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Case report

Severely infected pneumoceles of the frontal sinus in patients with mental retardation and brain atrophy treated by endoscopic sinus surgery

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ABSTRACT

We herein present three cases of abnormally expanded frontal sinuses (pneumoceles) with severe infection in patients with mental retardation and brain atrophy. Two patients previously underwent laryngotracheal separation surgery, and bacteriological examinations of purulent nasal discharge revealed infections caused by drug-resistant bacteria such as *Pseudomonas aeruginosa* and *Acinetobacter baumannii*. As conservative medical treatments were ineffective, all three patients were treated by computed tomography-guided endoscopic sinus surgery. This navigation system is useful for safer surgery in the area of anatomic deformity. The clinical findings, possible etiologies and surgical treatment of these cases are discussed.

1. Introduction

Pneumocele of the frontal sinus is a rare entity that results from abnormal expansion of sinuses beyond the margins of the containing bone with marked osseous thinning and dehiscence [1,2]. We herein present three cases of frontal sinus pneumoceles with severe infection in patients with mental retardation and brain atrophy. All three patients were successfully treated by computed tomography (CT)-guided endoscopic sinus surgery (ESS). To our knowledge, this is the first report of treatment of patients with frontal pneumoceles and severe infection by ESS. The etiology and management of severely infected frontal sinus pneumoceles are discussed.

2. Case reports

The clinical findings and management of three cases of severely infected frontal sinus pneumoceles are summarized in Table 1.

2.1. Case 1

A 65-year-old man was referred to our hospital with a 1-month history of left ocular displacement (Fig. 1A). He had mental retardation caused by high fever when he was 4 years of age. His bilateral nasal middle meatus were filled with polyps, and CT revealed brain atrophy and bilateral pneumoceles of frontal sinuses with sinusitis. The posterior wall and inferior wall of the left pneumocele were destroyed and thin (Fig. 1B–D), and the crista galli was surrounded by pneumoceles (Fig. 1E). Preoperative bacteriological examination of purulent nasal discharge revealed the presence of *Moraxella catarrhalis*. As conservative medical treatment was ineffective, ESS with the modified Lothrop procedure was performed using a CT-guided navigation

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Table 1
Clinical characteristics of the three patients. ESS: endoscopic sinus surgery, MLP: modified Lothrop procedure.

Case	1	2	3
Age	65	19	30
Sex	Male	Male	Female
Symptom	Ocular displacement	Frequent fever	Orbital cellulitis Subcutaneous abscess
Brain atrophy	Mild	Moderate	Moderate
Enlargement of mastoid air cells	+	+	–
Calvarial thicking	+	+	+
Laryngotracheal separation	–	+	+
Approach	MLP	ESS 2 times	ESS
Recurrence	–	–	+

system under general anesthesia. Bilateral sinuses including maxillary, ethmoid, and sphenoid sinuses, and frontal pneumoceles were widely opened. The frontal pneumoceles contained purulent retention. A common wide pathway from both frontal pneumoceles to the nasal cavity was created (Fig. 1F). Low-dose and long-term treatment of 14-membered macrolide (macrolide therapy) was used for postoperative medication, which is a standard post-ESS therapy in Japan. The postoperative course was uneventful, and the frontal pathway was open at postoperative 2 years (Fig. 1G) without ocular displacement (Fig. 1H). CT showed no recurrent infection (Fig. 1I–K).

2.2. Case 2

A 19-year-old man was referred to our hospital with a 3-month history of frequent fever and purulent rhinorrhea. He had severe psychomotor retardation since the onset of epilepsy at 10 months of age. He had used mechanical ventilation after undergoing laryngotracheal separation surgery at 12 years of age. Preoperative bacteriological examination of left purulent nasal discharge revealed the presence of *Pseudomonas aeruginosa* and *Citrobacter koseri*. CT revealed brain atrophy and pneumoceles of frontal sinuses with sinusitis (Fig. 2A–C). As conservative medical treatment was ineffective, bilateral ESS was performed using the CT-guided navigation system under general anesthesia. Bilateral all sinuses were widely opened. There were two different pneumoceles on the right side with serous retention, pneumocele of frontal sinus and pneumocele of supraorbital ethmoid cell (Fig. 2D). On the left side, there was a large pneumocele of frontal sinus containing purulent retention (Fig. 2E). Macrolide therapy and daily irrigation of the nasal cavity with saline continued after surgery, however, sinusitis of the left pneumocele recurred within 3 months. A second ESS was performed at 6 months after the first ESS. Besides macrolide therapy and daily irrigation, frequent prone positioning was performed to promote drainage from the frontal pneumoceles, resulted in no recurrent infection up to 5 months after the second ESS (Fig. 2F–H).

2.3. Case 3

A 30-year-old woman was referred to our hospital with a 3-day history of left orbital cellulitis and a subcutaneous abscess on the left side of her forehead. She had severe psychomotor retardation following accidental drowning at 6 years of age. She had used mechanical ventilation after undergoing laryngotracheal separation surgery at 16 years of age. CT revealed brain atrophy and pneumoceles of frontal sinuses with sinusitis. The lateral wall of the left pneumocele was destroyed and thin (Fig. 3A–C). Bacteriological examination of purulent nasal discharge revealed the presence of *Acinetobacter baumannii*. As conservative medical treatment was ineffective, ESS was performed using the CT-guided navigation system under general anesthesia. Bilateral all sinuses were widely opened. Purulent retention was noted in the pneumoceles on both sides (Fig. 3D, E). Although macrolide therapy and daily irrigation with saline continued after surgery, recurrent infection continued. However, there was no recurrence of orbital cellulitis and the subcutaneous abscess up to 2 years after ESS.

3. Discussion

Hypersinus, pneumosinus dilatans and pneumocele are terms to describe an abnormal enlargement of aerated paranasal sinuses [1]. Hypersinus is an enlargement of the sinus with normal bone thickness. Pneumosinus dilatans is an enlarged sinus outwardly displaced to cause intracranial, nasal or orbital expansion without localized bone destruction. Pneumocele is a pathological expansion of sinuses, which thins the bony sinus and displaces neighboring structures. The frontal sinus is most frequently involved bilaterally, followed by the sphenoid, maxillary and ethmoidal sinuses [3]. Although pneumoceles are generally asymptomatic, some patients develop local pressure symptoms such as frontal bossing, diplopia and headache. In case 1, a frontal pneumocele with an orbital roof defect caused ocular displacement, however, the patient had no visual disturbance.

The size of the frontal sinus is dependent on the relationship between the cessation of frontal lobe growth and the development of the frontal sinus [4]. At birth, the frontal sinus is not present or is incompletely developed. Anterior ethmoidal cells begin to invade the frontal bone at 2 years of age. Frontal lobe growth ceases by 7 years of age; at that time, the inner frontal table stops its forward migration. Pneumatization of the frontal sinus begins with growth of the outer table and is usually completed by 15 years of age [5]. The etiology of an abnormally expanded frontal sinus remains unclear, however, previous reports showed two cases of pneumosinus dilatans in patients with brain atrophy and mental retardation [6,7]. These reports

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