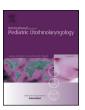
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#### Case Report

# Infection of the spheno-occipital synchondrosis: A morbid complication following adenoidectomy



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#### ABSTRACT

Two 2-year-old males presented post-operatively following adenoidectomy with persistent fever and neck stiffness. After multiple office visits, both patients were admitted and found to have a widened spheno-occipital synchondrosis and other imaging findings indicative of skull base osteomyelitis. Treatment with antibiotics allowed for recovery with good long-term outcomes. Infection involving the spheno-occipital synchondrosis is rare and its circuitous presentation of these two children no doubt led to delayed diagnosis.

#### 1. Introduction

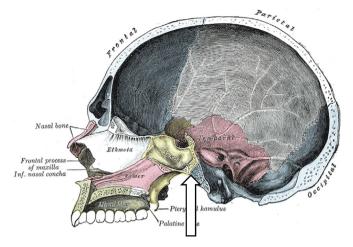
Pediatric adenoidectomy is a common, relatively safe procedure, with mortality rates similar to those of general anesthesia (less than 1 in 35,000), while complication rates range from 0 to 12% in the literature [1-6]. Typical postoperative course-though some authors classified as complications-is often self-limited and includes post-operative emesis, fever, halitosis, and neck pain and stiffness [6-9]. Post-operative neck pain is thought to be attributable to cervical muscle irritation and inflammation of adjacent tissues [9,10]; however, in rare cases, neck pain and stiffness has heralded serious underlying infections including necrotizing fasciitis, meningitis, and even cervical osteomyelitis [10-13]. Neck stiffness and torticollis following adenoidectomy may also represent Grisel syndrome, one of the most morbid but extremely rare complications [10,14]. Beyond the subluxation of the atlanto-axial joint in Grisel syndrome and two isolated reports of cervical osteomyelitis, there have been limited reports of skeletal complications secondary to adenoidectomy. To our knowledge, osteomyelitis of the skull base following pediatric adenoidectomy has only been reported twice in the first half of the 20th century [13,15]. Both patients developed otitis media following adenoidectomy, with subsequent clinical deterioration ultimately resulting in mortality. Importantly, one case occurred in the pre-antibiotic era, and both occurred prior to the advent of high-resolution imaging. They were likely only temporally related to adenoidectomy. We present two cases of osteomyelitis of the sphenooccipital synchondrosis attributable to complications of adenoidectomy. The spheno-occipital synchondrosis is a potential space at the junction of the sphenoid and occipital bones which ossifies during late childhood (Fig. 1).

#### 2. Case 1

A 2-year-old male with a history of congenital diplegia underwent an uncomplicated adenoidectomy and bilateral myringotomy with ventilation tube placement for adenoid hypertrophy and recurrent acute otitis media. The patient also received onabotulinum toxin type A injections and phenol intramuscular neurolysis of the bilateral lower extremities. On post-operative day six, the mother called with concerns of bilateral shoulder pain, which was believed to be due to a recent motor vehicle accident (MVA) three days prior; over the counter analgesics were recommended as needed. On post-operative day 16, the mother called reporting fever, neck pain, sore throat, and behavioral changes. On post-operative day 33, the patient presented to clinic with persistent symptoms, now in addition to headaches. The neck pain was attributed to the MVA, and the child was placed in a cervical collar per recommendations by the pediatrician in consultation with neurosurgery; MRI was recommended but was not obtained.

On post-operative day 37, the patient presented to the emergency

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Spheno-occipital synchondrosis

Fig. 1. The arrow depicts the location of the spheno-occipital synchondrosis from a mid-sagittal view of the cranium and skull base.

room with worsening neck pain, inability to flex the neck, and decreased range of vertical eye movements; the patient was afebrile, but labs showed leukocytosis with a left shift. Cervical spine x-rays ruled out spinal pathology from the MVA; however, MRI of the cervical spine demonstrated a widened spheno-occipital synchondrosis and T2 hyperintensity through the anterior clival periosteum consistent with infection (Fig. 2A). CT demonstrated bony erosions with similar widening of the synchondrosis and heterogeneously enhancing material filling the widened space. Suspected infection was believed to be a complication of the adenoidectomy. The patient was admitted to the hospital, and treatment was initiated with ampicillin/sulbactam. Clinical improvement occurred over four inpatient days (partial improved neck range of motion), and the patient was transitioned to oral amoxicillin/clavulanic acid at the time of discharge. After one month of oral antibiotics, the patient was reevaluated and noted to have complete resolution of the neck pain and stiffness. 2.5 months after discharge, MRI demonstrated a normal-appearing spheno-occipital synchondrosis (Fig. 2B), and clinically, there were no residual complications.

#### 3. Case 2

A 2-year-old fully vaccinated male with a past history of neonatal torticollis underwent an uncomplicated adenoidectomy and bilateral myringotomy with ventilation tube placement for adenoid hypertrophy and chronic otitis media with effusion. The patient presented to the pediatrician on post-operative day five with fever, cough, drooling, and yellow-green nasal discharge, which was attributed to a viral upper respiratory infection. On post-operative day eight, the mother called reporting continued symptoms, in addition to a stiff neck. Approximately one week later, the patient was diagnosed with viral pharyngitis by the pediatrician. On post-operative days 26 and 28, the patient presented to general pediatrics and otolaryngology clinic respectively with fever, vomiting, poor feeding, neck pain, continued low energy, and irritability. Other than a fever, exam was unremarkable and a CBC was drawn demonstrating leukocytosis with a left shift.

After developing bilateral periorbital swelling on post-operative day 31, the patient presented to the emergency department where he was found to have nuchal rigidity and an elevated C-reactive protein (CRP) and erythrocyte sedimentation rate (ESR). Cerebrospinal fluid analysis demonstrated mild pleocytosis with a negative gram stain and culture. Cervical-spine MRI demonstrated a widened spheno-occipital synchondrosis containing T2 hyperintense material extending towards the nasopharynx through the anterior clival periosteum consistent with osteomyelitis (Fig. 3A). Axial MRI showed cavernous sinus thrombosis with bilateral ophthalmic vein thrombosis (Fig. 3B), while MRA showed narrowed cavernous portions of the bilateral internal carotid arteries. Treatment was started with enoxaparin and empiric intravenous vancomycin, ceftriaxone, and metronidazole. Antibiotic therapy was narrowed after blood cultures were positive for nontypeable Haemophilis influenza and beta-lactamase negative Streptococcus anginosus susceptible to ceftriaxone and metronidazole. Improvement in inflammatory markers was followed by clinical improvement, and the patient was discharged on hospital day 14 with instructions to continue intravenous antibiotics and enoxaparin. On latest follow-up 70 days after discharge, the patient was asymptomatic; repeat MRI demonstrated cavernous sinus thrombosis had improved. No other long-term complications have been noted.

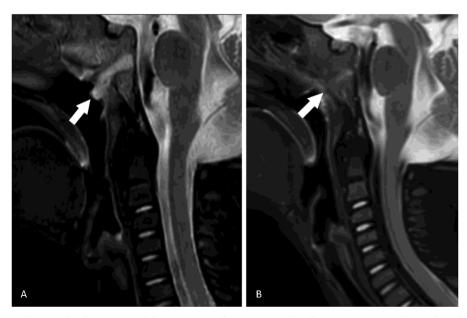


Fig. 2. A) Sagittal fat-suppressed T2-weighted MR image of the craniocervical junction reveals inflammatory tissue widening the spheno-occipital synchondrosis (arrow) consistent with osteomyelitis. B) Similar imaging, following therapy, demonstrates resolution of inflammation (arrow).

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