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Premaxillary abscess without bony erosion: An unusual complication of pediatric acute maxillary sinusitis



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ABSTRACT

Objectives: To report an unusual complication of pediatric acute maxillary sinusitis: premaxillary abscess. To describe clinical, radiological and biological presentation, treatment strategy and progression.

Material and methods: A retrospective study included all pediatric patients treated for premaxillary abscess complicating acute maxillary sinusitis in two ENT reference centers between 1999 and 2017. Disease history, clinical presentation, biological and radiological findings, treatment modalities and progression were studied.

Results: Ten patients were included, with a mean age of 10 ± 4.2 years. All presented with fever, rhinorrhea and premaxillary edema. Contrast-enhanced CT scan systematically found complete opacity of the maxillary sinus, without bone lysis, and extensive effusion along the intersinonasal wall up to the premaxillary region, extending in 3 cases back toward the parapharyngeal space. Bacteriology isolated *Streptococcus anginosus* most frequently ($n = 4$; 40%). Treatment comprised intravenous wide-spectrum antibiotics, with surgical drainage of the abscess if > 10 mm ($n = 9$; 90%). Seven of these 9 patients (78%) had recurrent abscess requiring surgical revision and 3 (33%) required a third drainage. All patients were cured without sequelae at 1 month.

Conclusion: In case of acute maxillary sinusitis with premaxillary edema, premaxillary abscess should be suspected. The high recurrence rate argues for maximalist surgery associated to close clinical monitoring with radiological examination.

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1. Introduction

Pediatric acute bacterial sinusitis is frequent and complications are rare. In a large-scale American study using the Nationwide Emergency Department Sample database for 2008, with 101,660 children, only 0.7% of those treated in emergency for acute bacterial sinusitis showed complications [1], which comprised orbital abscess (76%), periorbital cellulitis (15%) and intracranial complications (9%). Cellulitis and orbital abscess mainly occur in ethmoiditis in under-5 year-olds, whereas intracranial complications (thrombophlebitis, empyema and abscess) occur more often in pansinusitis in

male adolescents [2–5]. These regional complications are classically related to bony erosion, septic thrombophlebitis of the perforating veins, or osteomyelitis [6–10]. Acute childhood maxillary sinusitis rarely shows complications and, to our knowledge, was never associated with specific complications [2,3].

The present study reports the first pediatric series of acute maxillary sinusitis complicated by premaxillary abscess, without maxillary sinus wall bone lysis. We discuss pathophysiology, clinical and radiological presentation, treatment and progression in this unusual complication.

2. Material and methods

The study was reported following STROBE guidelines [11].

A retrospective study included all children (age < 18 years) managed for acute bacterial maxillary sinusitis complicated by

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premaxillary abscess in two ENT reference centers between January 1999 and January 2017.

Inclusion criteria comprised:

- acute maxillary sinusitis, defined by associated fever ($\geq 38.5^\circ\text{C}$), unilateral rhinorrhea (whether purulent or not) with ipsilateral maxillary sinus opacity on CT;
- premaxillary abscess, defined by facial edema and premaxillary liquid effusion (Hounsfield unit (HU) < 32) with partial or total peripheral uptake on contrast-enhanced CT [12,13].

Exclusion criteria comprised:

- sinusitis of dental origin;
- missing clinical or radiologic data.

Clinical, biological, bacteriological and radiological data were retrieved from our institutional medical information database. Files were read over to collect: age, gender, medical history, disease history and symptoms at admission, leukocyte and CRP (C-reactive protein) levels, bacteriological samples, CT reports, medical and surgical treatments, and clinical progression. CT scans were analyzed by 2 independent investigators: 1 pediatric ENT radiologist, and 1 pediatric ENT physician.

Data were analyzed on Microsoft Excel (Microsoft, Redmond, WA, USA). Quantitative variables were reported as mean and standard deviation and qualitative variables as percentage.

Parents signed a consent form for surgery. Patients and parents received oral information on the study, which had local review board approval.

3. Results

Ten patients were managed in the study centers for acute maxillary sinusitis complicated by premaxillary abscess during the study

period: 8 boys, 2 girls; mean age, 10 ± 4.2 years (range: 2.5–17 years).

3.1. Clinical presentation

All patients showed fever ($\geq 38.5^\circ\text{C}$), rhinorrhea and premaxillary edema. Eight had clinical signs of associated ethmoiditis (palpebral edema, plus exophthalmia, diplopia and visual acuity loss in 3 cases). Three had trismus and dysphagia without pharyngitis on intraoral examination. There were 7 cases of left involvement and 3 of right. None had history of sinusitis. Endoscopy systematically found unilateral mucous edema of the intersinonasal wall with pus in the ipsilateral middle meatus.

3.2. Biological findings

Leukocyte levels were elevated, with a mean value of $14,000 \pm 4,000 \times 10^6/\text{L}$ (range, $8,000\text{--}19,000 \times 10^6$) as was CRP, (Mean value: 158 mg/L; range: 29–306).

3.3. Radiologic findings

All patients underwent cerebral and sinus contrast-enhanced CT, which systematically found complete maxillary sinus opacity with ipsilateral liquid effusion and partial or total peripheral submucosal contrast uptake in the intersinonasal wall, extending inward to the piriform orifice and laterally to the premaxillary space; Fig. 1 shows a typical example. There were no cases of sinus wall bone lysis. One patient had nasal bone lysis adjacent to the premaxillary effusion (Fig. 2). In 3 children, intersinonasal wall submucosal effusion extended posteriorly along the nasal cavity to the parapharyngeal space (Fig. 1). All patients showed partial or total ipsilateral anterior ethmoid sinus opacity, and 4 showed orbital cellulitis with periosteal abscess.

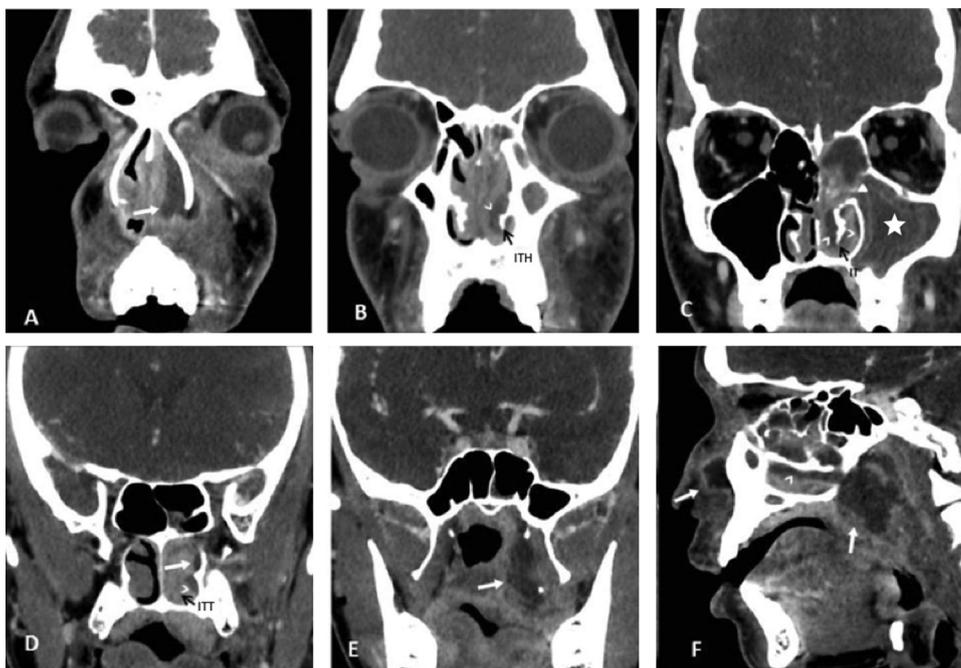


Fig. 1. Premaxillary and parapharyngeal abscesses complicating left maxillary sinusitis in a 17 year-old boy (patient #8). (A,B,C,D). Contrast-enhanced CT scan, coronal slice, tissue window. White arrow: low-density effusion in the submucosal space of lateral nasal wall, extending forward along the inferior turbinate (chevron); *: maxillary sinus opacity; ▲: obstruction of the ostium of the maxillary sinus. E. Contrast-enhanced CT scan, coronal slice, tissue window. Arrow: posterior spread of infection to the parapharyngeal space. F. Contrast-enhanced CT scan, sagittal slice, tissue window. Anterior premaxillary and posterior parapharyngeal spread (arrow) of intersinonasal wall submucosal infection (chevron). ITH: inferior turbinate head; IT: inferior turbinate; IIT: inferior turbinate tail.

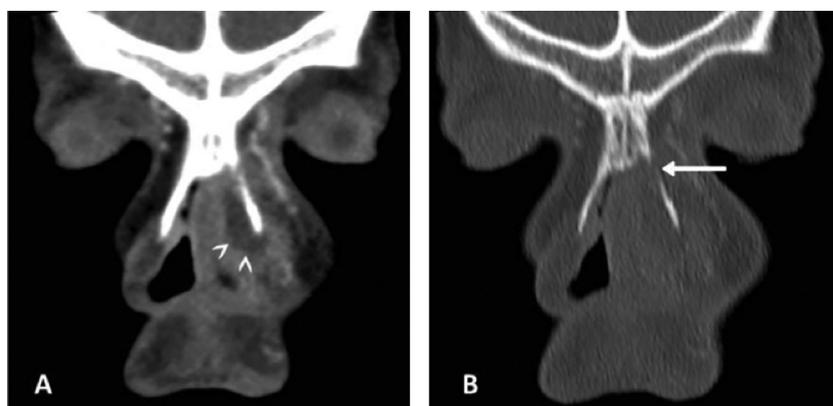


Fig. 2. Premaxillary abscess with nasal bone lysis complicating acute maxillary sinusitis in a 2.5 year-old (patient #10). Contrast-enhanced CT, coronal slice. A. Tissue window. Chevron: premaxillary abscess. B. Bone window. Arrow: nasal bone lysis.

3.4. Microbiology findings

Table 1 shows bacteriology findings. Pus samples were positive in 7 cases (70%), for 10 different bacteria, including 5 *Streptococci* (50%), 4 of which were *Streptococcus anginosus*.

3.5. Treatment

All patients received intravenous wide-spectrum antibiotic therapy, detailed in Table 1. According to our departments' protocol for complicated childhood sinusitis, antibiotherapy began with intravenous wide-spectrum probabilistic antibiotics associating ceftriaxone and clindamycin ($n=8$) or ceftriaxone and metronidazole ($n=2$) (Table 1); as appropriate, this first-line treatment was modified according to the antibiogram. Mean duration was 9 ± 6 days (range, 3–21 days). Following discharge, oral relay was prescribed for a mean 13 ± 6 days (range, 7–21 days), with amoxicillin/clavulanic acid ($n=8$), clindamycin plus amoxicillin/clavulanic acid ($n=1$) or metronidazole/clindamycin ($n=1$), depending on the department and the antibiogram.

For abscesses larger than 10 mm, surgical drainage was performed [13,14]. Only 1 patient had a 5 mm abscess responding to probabilistic antibiotherapy without need of drainage; in the other 9, drainage used a combined approach with endoscopic middle meatotomy and vestibular route drainage. The vestibular mucosa facing the maxillary was incised and the anterior maxillary wall periosteum was incised and elevated to drain the premaxillary abscess. A Delbet drainage blade (Peters Surgical®, Bobigny, France) was positioned for povidone-iodine (Betadine®) irrigation twice daily for at least 2 days.

The 3 parapharyngeal abscesses were drained by intraoral route and the 4 patients with intraorbital subperiosteal abscess underwent endoscopic anterior ethmoidectomy and external drainage via a skin incision at the medial canthus and subperiosteal dissection of the lamina papyracea for abscess drainage and a drain for twice-daily irrigation with diluted povidone-iodine.

Seven of the 9 patients initially treated by first-line surgery and antibiotic therapy showed recurrence of premaxillary abscess (78%), after a mean delay of 5 days after surgery (range, 1–14 days). Clinical suspicion was founded on persistence or recurrence of fever with pain and facial edema, confirmed by repeat contrast-enhanced sinus CT scan. In 6 cases, recurrence followed drainage blade removal. One child had early recurrence the day after surgery, with the blade still in place. Recurrence occurred before the antibiotherapy was modified in 6 cases, and after in only 1 (at 14 days). The 4 subperiosteal abscesses and 3 parapharyngeal abscesses also showed recurrence. These 7 patients underwent secondary

surgical drainage; 3 required a third, for 2 recurrences of premaxillary abscess and 1 of orbital periosteal abscess. Despite these frequent recurrences, all patients were cured by 1 month, without sequelae.

4. Discussion

To the best of our knowledge, this is the first reported series of acute bacterial maxillary sinusitis complicated by premaxillary abscess in children. Contrast-enhanced CT showed submucosal intersinonasal wall liquid effusion with peripheral uptake, extending forward to the premaxillary region and sometimes backward to the parapharyngeal region, without sinus wall bone lysis. We hypothesize that the abscesses arise from diffusion of pus from the maxillary sinus, via the ostium, to the submucosal space of the intersinonasal wall and then forward and backward to the premaxillary and parapharyngeal regions. This pattern differs from classic theories, according to which regional complications of sinusitis are due to bony erosion, septic thrombophlebitis of the perforating veins or osteomyelitis [6–10]. This complication is also to be distinguished from childhood maxillary osteomyelitis, which induces fever and facial edema but with a different clinical presentation [15–19]. It mainly affects neonates and young infants, and is of dental origin or results from hematic spread of remote infection typically implicating *Staphylococcus aureus*. Orbital abscess, but not premaxillary abscess, is very frequent in childhood maxillary osteomyelitis; moreover, effusion in the submucosal space of the intersinonasal wall has never been reported.

Three patients had parapharyngeal abscess without pharyngitis on intraoral examination. This was due to posterior spread of effusion along the intersinonasal wall. Anterior ethmoiditis was frequently associated with maxillary sinusitis in the present series, accounting for the 4 orbital abscesses. Four of the 6 isolates were of the *S. anginosus* family, frequently implicated in invasive infection [20]. A study of 17 children showed that *S. anginosus* was the most frequent pathogen in sinusitis complicated by sub- or extra-dural empyema [6], whereas non-complicated acute bacterial sinusitis classically involves *S. pneumonia* (30%), *H. influenza* (30%), or *M. catarrhalis* (10%) [21].

Surgical drainage was frequent in the present series, suggesting that a more aggressive treatment strategy may be justified in first line. Some authors reported that drainage was not always necessary in ethmoiditis complicated by orbital abscess [22]. The situation seems to be different in acute maxillary sinusitis complicated by premaxillary abscess due to the high risk of recurrence despite well-conducted surgery. We therefore recommend systematic drainage for 2 or 3 days, with drains positioned submucosally

Table 1
Bacteriology results and treatments.

Isolates	1st line IV antibiotherapy		2nd line antibiotherapy			3rd line antibiotherapy			4th line antibiotherapy			5th line antibiotherapy		
	MOL	TD	MOL	R	DT	MOL	R	TD	MOL	R	TD	MOL	R	TD
<i>Strepto. constellatus</i>	CRO + CLI + MTZ	5	AMC	PO	7	CRO + CLI	IV	7	TZP	IV	9	AMC + CLI	PO	21
<i>Strepto. mitis, Haemophilus influenzae</i>	CTX + MTZ + FOF	8	AMC	PO	6	CTX + MTZ + FOF	IV	8	AMC	PO	10	-	-	-
<i>Strepto. intermedius</i> and <i>Staph. intermedius</i>	CRO + CLI	4	AMC	PO	10	-	-	-	-	-	-	-	-	-
<i>Staph. aureus</i>	CRO + CLI	7	AMC	PO	15	-	-	-	-	-	-	-	-	-
<i>Strepto. milleri, Actinomyces odontolyticus</i>	CRO + CLI	8	SXT + CLI	PO	10	-	-	-	-	-	-	-	-	-
Negative	CRO + CLI	7	AMC	PO	10	-	-	-	-	-	-	-	-	-
<i>Veillonella sp.</i>	CRO + CLI	7	AMC	PO	7	-	-	-	-	-	-	-	-	-
Negative	CRO + MTZ	3	AMC	PO	8	-	-	-	-	-	-	-	-	-
<i>Strepto. anginosus</i>	CRO + CLI	10	AMC	PO	10	-	-	-	-	-	-	-	-	-
No bacteriological sample	CRO + CLI	5	AMC	PO	15	-	-	-	-	-	-	-	-	-

TD: treatment duration (days); MOL: molecules; R: administration route; Staph.: *Staphylococcus*; Strepto.: *Streptococcus*; sp.: species; IV: intravenous; AMC: amoxicillin + clavulanic acid; CTX: cefotaxime; CRO: ceftriaxone; CLI: clindamycin; FOF: fosfomicin; MTZ: metronidazole; SXT: trimethoprim sulfamethoxazol; TZP: piperacilline-tazobactam; PO: per os.

facing the intersinonasal wall, which requires a direct approach in the inferior meatus and at premaxillary level via a vestibular incision. Moreover, given this high risk of recurrent abscess, we would stress the importance of close clinical monitoring associated, depending on progression, to contrast-enhanced CT.

The study limitations lie in the retrospective design and small number of patients. However, the latter point relates to the rarity of this complication of acute childhood maxillary sinusitis. Further studies will be needed to determine optimal treatment strategy.

5. Conclusions

Acute childhood maxillary sinusitis may be complicated by premaxillary abscess. In our experience, this rare complication involves an exceptionally high rate of failure for surgical drainage. The present results advocate maximalist drainage strategy associated to close clinical monitoring with early radiologic examination.

Disclosure of Interest

The authors declare that they have no competing interest.

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