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Short communication

Two rare cases of maxillary pneumocoele

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Abstract

Pathological expansion of the paranasal sinus by air is rare, and may cause severe deformation of the overlying bone and tissue. We present two cases of young adults with maxillary pneumocoeles that were diagnosed by computed tomography, and discuss the differential aetiology.

Keywords: Maxillary sinus; pneumocele; aetiology

Introduction

Pathological dilatation of the paranasal sinuses by air is rare, and only about 100 cases have been reported since it was first described by Meyes in 1898.[1] The currently accepted classification was developed by Urken et al in 1987 based on the anatomical changes.[2] Hypersinus refers to a dilated sinus that does not extend beyond the normal anatomical boundaries of the sinus; pneumosinus dilatans describes a dilated sinus that extends beyond the normal boundaries, and if the dilated sinus extends not only beyond the normal anatomical boundaries but also causes erosion of the surrounding bony walls, it is classified as a pneumocoele. Despite the fact that it was first characterised a century ago, the aetiology remains conjectural.

Case reports

Case 1

A 21-year-old woman was admitted to our department with a four-year history of intermittent bilateral nasal obstruction, together with nasal itching, sneezing, rhinorrhoea, and olfactory dysfunction. She had a four-year history of hyperthyroidism, and there was no history of trauma, surgery, or other serious medical illness. Physical examination showed slight facial asymmetry. All laboratory tests were within normal limits, including concentrations of thyroid hormones. Nasal endoscopy showed polyposis in the right middle meatus, septal deviation to the left side, and enlargement of the right maxillary sinus. There was appreciable narrowing of the right nasal cavity as a result of collapse of the medial wall of the right maxillary sinus. Computed tomography (CT) showed moderate enlargement of a well-aerated, right maxillary sinus, with bony thinning (Figs. 1 and 2).

She was treated under general anaesthesia by endoscopic septoplasty polypectomy of the right middle meatus, and right maxillary antrostomy, and a large proportion of the medial wall of the maxillary sinus was resected. There was no evidence of mucosal disease within the maxillary sinus, nor was the sinus obstructed (Fig. 3). Postoperative histopathological examination of the resected maxillary bone showed chronic infiltration of inflammatory cells within the mucosal interstitium and fibrous hyperplasia on the bony surface (Fig. 4). At short-term follow up her symptoms had improved subjectively and she was able to breathe through her nose, but the facial asymmetry still existed. Endoscopic assessment showed a well-healed maxillary sinus with mild mucosal oedema.

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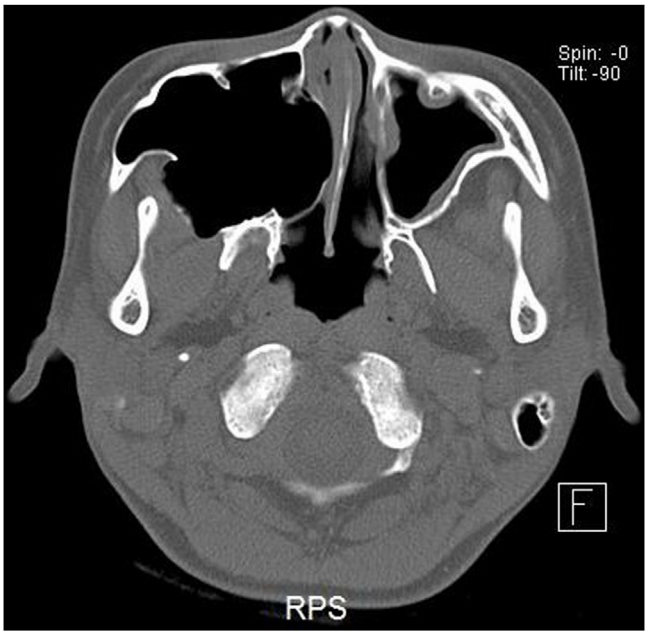

Fig. 1. Axial view of a non-contrast computed tomographic image showing septal deviation and dilatation of the right maxillary sinus with cortical bone thinning into the right nasal cavity.

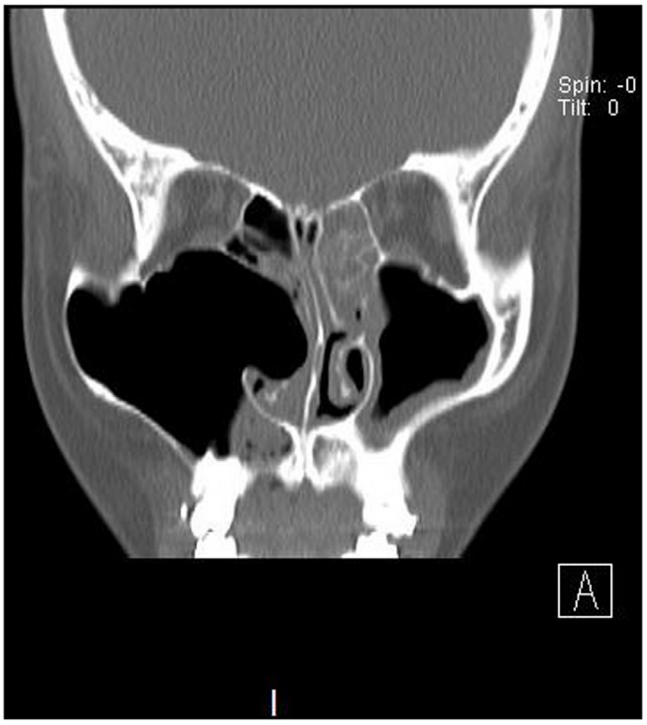

Fig. 2. Coronal view of a non-contrast computed tomographic image showing septal deviation and dilatation of the right maxillary sinus with cortical bone thinning into the right nasal cavity.

Case 2

A previously healthy 16-year-old boy came to our outpatient department complaining of a six-month history of intermittent right nasal obstruction. Clinical examination showed mild bilateral bony swelling of the cheeks with no evidence of diplopia, paraesthesia, or oroantral fistula. A CT scan showed septal deviation to the right as well as bilateral hypertrophy of the inferior turbinates. Pneumatic dilatation of the bilateral

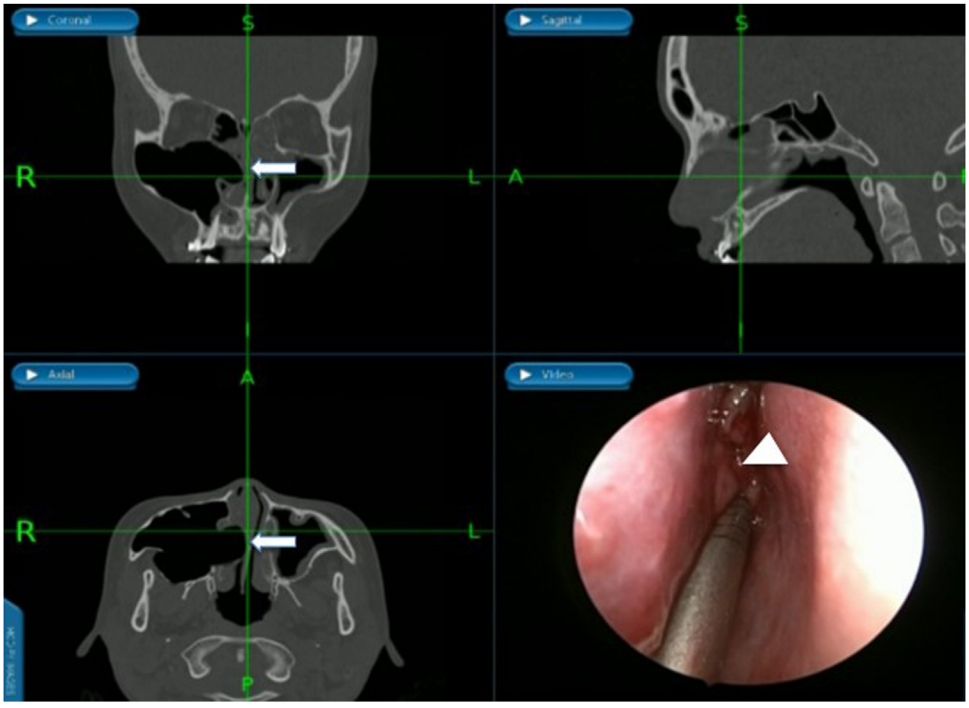

Fig. 3. Intraoperative view of the right middle meatus showing expanding of the right maxillary sinus causing narrowing of the right nasal cavity with no evidence of obstruction.

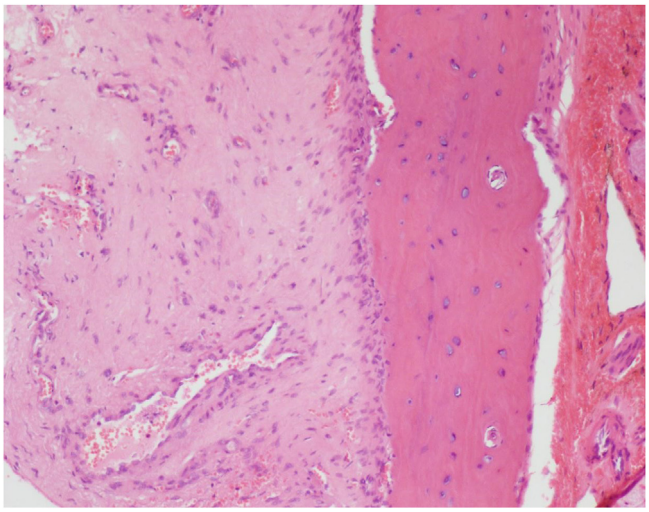

Fig. 4. Histopathological examination of resected maxillary bone showing infiltration with chronic inflammatory cells within the mucosal interstitium and fibrous hyperplasia on the surface of the bone (haematoxylin and eosin, original magnification × 100).

Fig. 5. Axial view of the computed tomographic scan of case 2 showing bilateral dilatation of the maxillary sinuses, with the right side slightly larger than the left, and thinning of cortical bones.

maxillary sinuses was also present, with obvious thinning of the walls (Figs. 5 and 6). The patient refused surgical intervention.

Discussion

Ricci reviewed all the published papers about pathological dilatation of the paranasal sinuses,[3] and summarised the leading theories about its aetiology, which include a

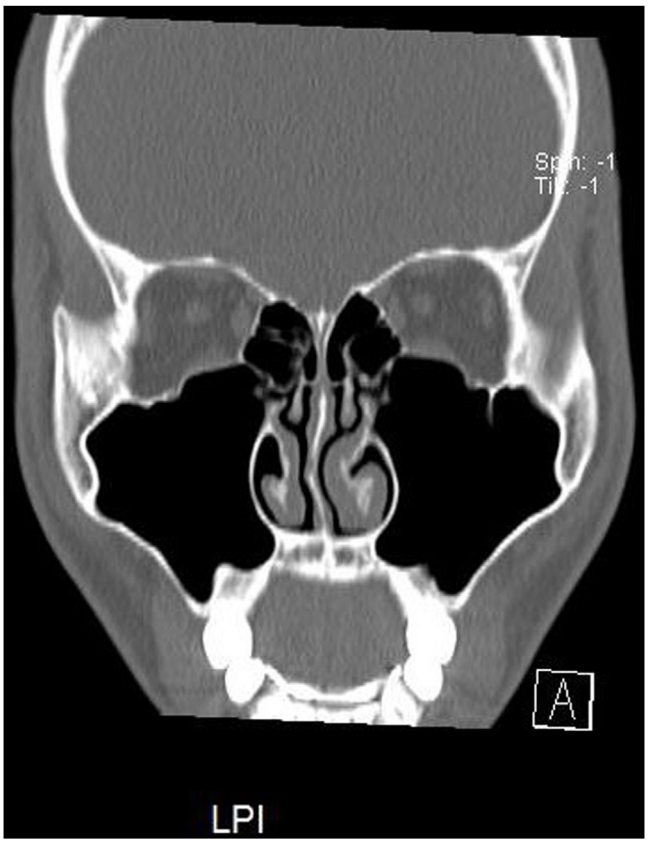

Fig. 6. Coronal view of the computed tomographic scan of case 2 showing bilateral dilatation of the maxillary sinuses, with the right side slightly larger than the left, and thinning of cortical bones.

ball-valve mechanism; fibro-osseous dysregulation; draining mucocoele; genetic predisposition with a hormonal trigger during puberty; gas-forming bacteria; and hormonal dysregulation of bone metabolism. Case1 had an obstructive lesion in the right middle meatus, but the polyp did not obstruct the outflow tract of the maxillary sinus.[4] The one-way valve theory has been tested by Wolfensberger[5] but that did not work in our patients. Most case reports involve only a single sinus,[3] while in our second case, both maxillary sinuses were affected, the right side slightly more than the left.

Pathological examination of the resected maxillary bone in the first case showed fibrous hyperplasia over the bone surface, which is in accordance with the report by Lloyd,[6] indicating that a pathophysiological process within the bony sinus caused the pneumocoele. As well as the pathological findings, it is noteworthy that the first patient had concurrent hyperthyroidism when her nasal symptoms presented at the age of 17, which may have aggravated her hormonal imbalances and increased her predisposition to the disease. Analysis shows that people aged 16-25 are affected two to four times more often than other age groups.[3] Peripubertal patients experience more changes in hormone concentrations, which suggests that hormone imbalances are likely to have a role in the pathophysiology of the disease.

In the first case, the patient did not complain about cosmesis, and therefore her facial asymmetry was left untreated. However, multiple treatments are available for restoration of a normal complexion. Open procedures that involve removal of the anterior sinus wall with or without plates made of either bone or artificial material, were described by Bouguila et al.[7] Endoscopic osteotomy is also a good choice for correcting alterations to the maxillary or orbital contours through Caldwell-Luc and infraorbital approaches, respectively.

The prognosis of a pneumocoele is excellent as we know of no reported cases of relapse, and our first patient reported relief of her symptoms of nasal obstruction during postoperative follow up. Both endoscopic inspection and CT were used for follow-up examinations. While endoscopy provides a direct view of structures within the nasal cavity, CT was better for observing bones.

Conclusion

We have described two cases of maxillary pneumocoele, a rare type of sinus condition the aetiology of which is unknown. The disease is diagnosed when CT shows enlargement of the maxillary sinus and the presence of air alone within the walls of the sinus. Pneumocoele has an excellent prognosis, with various treatments available.

Conflict of interest

We have no conflicts of interest.

Ethics statement/confirmation of patient's permission

Ethics approval not required. The patients' permissions have been obtained.

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