



CASE REPORT

Odontoid osteomyelitis with atlantoaxial subluxation in an infant

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Abstract

Purpose To report a rare case of odontoid osteomyelitis with atlantoaxial subluxation in a 6-month-old infant.

Background Odontoid osteomyelitis with atlantoaxial subluxation is extremely rare in children. Although several cases have been reported, there have been no studies concerning proper surgical drainage and immobilization in this disease.

Methods A 6-month-old infant with odontoid osteomyelitis with atlantoaxial subluxation was surgically treated. The patient underwent a 3-month intravenous and oral antibiotic course and the Minerva body jacket cast was used for 3 months. Follow-up was carried out with computed tomographic scans and a cervical spine dynamogram.

Results At 18 months post-surgery, the patient had completely recovered with no cervical instability. Computed tomographic scans revealed complete fusion of odontoid synchondrosis. The infant remained asymptomatic with a full range of head movement.

Conclusion Surgical drainage and proper immobilization appears to be a satisfactory treatment for pyogenic osteomyelitis of odontoid synchondrosis secondary to retropharyngeal abscess and atlantoaxial subluxation.

Level of evidence N/A.

Keywords Odontoid · Osteomyelitis · Atlantoaxial subluxation · Pediatrics · Retropharyngeal abscess

Introduction

Osteomyelitis of the odontoid is an extremely rare disease in children that may cause instability between the atlas and axis. Although several cases have been reported, only one shows definite atlantoaxial subluxation in an infant [2, 4, 10, 12, 17]. We report a case of an infant with odontoid osteomyelitis with retropharyngeal abscess (RPA) that led to atlantoaxial subluxation.

Case report

A 6-month-old infant was taken to the emergency room with a fever of 39.5 °C persisting for 4 days. The physical examination of the chest and abdomen was not remarkable. Initial laboratory evaluation revealed an erythrocyte sedimentation rate of 83 mm/h (normal = 0–15), a white blood cell count of 27,440/µL with 67.5% polymorphonuclear leukocytes, and C-reactive protein levels of >160 mg/L (normal = 0.1–6.0). The neurological examination and cerebrospinal fluid examination were normal. The infant was admitted to the pediatric department and blood cultures were obtained due to the possibility of sepsis.

Day 5 after admission, the infant still had a high fever and the parents noted that the infant displayed reduced cervical motion and guarded his neck. The infant was referred to our orthopaedic department. Light touching over the posterior upper cervical spine caused irritability in the patient. The parents recalled no preceding trauma. The cervical plain film showed a loss of continuity of odontoid

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synchondrosis suggesting a fracture and subluxation of the atlas on the axis and the instability of atlantoaxial joint (Fig. 1). Noncontrast computed tomography (CT) and magnetic resonance imaging (MRI) with gadolinium enhancement were performed for further investigation. CT demonstrated a bony alignment change between the odontoid process and C2 body and retropharyngeal space soft tissue swelling and edema (Fig. 2). MRI revealed a multilocular abscess around the odontoid process and diffuse soft tissue inflammatory processes at epidural, paraspinal, and prevertebral retropharyngeal areas. The signal change and gadolinium enhancement of dens suggesting evidence of osteomyelitis were seen (Fig. 3). Blood cultures were positive for methicillin-resistant *Staphylococcus aureus*. Intravenous administration of teicoplanin and cefotaxime was initiated.

The diagnosis was odontoid osteomyelitis and atlantoaxial subluxation secondary to RPA without neurologic dysfunction. Intravenous antibiotic therapy with immobilization of neck was one of treatment options. However, we chose surgical drainage of RPA with immobilization of neck because we were concerned about airway obstruction due to large RPA and detrimental effects of RPA on the atlantoaxial stability.

Day 10 after admission, the infant underwent surgery under general anesthesia. The patient was positioned in a supine position and fluoroscopically controlled transoral surgery was performed. A vertical midline incision was made through the pharyngeal mucosa. Mucosal flaps and pharyngeal constrictor muscular layer were divided and retracted on each side. Complete evacuation of the abscess was carried out and all granulation tissue was removed. Wounds are thoroughly irrigated with saline solution. Loose approximation of the wound edges was done with the Vicryl 4-0, no suction drains were placed. After surgery, we used the individually formed Minerva cast made of two-component polyurethane foam (Neofrakt, Schumacher, Krefeld, FRG). Our cast covers the frontal and



Fig. 2 The computed tomography sagittal image reveals retropharyngeal swelling and destruction of odontoid synchondrosis with atlatoaxial subluxation

occipital regions of the skull and extends over the neck, chest, back, abdomen, and iliac crest. The face, ears, the top of the head and hair, and extremity are exposed. To facilitate toileting or diaper changing and hygienic cleaning, the buttocks and pubic areas are also exposed. The Neofrakt casting procedure required the use of padding and cotton stockinette similar to the Scotchcast procedure. The double-layered textile, evenly filled with polyurethane, is applied to the patient and closed by means of a zipper on supine position. After molding cast, the alignment of the atlas on the axis was checked with an image intensifier (Fig. 4). We adjusted the degree of neck extension without traction before hardening of cast material. This cast was maintained for 24 h in a day (Fig. 5).

The infant was started on a liquid diet on the first postoperative day through the Levin tube. After 2 weeks, soft diet was stated. Results of microbiology tests confirmed the infectious agent. Same intravenous antibiotics were administrated during his hospitalization. Over the

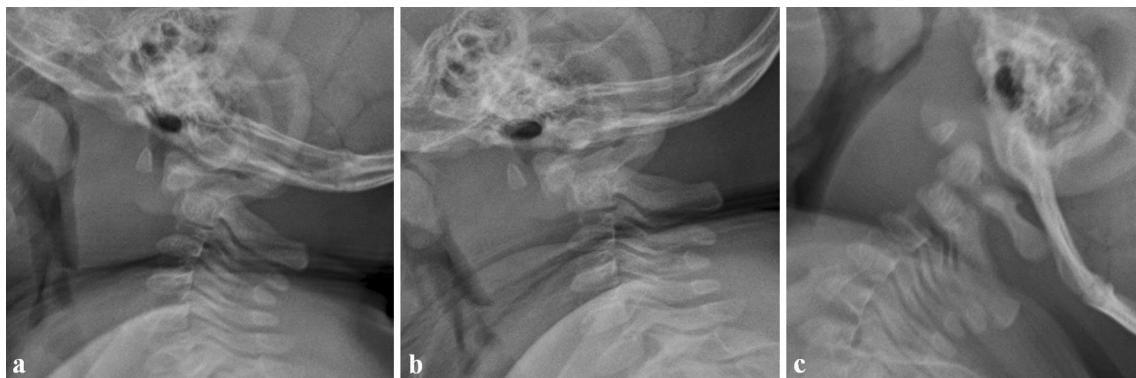


Fig. 1 **a** A lateral view of the cervical radiograph shows atlantoaxial subluxation and prevertebral soft tissue swelling. The displacement between the odontoid process and the body of axis widened on flexion view (**b**) and reduced on extension view (**c**)

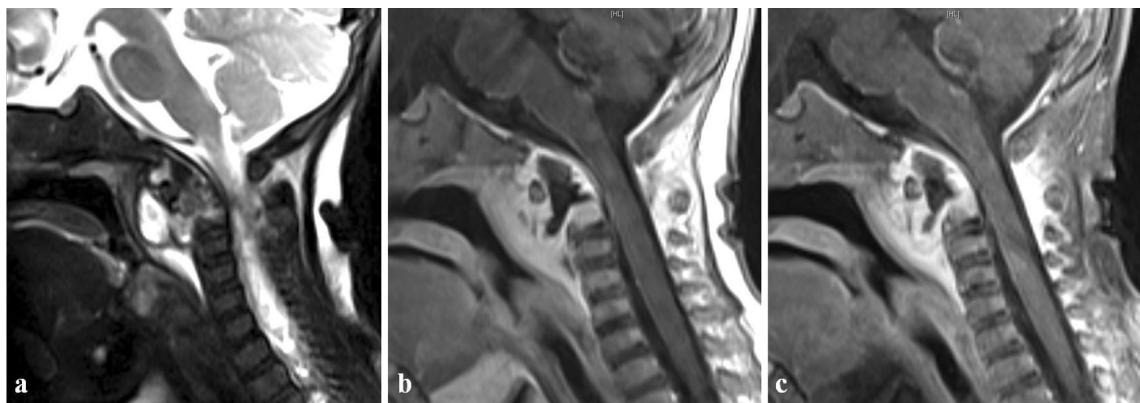


Fig. 3 **a** The sagittal T2-weighted image consistent with large abscess in retropharyngeal space. **b** The sagittal T1-weighted image shows a large retropharyngeal abscess around the odontoid process

and hypointensity of the odontoid process. **c** The sagittal T1-weighted image after gadolinium injection showing enhancement of the dens

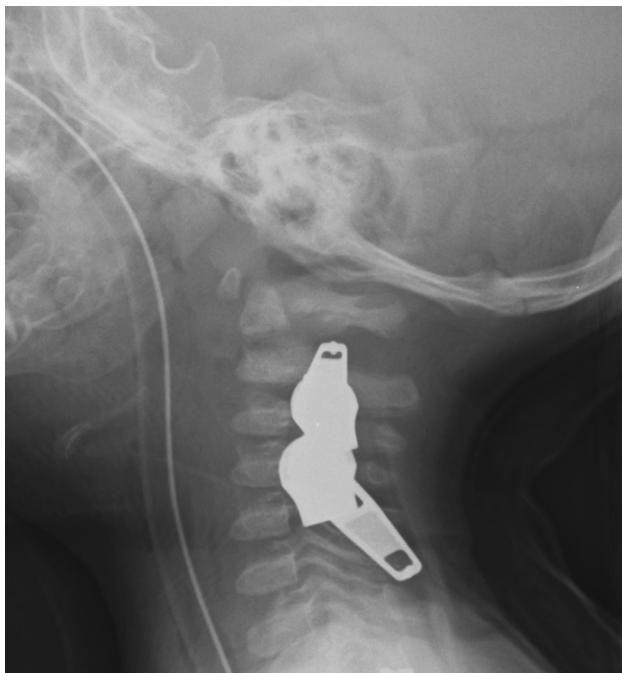


Fig. 4 An immediate postoperative lateral view of the cervical radiograph shows good alignment of the atlatoaxial joint with the Minerva cast

next 2 weeks, the patient had no further febrile episodes and all laboratory tests were normal. The patient was discharged after 24 days of uncomplicated hospitalization and remained on oral antibiotics for the next 3 months.

After 3 months, the Minerva cast was removed. A follow-up plain radiograph 15 months later showed sclerotic changes between the body and dens of C2. There was no instability identified by cervical spine dynamogram and CT scans demonstrated bony union of the odontoid synchondrosis (Fig. 6). The infant remains asymptomatic with a full range of head movement.



Fig. 5 The patient was immobilized with the Minerva cast for 3 months

Discussion

In the pediatric population, the odontoid process is an extremely rare site for osteomyelitis. Only six cases have been reported (Table 1) [2, 4, 10, 12, 17]. There were two cases showing abscess formation and one case showing atlantoaxial subluxation. One patient with multilocular RPA was surgically treated using a transmastoïd approach [12], four patients without abscess were treated with intravenous antibiotic administration only [4, 10, 17], and one infant showing fatal atlantoaxial dislocation and large RPA abscess died [2]. There was limited experience with treatment of odontoid osteomyelitis, but intravenous antibiotic treatment can be recommended in patients without abscess formation and surgical drainage may be considered in patients with RPA. Therefore, we performed

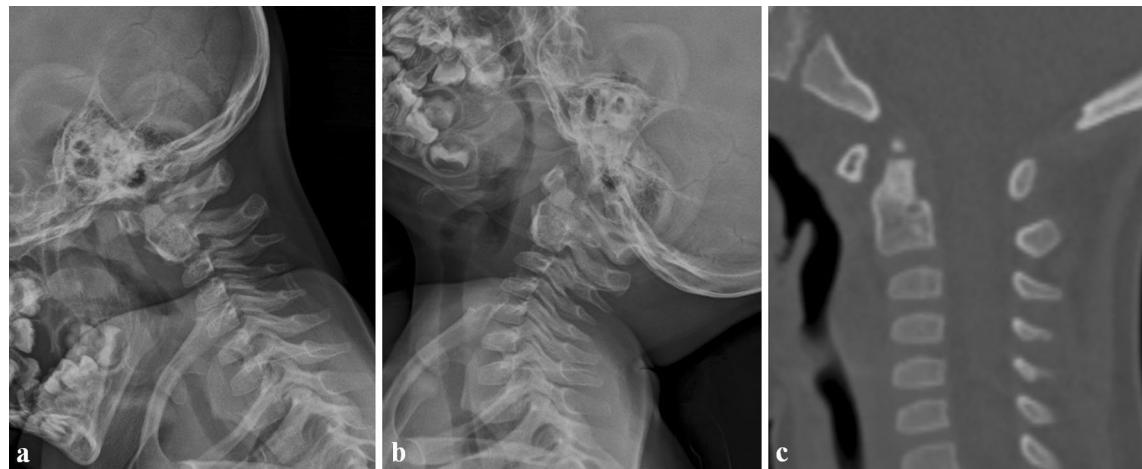


Fig. 6 Follow-up analysis 18 months after the surgery revealed **a, b** no radiologic instability via cervical spine dynamogram and **c** union of odontoid synchondrosis via computed tomography sagittal

Table 1 Review of previous case reports about odontoid osteomyelitis

References	Age	MRI findings	Abscess	Atlantoaxial subluxation	Treatment	Outcome
Chen et al. [2]	9 M	Large retropharyngeal abscess extending intracranially	Yes	Yes	Death before treatment	Death
Papp et al. [12]	1 M	Multilocular abscess in mastoidal area with the destruction of C1 and C2	Yes	No	Surgical drainage and cervical immobilization with custom-made device	Improved
Dimaala et al. [4]	1 Y 6 M	Enhancement of odontoid, thickening and enhancement of the prevertebral tissue, retropharyngeal inflammation or infection without a ring-enhancing abscess	No	No	Intravenous antibiotics with cervical immobilization	Improved
Nolting et al. [10]	1 Y 8 M	Increased signal in the odontoid process with surrounding edema	No	No	Intravenous antibiotics with soft collar	Improved
Zimmermann et al. [17]	1 Y 2 M	Edema of the basis of the odontoid peg with enhancement	No	No	Intravenous antibiotics only	Improved
Zimmermann et al. [17]	2 Y 8 M	Marked enhancement of the odontoid	No	No	Intravenous antibiotics only	Improved
Our study	6 M	Multilocular abscess around the odontoid process, signal change and enhancement of dens	Yes	Yes	Surgical drainage and cervical immobilization with Minerva cast	Improved

Y Years, M Months

surgical drainage for large RPA and cervical immobilization with Minerva cast for definite atlantoaxial subluxation. The result of treatment for our case was successful.

A high index of suspicion is required for the diagnosis of odontoid osteomyelitis, as it has variable clinical symptoms and initial investigation may be misleading. Most cases in previous studies were not initially diagnosed [2, 4, 10, 17]. Our case was also diagnosed at day 5 after admission. Further evaluation of neck area is necessary in nonverbal children with reduced head movement [17]. Radiography does not always provide an accurate diagnosis, making MRI the ideal modality for diagnosis of odontoid

osteomyelitis [9, 16]. However, our case was easily detected by plain radiography due to atlantoaxial subluxation. Nevertheless, MRI images were necessary to find the cause of C1–2 subluxation.

The standard treatment of vertebral osteomyelitis in children is administration of appropriate antibiotics. However, if an abscess is associated with an established osteomyelitis, surgical drainage should be considered. RPA is uncommon and usually found in pediatric population because the lymph nodes in this space during childhood are prone to infection [6]. Traditional management of RPA has been surgical drainage, [7] but some pediatric patients were

treated with antibiotics alone [1]. Although nonoperative treatment may be considered in some cases with RPA in which the abscess is small, surgical drainage is mandatory in patients who have large abscess because the complication of RPA involve serious morbidity including airway obstruction and subsequent sequela such as os odontoideum, pseudoarthrosis, and chronic atlantoaxial instability [3, 14]. As illustrated by our case with odontoid myelitis and large RPA, we think that surgical drainage of RPA could be effective for odontoid synchondrosis union and to prevent instability of atlantoaxial joint.

Atlantoaxial subluxation secondary to RPA involves destruction of bony structure or loosening of atlantoaxial ligaments as a result of the inflammatory process [15]. Grisel's syndrome also shows non-traumatic atlantoaxial subluxation, because upper respiratory tract infection can lead to relaxation of the transverse ligament and subluxation of the atlantoaxial joint [5, 13]. However, most patients with Grisel's syndrome presents with torticollis due to atlantoaxial rotatory subluxation [11]. Our case showed definite anteroposterior instability caused by destruction of synchondrosis of odontoid process without torticollis and was different from the Grisel's syndrome.

If there is instability of the atlantoaxial joint, appropriate cervical immobilization should be considered [8]. Proper treatment of cervical instability may include traction, bracing, or surgery. Because our case occurred in a very young infant, we used the Minerva cast to stabilize the atlantoaxial joint externally. The Minerva cast was applied with the neck placed in an extended position. Union at the odontoid chondrosis was observed at final follow-up.

Conclusions

Surgical drainage and proper immobilization appear to be satisfactory for treatment of pyogenic osteomyelitis of odontoid synchondrosis with RPA that led to atlantoaxial subluxation.

Compliance with ethical standards

We did not receive grants or outside funding in support of this research or for preparation of this manuscript. We did not receive payments or other benefits or a commitment or agreement to provide such benefits from a commercial entity. No commercial entity paid or directed, or agreed to pay or direct, any benefits to any research fund, foundation, educational institution, or other charitable or nonprofit organization with which we are affiliated or associated.

Conflict of interest None of the authors has any potential conflict of interest.

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