crystal deposition-related CDS (15,16). The imaging characteristics exhibited by these two conditions are similar; therefore, differentiation based solely on imaging findings may not be feasible (14,17).

CDS may manifest with a range of clinical symptoms; however, the typical presentations during the acute phase often include acute neck pain, neck stiffness and elevated levels of inflammatory markers such as ESR, CRP and white blood cell count. As the disease progresses, substantial calcified lesions can evolve into space-occupying masses that progressively compress the cervical spinal cord, ultimately resulting in cervical spondylotic myelopathy. Patients may exhibit signs of nerve damage, including diminished muscle strength in the limbs, paresthesia, abnormal bowel and urinary function, as well as positive pathological findings (18). Consequently,

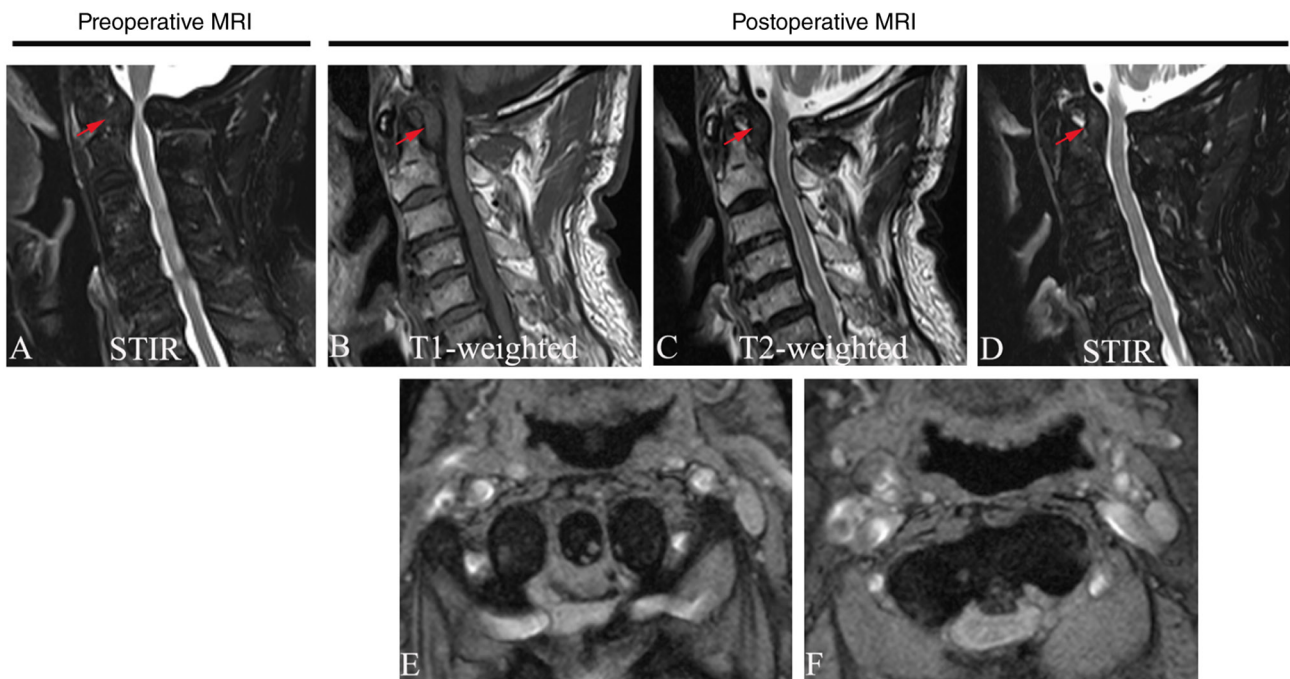


Figure 3. MRI data of case 2 with crowned dens syndrome. (A) Preoperative MRI: Thickening of the posterior ligament of the odontoid process, secondary spinal canal stenosis and ischemic degeneration of local spinal cord compression (arrow). (B-F) Postoperative MRI: Partial resection of the posterior arch of the atlas was performed, which relieved the pre-existing local spinal stenosis and cervical cord compression (arrows). MRI, magnetic resonance imaging; STIR, short-TI inversion recovery.

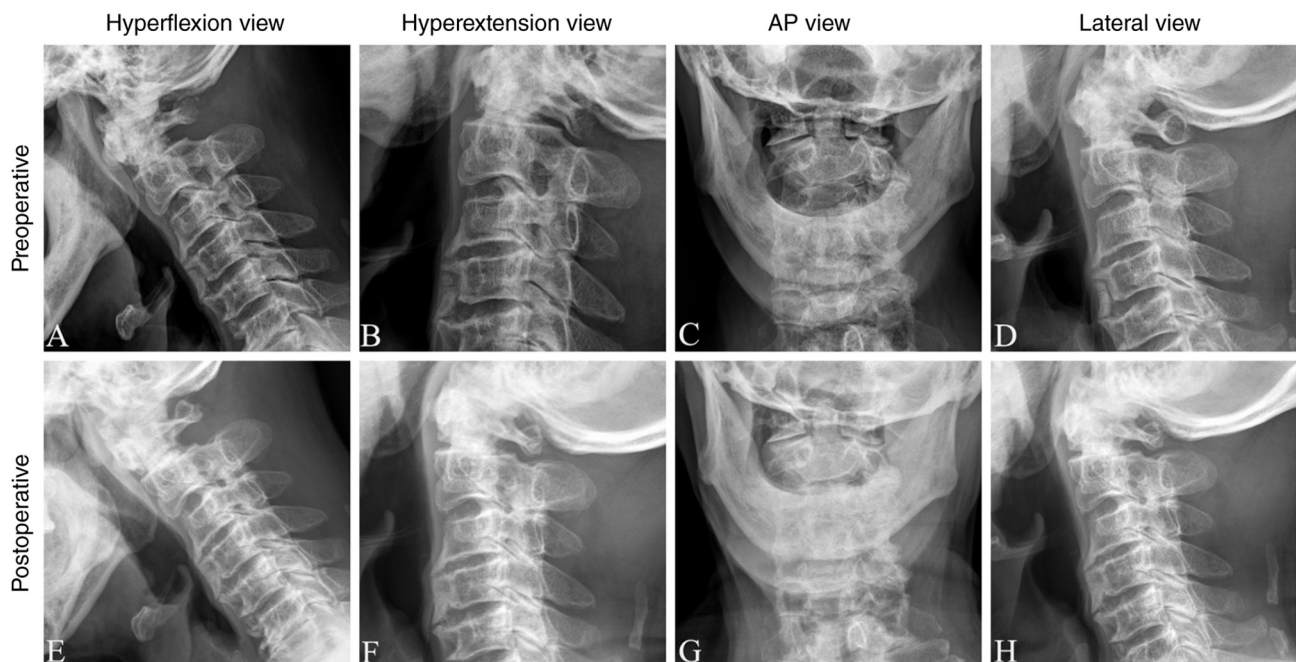


Figure 4. X-ray imaging data of case 2 with crowned dens syndrome. (A-D) Anterior and lateral dynamic position of the cervical spine before surgery: Degenerative lesion of the cervical spine without cervical instability. (E-H) Dynamic position of the cervical spine in the anteroposterior and lateral position after surgery: Degenerative lesions of the cervical spine, without cervical instability. AP, anteroposterior.

it is essential to differentiate CDS from various other conditions such as trauma, meningitis, cervical discitis or giant cell arteritis, epidural abscesses, osteomyelitis, polymyalgia rheumatica, metastatic bone disease and tumors. Clinically relevant differential diagnoses can be established through a thorough patient medical history review coupled with

meticulous physical examinations and appropriate auxiliary investigations (4).

A neck CT scan is considered the gold standard for diagnosing CDS due to its high resolution, which allows for the clear visualization of calcification morphology and distribution along with assessing spinal canal stenosis severity (19).

In contrast to CT scans, while MRI cannot directly identify crystal deposition, it plays a crucial role in evaluating the extent of spinal cord compression and excluding tumors or infections among other diseases (20). Dynamic X-ray imaging serves a key purpose by aiding in the assessment of cervical spine stability prior to surgical intervention while also aiding in the prevention of secondary deformities post-operatively (5). In the present study, the diagnosis of CDS in case 2 was established based on a combination of characteristic clinical features, pathognomonic imaging findings and systematic exclusion of differential diagnoses. The diagnosis of CDS in case 2 fulfilled the currently established diagnostic criteria, including: i) Acute severe neck pain; ii) marked limitation of cervical motion, particularly rotation; iii) elevated inflammatory markers, such as elevated CRP levels; iv) CT evidence of calcium deposition around the odontoid process; and v) absence of trauma history, with exclusion of other inflammatory diseases and tumors (21,22). Among these, cervical CT scan remains the gold standard for diagnosing CDS, as its high resolution enables clear visualization of the morphology and distribution of calcifications and accurate assessment of spinal canal stenosis. The most characteristic imaging finding of CDS is the coronal or horseshoe-shaped calcification deposited around the top and lateral aspects of the odontoid process, which was clearly demonstrated on the cervical CT scan of case 2. Although MRI cannot directly identify crystal deposits, it plays an irreplaceable role in evaluating the degree of spinal cord compression and excluding neoplastic or infectious conditions, while dynamic cervical radiographs are valuable for preoperative assessment of cervical stability to prevent postoperative secondary deformity.

Notably, the CT scans in case 2 revealed multiple bone erosions around the odontoid process and atlantooccipital joint. Such erosive changes are recognized manifestations in patients with CDS, likely resulting from chronic inflammatory stimulation caused by CPPD or HA crystal deposition. However, these erosions exhibit a non-aggressive, localized pattern and lack the permeative or destructive characteristics typical of neoplastic or infectious diseases, thereby supporting the diagnosis of CDS rather than other invasive pathologies.

In clinical practice, CDS must be differentiated from trauma, meningitis, cervical myofascial pain syndrome, polymyalgia rheumatica (PMR), epidural abscess, osteomyelitis, metastatic bone disease and tumors through detailed history taking, meticulous physical examination and appropriate ancillary investigations (23). In the present study, trauma was excluded on clinical and imaging grounds, as case 2 had no history of recent neck trauma and cervical CT revealed no evidence of atlantoaxial dislocation or fracture. Meningitis, one of the most critical differential diagnoses, was also excluded on clinical and imaging grounds; although meningitis closely mimics CDS in clinical presentation, an important diagnostic criterion for meningitis is the demonstration of nodular or thickened linear meningeal enhancement on MRI, with meningeal enhancement >3 cm raising strong suspicion for meningeal pathology, yet no such meningeal changes were observed in case 2. Cervical myofascial pain syndrome was excluded on clinical and physical examination grounds, as this condition typically results from overuse of

cervical myofascial tissues or acute trauma, with palpable cord-like structures at the affected site and symptomatic relief achievable through manual massage, whereas case 2 had no predisposing factors, no abnormal cord-like structures were palpated and symptoms could not be relieved by manual massage. Polymyalgia rheumatica was excluded on clinical and laboratory grounds, as the typical manifestations of PMR, including stiffness and pain in the neck, shoulder and hip girdles, shoulder girdle fatigue in nearly all patients, neck girdle fatigue in $\sim 70\%$ of cases, and non-specific systemic features such as low-grade fever, fatigue, somnolence, generalized myalgia, anorexia and weight loss, were absent in case 2. Infectious conditions such as epidural abscess and osteomyelitis were excluded on laboratory and imaging grounds, as peripheral white blood cell and neutrophil counts were within normal limits upon admission, and cervical MRI showed no evidence of spinal canal abscess or vertebral body infection. Similarly, metastatic bone disease and primary cervical tumors were excluded on clinical and imaging grounds, as case 2 had no history of malignancy and cervical MRI revealed no malignant space-occupying lesions. In conclusion, the diagnosis of CDS in case 2 is well established based on the characteristic clinical presentation, pathognomonic imaging findings and systematic exclusion of relevant differential diagnoses through clinical evaluation, laboratory investigations and imaging studies.

As no pharmacological therapy is currently available to eliminate or prevent intra-articular crystal deposition, the management strategy for CDS emphasizes inflammation control and individualized treatment. For acute CDS, NSAIDs combined with short-term corticosteroids can effectively inhibit the inflammatory response and alleviate pain and stiffness. Colchicine may serve as a second-line option in refractory cases; however, its gastrointestinal side-effects could limit its use among elderly patients. Since there is no direct drug therapy available to eliminate or prevent the deposition of crystal deposits in the joints, the current focus of medication treatment is to reduce the associated inflammation, resulting in a certain probability of symptom recurrence. In different studies, the recurrence rate of symptoms following conservative treatment is mostly concentrated in the range of 20 to 35%, with follow-up periods typically ranging from 6 to 12 months. In the study by Goto *et al* (18), 40 patients with symptomatic CDS confirmed by CT were included, and the treatment regimen included NSAID monotherapy, prednisone monotherapy or a combination of both. At the 9-month follow-up, the symptom recurrence rate was 22.5% (9/40). In another study, the overall recurrence rate after conservative treatment was 34.62% (9/26); the recurrence time span ranged from 0.5 to 12 months, and patients who experienced recurrence could still achieve rapid symptom relief with the same type of medication (24). Within the present study, in case 1, after excluding relevant drug contraindications for the patient, a combination of diclofenac sodium sustained-release tablets and prednisone tablets was used for treatment. The symptoms of the patient were markedly relieved, consistent with the treatment outcomes reported in other cases, and no recurrence occurred during subsequent follow-up.

As observed in case 2 of the present study, the patient ultimately required surgical intervention due to the

compression of the cervical spinal cord caused by the formation of a posterior odontoid pseudotumor (8). Currently, the most commonly employed surgical methods include: The anterior approach, with microsurgical (transoral) and endoscopic (transnasal) techniques; and the posterior approach, wherein the lesion is directly excised through an epidural route during surgery, accompanied by posterior internal fixation. These surgical approaches have demonstrated favorable treatment outcomes for patients. Nonetheless, there remains no consensus on the optimal surgical strategy to date (25).

Considering elderly patients with complex underlying conditions, they may not tolerate the high risks, prolonged operative times and potential postoperative complications associated with transoral resection of calcification or isolated posterior fusion procedures. Within the present study, in case 2, a posterior approach was used for an atlas posterior arch resection. Before proceeding with the resection, a dynamic fluoroscopic evaluation was performed under anesthesia, including flexion-extension views, to confirm the absence of pathological translation or angulation at the atlantoaxial joint. During the surgical exposure, the integrity of the transverse atlantal ligament and the surrounding soft tissues was also directly inspected. Stability was defined as the absence of >3 mm of anterior translation of the atlas relative to the axis on lateral fluoroscopy and no evidence of ligamentous disruption on direct visualization. Preoperative imaging assessment revealed no atlantoaxial instability in the patient, so internal fixation was not performed. Simple atlas resection (without fusion) is a surgical procedure applicable to conditions such as Arnold-Chiari malformation (often combined with suboccipital craniectomy), intraspinal tumors at the C1 level and retrodental pseudotumors without instability. The total width of the resected atlas posterior arch was 3 cm (below the safe exposure width of ~3.5 cm for the atlas posterior arch), which avoided intraoperative injury to the vertebral artery (26). The postoperative neurological improvement rate was ~66.7%, and the improvement rate at 3 years postoperatively reached ~88.9%. Long-term stress fractures of the atlas anterior arch after an atlas posterior arch resection are rare, but severe cases may result in atlantoaxial joint instability, necessitating a reoperation for internal fixation (26-28). In the present case, the 3-year follow-up imaging assessment revealed no enlargement or absorption of calcified lesions around the odontoid process, no anterior arch stress fracture, no cervical instability and no other complications.

In conclusion, the management of cervical degenerative stenosis should be tailored to the extent of calcification and nerve compression present in each patient. For the majority of individuals with acute CDS, conservative treatment is deemed appropriate; however, resection of the posterior arch of the atlas has been shown to effectively alleviate symptoms associated with chronic myelopathy. Based on the single surgically treated case described within the present study, a posterior arch resection appears to be a potential treatment option; however, its effectiveness cannot be validated at this stage. Further reports with a greater number of cases are necessary to draw definitive conclusions.

Acknowledgements

Not applicable.

Funding

The present study was financially supported by the Guangzhou Municipal Science and Technology Project (grant nos. 2025A03J3363, 2023A03J0571 and 2023A03J0574), the Guangzhou Traditional Chinese Medicine and Integrated Traditional Chinese and Western Medicine Project (grant no. 20242A011014), and the Technology Project of Guangzhou Municipal Health Commission (grant no. 20261A010032).

Availability of data and materials

The data generated in the present study are included in the figures and/or tables of this article.

Authors' contributions

YL contributed to study conception and design, collated clinical cases and wrote the original manuscript. XL obtained medical images (MRI and CT scans), analyzed patient data and drafted the manuscript. ZW obtained X-ray images, and performed the diagnostic analysis. XX contributed to study conception and design, and revised the manuscript. SH and HM contributed to study conception and design, and provided critical revision and writing guidance. XX and HM confirm the authenticity of all the raw data. All authors have read and approved the final version of the manuscript.

Ethics approval and consent to participate

Informed consent for the use of their samples in scientific research was obtained from all patients. The present study was conducted after the protocol was approved by the Medical Ethics Committee of Guangzhou Red Cross Hospital of Jinan University (Guangzhou, China; approval no. 2022-091-01).

Patient consent for publication

Written informed consent was obtained from the patients for publication of the present case report and associated images.

Competing interests

The authors declare that they have no competing interests.

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