

Rapidly expanding maxillary pneumosinus dilatans*

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SUMMARY

Maxillary pneumosinus dilatans is a rare and difficult to diagnose condition at its outset. The natural history of a case where a computer tomography (CT) scan was performed 6 months prior diagnosis raises the possibility that pneumosinus dilatans and sinus pneumocele may be a continuum of the same disease process. The classification and aetiology are discussed.

Key words: Pneumosinus dilatans, pneumocele, hypersinus, CT.

INTRODUCTION

The abnormal dilatation of paranasal sinuses is a very rare condition usually involving a single sinus cavity. Most frequently affected are the frontal sinus (76%), the ethmoidal sinuses (10%), the sphenoid sinus (10%) with the maxillary sinus being least commonly affected (4%)⁽¹⁾. The condition was first described by Mayes in 1898 but was named pneumatocoele. In 1918, Benjamin coined the term pneumosinus dilatans and described the difference between pneumosinus dilatans and a pneumatocoele. Pneumosinus dilatans is characterized by the benign expansion of an aerated sinus beyond its normal margin. The sinus wall and mucosa remain intact, and retain normal thickness. This differs from a pneumatocoele which defined as a pocket of air in a soft tissue envelope, extending beyond the boundaries of the sinus through a defect in the sinus wall⁽¹⁻⁴⁾. Symptoms at presentation depend partly on the local sinus involved but commonly include toothache, headache, nasal obstruction, reduced visual acuity^(3,5), double vision, cosmetic deformity^(3,4,6), parasthesia and neuropathic pain⁽⁵⁾. Both of these entities must be distinguished from the hyperpneumatized sinus^(3,4) defined as a larger sinus or sinuses without extension beyond normal boundaries, which can be associated with erosion of bony walls and clinical symptoms^(3,4,7). In contrast this condition is usually asymptomatic and usually detected as an incidental finding on a computer tomograph scan (CT) (Figure 1).

CASE REPORT

In 2004, a 33-year-old man presented to the Department of Otorhinolaryngology of Palacky University, Olomouc with a 12-month history of parasthesia in his left cheek and intermittent toothache, along with a 6-month history of facial asymmetry, left nasal obstruction and left exophthalmous. There was

no previous history of upper respiratory infection, sinusitis or head injury. The patient had no significant past medical history, and had never received any nasal or sinus surgery. He had previously had a CT scan of the sinuses performed in another centre which at the time had been reported as normal (Figure 2). Our examination by rhinoscopy showed that the lateral nasal wall including the inferior turbinate was displaced medially towards the nasal septum causing severe compromise of the left nasal airway. A second CT scan (Figure 3) demonstrated massive dilatation of the left maxillary sinus. Treatment comprised endoscopic subtotal resection of the medial wall of maxillary sinus and a subsequent lateralisation of the displaced



Figure 1. Coronal CT scan of hyperpneumatized frontal sinuses.

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Figure 2. Coronal CT scan: Slightly enlarged left maxillary sinus without noticeable bony erosion.

of inferior turbinate. Histologic examination of the resected medial maxillary sinus wall, which included bone and mucosa, disclosed no significant abnormalities. At late follow up 14 months post surgery, the patient remains asymptomatic.

DISCUSSION

Since pneumosinus dilatans is a rare condition occurring as sporadic cases the classification criteria have not been unified. Several previous authors haven't adopted the criteria recommended by Urken, and have described pneumosinus dilatans as an abnormal dilatation of the paranasal sinuses with bony erosion^(1,8). We were fortunate in having an opportunity to compare CT scans in the six-month period prior to diagnosis. In our case the early pre-diagnostic scan shows a noticeable mild enlargement of left maxillary sinus and a change of sinus shape but without bone erosion (Figure 2), this is completely consistent with pneumosinus dilatans. The second scan (Figure 3) demonstrates the characteristic of a pneumocele and exhibits bone erosion. We propose that the pneumosinus dilatans may be the first stage in a continuum of pneumatic dilatation, which without intervention may result in a pneumocele.

It is important to distinguish an idiopathic (or primary) pneumosinus dilatans from secondary pneumosinus dilatans^(3,9). The later condition is associated with planum meningiomas and optic nerve sheath meningioma⁽¹⁰⁾, where tumour invasion causes a weakening of the abutting bone, leading to sinus expansion. The occurrence of secondary pneumosinus dilatans is also reported in association with fibrous dysplasia, ossifying fibroma, Sylvian arachnoid cyst, cerebral hemiatrophy (ex-vacuo pneumosinus dilatans)^(1,4,6,9).

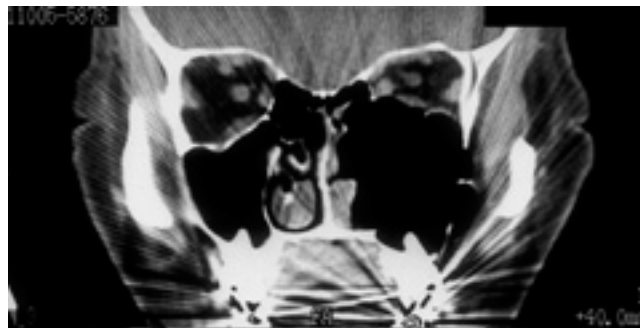


Figure 3. Coronal CT scan: Enlarged air-filled left maxillary sinus with bony erosion.

The pathogenesis of idiopathic (primary) pneumosinus dilatans has been debated by many authors for a considerable period of time. Some theories such as the presence of a spontaneously draining mucocoele, dilatation by a gas-forming microorganism, or a previously unrecognised congenital malformation seem hardly likely. The one-way valve theory perhaps seems the most attractive and even has a limited evidence base. Wolfenger^(6,8) measured the antral pressure before and after Valsalva's manoeuvre on patient with pneumosinus dilatans. He discovered there was higher antral pressure in the affected sinus after manoeuvre and this persisted for several minutes. Some pneumosinus patients describe an increased susceptibility to sinus barotrauma^(3,6). The problem with the theory is that it fails to explain the normal mucociliary clearance of the affected sinuses of these patients. It is possible but only conjecture that the higher pressure in the affected sinus pressure (whether constant or intermittent) maybe leading to an imbalance of osteoblast / osteoclast activity.

CONCLUSION

The aetiology and pathophysiology of pneumosinus dilatans remains obscure. We postulate that pneumosinus dilatans and pneumocele may be a spectrum of the same pneumatic disease process.

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