

Rhinolithiasis: an unusual cause of sinusitis complicated with frontal osteomyelitis and epidural abscess

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Rhinoliths are rare foreign bodies of the nose formed by *in situ* mineralization of endogenous or exogenous foreign material. They are often asymptomatic but may have various clinical presentations, with purulent rhinorrhea and nasal obstruction being the most common. They may go unnoticed for a long period and be diagnosed accidentally during a routine examination. We report a 6.5-year-old girl with rhinolithiasis complicated with sinusitis, frontal osteomyelitis and epidural abscess; she had a history dating back four years.

Key words: rhinolithiasis, sinusitis, frontal osteomyelitis, epidural abscess.

Rhinoliths are mineralized masses that precipitate on an intranasal foreign body¹⁻¹¹. Any material lodged in the nasal cavity can act as a nidus for deposition of salts^{1,2,5-9,12}. The encrusted nidus can be exogenous or endogenous. Dried blood clots, teeth, sequestra, desquamated epithelium, dried pus, and bone fragments are considered endogenous^{2,3,5,7-12}. Fruit seeds, buttons, beads, peas, dirt, pebbles, glass, wood, and the remains of a gauze tampon are examples of exogenous sources^{2,3,5,7-13}. Although nasal obstruction, discharge, malodor and headache are among the common symptoms, some patients may remain asymptomatic for years¹⁻¹².

Complications secondary to rhinoliths have been described, including recurrent sinusitis, recurrent otitis media, recurrent dacryocystitis, destruction of the nasal mucosa, oronasal and oronasal fistula, septal deviation and perforation, palatal perforation, and bony destruction of the maxillary sinus^{1-4,7,8-10,12}. Sinusitis is not frequent, but when present it is usually treated palliatively and thus delays the correct diagnosis and treatment of rhinolithiasis, as in our case. In this report, we present a 6.5-year-old girl with rhinolithiasis complicated by sinusitis, frontal osteomyelitis and epidural abscess, with a history dating back four years.

Case Report

A 6.5-year-old girl was admitted to the Children's Emergency Room with the chief complaints of fever, cough, vomiting, forehead swelling, and nasal discharge. The patient had been hospitalized and treated with intravenous (iv) antibiotics for two weeks in another center prior to admission. She was referred to our tertiary center because of persisting fever and vomiting. During her initial assessment, she had fever and forehead swelling. There were no neurologic or ophthalmologic symptoms, and laboratory tests showed increased erythrocyte sedimentation rate and elevated C-reactive protein and white blood count. Cranial computed tomography (CT) examination revealed left frontal epidural abscess measuring 3x1.5 cm and pansinusitis (Fig. 1). The patient was consulted with neurosurgery and otolaryngology. Immediate surgery was not planned and the patient was hospitalized by the Pediatric Infectious Diseases Department and placed on iv cefotaxime and metronidazole treatment. She responded well to the treatment and a follow-up axial-cranial and coronal-paranasal CT scan repeated after four weeks showed significant decrease in diameters of the abscess, whereas paranasal CT showed sinusitis, bony defect secondary to

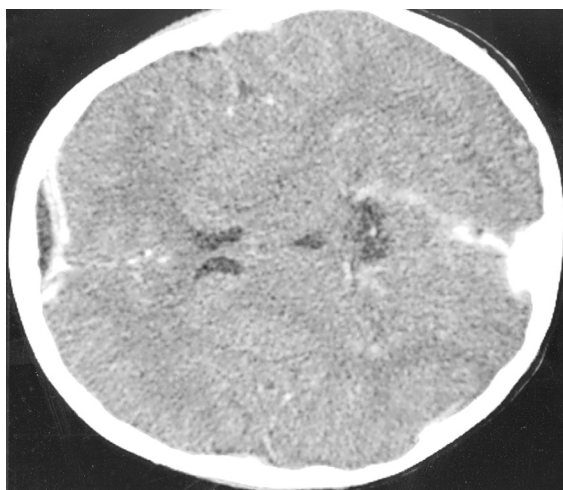


Fig. 1. Epidural abscess demonstrated as a hypodense collection located in the anterior part of the left frontal lobe extending to the right of the midline with peripheral contrast enhancement.

osteomyelitis at the posterior wall of frontal bone (Figs. 2a and 2b) and a hyperdense mass lodged on the floor of the nose on the left side, consistent with a rhinolith (Fig. 3). Further querying of the mother revealed a suspicious history of a left-sided nasal foreign body from four years ago. She clearly stated that her child had suffered left-sided malodorous purulent discharge since she was 2.5 years old and had been treated for sinusitis many times. The patient was operated to remove the rhinolith (Fig. 4) with bilateral middle meatal antrostomy, anterior-posterior ethmoidectomy, and left frontal sinusotomy. After surgery, the patient continued to receive iv antibiotic therapy for two weeks and was discharged home with oral amoxicillin-clavulanic acid.

Discussion

Rhinolithiasis is a rare clinical condition, with only some 700 cases reported since its first description by Bartholin in 1654^{1,3,9}. Although rare, understanding its nature will allow timely diagnosis and help to distinguish it from tumors of the nasal cavity⁵. Rhinoliths typically develop from an exogenous or endogenous nidus. The route of entry is usually anterior but some may enter through the choana secondary to sneezing, vomiting or coughing^{1,3,4,8,9}. The pathogenesis is not clear but chronic inflammatory reaction incited by the nidus is thought to result in deposition of mineral



Fig. 2a and 2b. Bony defect at the superoposterior wall of the left frontal sinus: a. coronal plane, b. axial plane.

salts, with calcium phosphate being the most common. Rhinoliths are generally lodged on the floor of the nose, halfway between the anterior and posterior portions of the nares, as in our

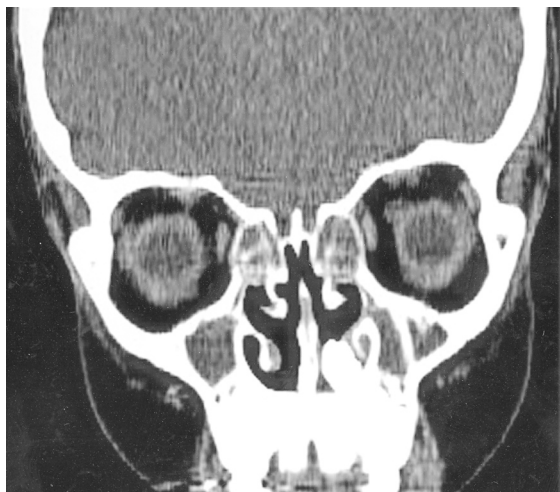


Fig. 3. A hyperdense mass located in the left nasal passage between the inferior concha and nasal septum foreign body. Opacification of bilateral maxillary sinuses and ethmoid air cells is seen.

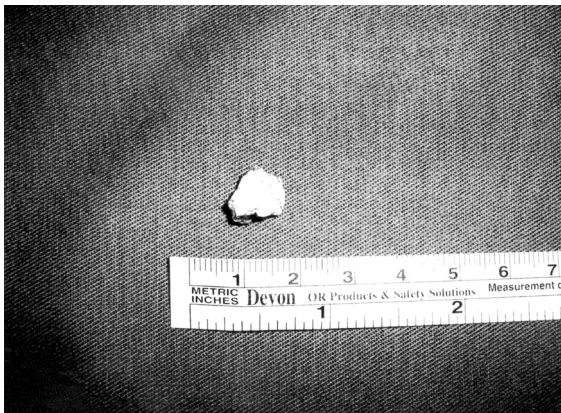


Figure 4. Rhinolith after surgical removal.

case^{1,2,4,7-9}. The most common symptoms are purulent nasal discharge, nasal obstruction and malodor¹⁻¹². Other findings may include nasal and facial swelling, pain, epistaxis, and anosmia.

The diagnosis of rhinolithiasis is usually overlooked⁵. The possibility of varying clinical presentations and history of symptoms dating back several years make the diagnosis challenging^{5,8}. Rhinoliths can appear as incidental findings on plain films and CT scans, as in our case. Presence of superimposed adjacent structures on plain films may result in diagnostic failure^{2,3,8-10,12}. A coronal CT scan is essential for complete evaluation since it does not superimpose adjacent structures. Our patient had been treated for sinusitis on several occasions. Several sinus radiographs were obtained; however, no paranasal sinus CT scan was performed. It is recommended by the American Academy of Pediatrics that imaging studies are not necessary to confirm a diagnosis of clinical sinusitis in children ≤ 6 years of age. However, CT of the sinuses should be reserved for children who present with complications of acute bacterial sinusitis or who have very persistent or recurrent infections and are not responsive to medical management¹⁴.

Intracranial suppurative complications are present in 3%-11% of all children hospitalized for sinusitis¹⁵⁻¹⁷. The incidence of intracranial complications is higher in cases of frontal sinusitis because of its unique anatomy. El Hakim et al.¹⁸ reported that the prevalence of intracranial complications was significantly higher (30.4%) in pediatric patients with frontal sinusitis compared to those without frontal

sinus involvement. Epidural abscesses are caused by osteomyelitis of the bony posterior wall of the frontal sinus. Osteomyelitis of the frontal bone in children is an extremely rare condition, most often found in neglected cases of frontal sinusitis. Severe osteomyelitis of the posterior wall of the frontal sinus can cause erosion into the epidural space, inducing an epidural abscess. Intracranial complications of sinusitis are most prevalent in young children, because this is the age at which the valveless diploic system is at its most vascular, providing a good conduit for infection, and the posterior wall of maturing frontal sinuses is a poor barrier to the spread of bacteria, causing sinus infection¹⁵. Epidural abscesses are reported as the predominant type of intracranial complications of sinusitis in children, have outcomes better than those of the other intracranial complications of sinusitis, and present without specific neurologic symptoms or signs, like in our case¹⁶. Epidural abscess resolved significantly with iv antibiotic treatment but the etiology of sinusitis could only be identified when the follow-up coronal CT scan showed the rhinolith.

A single case of meningitis as a complication of rhinolith-related orbital abscess was described by Polson in 1943¹⁹. In the literature, accompanying sinusitis has been reported in only 20-25% of the cases^{5,7,12}. In our case, we believe that rhinolithiasis was a predisposing factor for the development of frontal sinusitis and its complications, including frontal osteomyelitis and epidural abscess. To our knowledge, this is first case of rhinolithiasis causing frontal osteomyelitis and epidural abscess.

Rigid or flexible nasal endoscopy is also of great value in the diagnosis of rhinoliths, especially with posterior location, but its use is limited among the pediatric population. Therefore, flexible endoscopy must be done in all children with recurrent or persistent sinusitis. Careful patient follow-up with endoscopic evaluation is necessary to diagnose residual disease in the frontoethmoidal recess due to surgical scars and obliteration and prevent recurrence. The differential diagnosis includes osteosarcoma, chondrosarcoma and squamous cell carcinoma in the malignant tumor category; ossifying fibroma, osteoma, hemangioma, enchondroma,

dermoid, calcified polyps, odontoma, and nasal teeth in the benign tumor category; and syphilis and tuberculosis in the inflammatory category. Surgical removal of the rhinolith is the sole treatment. It can be achieved by an anterior approach assisted by rigid endoscopy under local anesthesia². General anesthesia should be reserved for children and adults having large rhinoliths where removal in a piecemeal fashion is necessary. Mink et al.²⁰ used lithotripsy to break a large stone into pieces. Concomitant cauterization of the granulation tissue, if present, and appropriate antibiotics to control the local infection will yield better healing.

In conclusion, rhinolithiasis is a rare condition and this is the first report to present a cascade of its complications.

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