

Pneumosinus dilatans, pneumocoele or air cyst? A case report and literature review

B M TEH, C HALL, S W CHAN

Department of Otolaryngology, Head and Neck Surgery, Western Health, Melbourne, Victoria, Australia

Abstract

Background: Pathological paranasal sinus expansion secondary to air is uncommon. However, this condition may be symptomatic or cosmetically apparent, requiring surgical intervention. Various terms have been used to describe this condition, and nomenclature is controversial.

Method: An 18-year-old man presented with right facial pain, and was subsequently found to have pneumosinus dilatans of the maxillary sinus. A search was conducted of the PubMed, Medline and Embase databases, using the key words 'pneumosinus dilatans', 'pneumoc(o)ele', 'pneumatoc(o)ele' and 'maxillary sinus'. Articles were also hand-searched. Relevant articles published in English were reviewed.

Results: The literature review identified 36 cases involving the maxillary sinus (including the present case), with 19 cases reported as pneumosinus dilatans, 12 as pneumocoeles, two as pneumatocoeles, two as pneumosinus dilatans multiplex and one as an air cyst. However, based on the strict classification described by Urken *et al.*, the majority of these cases should be reclassified as pneumocoeles.

Conclusion: Whilst the nomenclature of this pathology is confusing, distinctions of terminology do not alter the management. Hence, we support the use of the term 'air cyst', to incorporate all these lesions.

Key words: Paranasal Sinuses; Maxillary Sinus; Diagnosis; Pneumosinus Dilatans; Surgical Procedures, Operative

Introduction

Pathological air expansion of the paranasal sinuses is a rare cause of facial pressure symptoms and visual disturbance. An expansile, cystic lesion of the sinus was first described by Meyes in 1898, and was termed sinus pneumatocoele.¹ The two predominant terms used in the literature are pneumosinus dilatans and pneumocoele, the distinction being based on the presence of bone erosion, as proposed by Benjamin in 1918.¹ Other terms used include pneumatocoele, pneumosinus frontalis, sinus hypertrophy, aerocoele, hyperpneumatisation, sinus ectasia and sinus blistering. Over the years, inconsistencies in terminology definition have led to a tendency to use such terms interchangeably. It is quite possible that most of these lesions represent a spectrum of an essentially similar underlying pathology.

Most recent authors have adopted a classification system based on a spectrum of radiological and clinical findings, as described by Urken *et al.*,² which uses the terms hypersinus, pneumosinus dilatans and pneumocoele.

A hypersinus (an abbreviated form of 'hyper-pneumatised-sinus') is an aerated sinus with normal walls confined within the normal boundaries of the sinus, and is asymptomatic.

Pneumosinus dilatans is an aerated sinus that expands beyond the normal boundaries (either focally or generally), with an intact and normal wall thickness, and is clinically symptomatic.

A pneumocoele is an aerated sinus with thinning of, or a defect in, the bony sinus walls (either focal or generalised), and loss of integrity of the bone.

In addition, pneumosinus dilatans affecting all sinuses has been described as pneumosinus dilatans multiplex.^{3,4} Furthermore, a pneumatocoele is distinguished as a subperiosteal collection of air on the external wall of the sinus, usually due to trauma, infection, tumour or radiotherapy.

Controversy over the correct nomenclature of these abnormalities has resulted in the use of the term 'air cyst' of a paranasal sinus, in order to cover all lesions of this type.⁵

Case report

An 18-year-old, otherwise healthy man presented to an otolaryngology clinic with a three-month history of right facial pain. An initial dental review had led to a right upper molar extraction. However, the pain had progressed, with associated periorbital swelling and right nasal obstruction. There had initially been some bubbling through the extracted tooth socket, but this had settled?

Clinical examination revealed mild bony right cheek swelling, with no evidence of diplopia, paraesthesia or oroantral fistula. There was significant narrowing of the right nasal cavity due to medialisation of the right lateral wall complex posteriorly, including the inferior turbinate, with associated mild oedema.

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A computed tomography scan of the paranasal sinuses (Figure 1) demonstrated pneumatic dilatation of the right maxillary sinus, with bony erosion and a small degree of air within the periosteal fat. The lesion was reported as pneumosinus dilatans, despite obvious bony erosion.

A wide middle meatal antrostomy was performed endoscopically under general anaesthesia, including resection of the posterior inferior turbinate, without complication. There was no evidence of mucosal disease within the maxillary sinus or nasal cavity.

The patient made an uncomplicated recovery, with complete resolution of symptoms.

We conducted a literature search of the PubMed, Medline and Embase databases, using the key words 'pneumosinus dilatans', 'pneumoc(o)ele', 'pneumatoc(o)ele' and 'maxillary sinus'. Articles were also hand-searched. Relevant articles published in English were reviewed. The results of this review are shown in Table I.

Discussion

The maxillary sinus is filled with fluid in utero, and only becomes pneumatized at birth. It grows rapidly during the first three years of life, and again between the ages of seven and 12 years. The adult maxillary sinus is pyramidal in shape.

Enlargement of the maxillary sinus is rare, and can be due to air (pneumosinus dilatans or pneumocele), mucus (mucocele) or tumour.⁶

Pneumosinus dilatans and pneumocoels can arise in any of the paranasal sinuses, and can be bilateral. However, they are relatively unusual in the maxillary sinuses, compared with the frontal and sphenoid sinuses.

In our literature review data, the mean age of presentation was 27 years (range, nine to 62 years), with a sex ratio slightly in favour of males (23 males to 13 females). We identified a total of 36 reported cases (including the present case) involving the maxillary sinus, with 19 cases reported as pneumosinus dilatans, 12 as pneumocoels, two as pneumatocoels, two as pneumosinus dilatans multiplex and one as an air cyst.

However, if the classification proposed by Urken *et al.*² is strictly applied, the presence of bone erosion in the majority of these cases (including our own case) suggests that they should be reclassified as pneumocoels. This implies that lesions at the more severe end of the spectrum are relatively more common in the maxillary sinus. However, an alternative explanation may be that because there is more room for expansion in the maxillary sinus, compared with the frontal and sphenoid sinuses, these lesions become more advanced before the onset of symptoms.

Aeration of the maxillary sinus most commonly results in expansion postero-medially into the infratemporal fossa and superiorly into the orbital floor. The right maxillary sinus is more commonly affected. Clinically, lesions become symptomatic when expansion results in pressure effects on local structures, or becomes cosmetically apparent. The commonest symptoms affecting the maxillary sinus are a cheek mass or swelling, pain, fullness, numbness, facial asymmetry, eye symptoms (e.g. proptosis), and nasal obstruction. Sphenoid lesions may present with increasing loss of vision and reduction of visual fields, and are often found in association with meningiomas. In fact, sphenoid pneumosinus dilatans can represent the first sign of a meningioma at the tuberculum sellae or the planum sphenoidale.^{7,8}

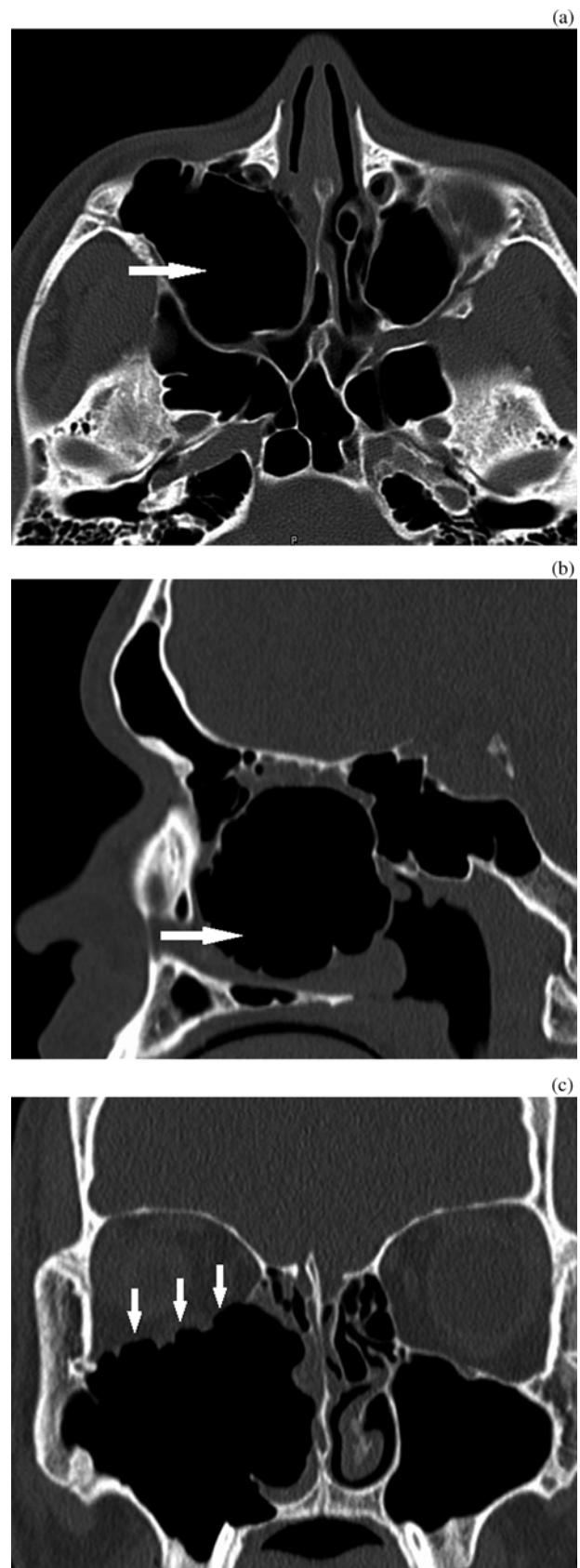


FIG. 1

(a) Axial and (b) sagittal computed tomography (CT) images of a right maxillary air cyst (arrow), reported as pneumosinus dilatans. (c) Coronal CT image showing thinning and erosion of the bone in the roof of the maxillary sinus (arrows), suggesting that this is a pneumocele rather than pneumosinus dilatans.

TABLE I
REPORTED CASES* OF PNEUMOSINUS DILATANS AND PNEUMOCOELE

Study	Pt age (yrs)/sex	Reported as	Location	Presenting symptoms	Radiological description
Noyek & Zizmor ⁹	35/M	Pneumocoele	R max antrum	6 mths: intermittent R cheek pressure & pain	Erosion of inf-lat antral wall
Zizmor <i>et al.</i> ¹⁰	13/F	Pneumocoele	R max sinus	3 mths: R facial swelling, nasal obstruction & proptosis	Thinning of bony wall, bony destruction in antral roof & nasoantral wall
Vines <i>et al.</i> ¹¹	62/F	Pneumocoele	R max sinus with expansion into orbit, med sinus wall & ethmomaxillary angle	5 mths: gradual R eye proptosis	Dehiscence of inf-lat wall
Morrison <i>et al.</i> ¹²	45/M	Pneumocoele	L max sinus extending to zygomatic process and alveolar process	Eustachian tube obstruction & serous otitis media	Erosion of ant cortical margin of inf-lat wall
Meyers & Burtzsch ¹³	25/M	Pneumocoele	R max sinus	3 mths: R cheek swelling, R nasal obstruction	Wall thinning, erosion of inf-lat wall
Lloyd ¹⁴	17/F	Pneumosinus dilatans	Bilat max antra, floor of orbits	Proptosis, prominence of nasolabial fold	Abnormal bone formation
	21/F	Pneumosinus dilatans	Bilat max antra	Bilat exophthalmos, mandible swelling	Bone thickening of both max antra (due to fibrous dysplasia)
Dhillon & Williams ¹⁵	16/F	Pneumosinus dilatans	Bilat max antra	18 mths: R proptosis, some visual loss, fullness of infraorbital region	General expansion of max antra with new bone formation, soft tissue swelling in orbital wall
Wolfensberger ¹⁶	15/M	Pneumocoele	R max sinus	2 mths: upwards displacement of R eye, exophthalmos, dull pain & pressure in R cheek	Hyperlucent, expanded wall with thinning of orbital floor
Komori & Sugisaki ¹⁷	36/F	Pneumosinus dilatans	R max sinus extending to alveolar process	Few yrs: alveolar swelling in R upper quadrant	Thin inf-lat bony wall
Tovi <i>et al.</i> ⁵	20/M	Air cyst	L max sinus	6 yrs: progressive L facial mass	Thin ant sinus wall, outward bulging of lat bony wall
Chan <i>et al.</i> ¹⁸	33/M	Pneumatocoele	R max sinus extending into zygomatic process & infratemporal fossa	R cheek fullness & numbness	Thin post wall of zygomatic process
	51/M	Pneumatocoele	L max sinus to infratemporal fossa	L facial fullness & pain	Bony defect of post-inf aspect of lat wall
Stretch & Poole ¹⁹	17/M	Pneumosinus dilatans	Ethmoid, sphenoid, frontal & max sinuses	4 mths: diplopia, progressive bilat visual loss	
Omnell & Rohlin ²⁰	27/F	Pneumocoele	R max sinus	Midface trauma, palatal asymmetry	Thin bony wall (no erosion), encroachment of nasal & oral cavity
Breidahl <i>et al.</i> ²¹	12/F	Pneumosinus dilatans	R ant-med max sinus wall	6 yrs: R infraorbital swelling	R orbital wall thinning
	42/M	Pneumosinus dilatans	R sup-med max sinus wall, sphenoid, frontal & ethmoid sinuses	Many yrs: R cheek mass, proptosis	Extensive aeration of max sinus extending into orbit
Flanary & Flanary ²²	9/M	Pneumocoele	L max sinus, with erosion of orbital floor & lamina papyracea	L proptosis	Erosion of lamina papyracea & orbital floor
Dillard & Sillers ²³	26/M	Pneumocoele	R max sinus, with med displacement of med wall	5 mths: 'lazy eye'	Zygoma thinning, sup displacement of orbital floor

Hwang <i>et al.</i> ³	27/M	Pneumosinus dilatans multiplex	All paranasal sinuses & mastoid air cells	L malar pain, infraorbital nerve hypoesthesia, facial asymmetry, retruded mandible, malocclusion	Enlargement of all paranasal sinuses, without bone destruction, hyperostosis or mucosal disease
Mauri <i>et al.</i> ²⁴	17/M	Pneumosinus dilatans	L max sinus	3 yrs: progressive L infraorbital swelling, nasal obstruction, pain	Expansion of med bony wall to nasal septum, erosion of ant max wall with an air pocket
Juhl <i>et al.</i> ²⁵	40/M	Pneumosinus dilatans	R max sinus, bulging into alveolar process, sphenoid sinus, nasal cavity & orbit	Few mths: R exophthalmos, facial asymmetry, R eye displacement	Ant & med max sinus wall thinning, expansion into orbit (close to optic nerve & alveolar process)
Ganly & McGuinness ^{26,27}	24/M	Pneumosinus dilatans	All paranasal sinuses especially on R	6 mths: R nasal congestion, max pain, intermittent facial swelling, dull ache of premolar & molar teeth	Erosion of bony septa in ethmoid, R side of sphenoid med & inf orbital wall, & lat max sinus wall
Hsu & Wang ²⁸	24/M	Pneumosinus dilatans	R frontal, ethmoidal & max sinuses	5 mths: progressive L proptosis	Large orbital mass with marginal enhancement
Trimarchi <i>et al.</i> ²⁹	33/F	Pneumosinus dilatans	R max sinus	6 yrs: R nasal obstruction, R max region pain	Normal sinus wall thickness, no loss of wall integrity, med wall of max sinus displaced outward
Dogan <i>et al.</i> ³⁰	25/M	Pneumosinus dilatans	L frontal, max & ethmoidal sinuses	Incidental CT finding	Extensive pneumatization of frontal sinus, some dilatation of ethmoid & max sinuses
Karlidag <i>et al.</i> ³¹	33/M	Pneumosinus dilatans	Bilat ant & med walls of max sinus	10 yrs: slowly enlarging L facial mass	Continuity of max sinus with ant air pockets
Knapp & Klenzner ³²	49/F	Pneumocoele	R max sinus, extending into ventral maxilla & infratemporal fossa	R upper lip & R nasal flank numbness, R cheek pressure, upper jaw dental pain	Sinus wall thinning with bony defect
Sanjari <i>et al.</i> ³³	13/F	Pneumosinus dilatans	Max, frontal, ethmoid & sphenoid sinuses	1.5 yrs: gradual painless bilat loss of vision	Significant expansion of paranasal sinuses
Viehweg & Hudson ³⁴	27/F	Pneumosinus dilatans	Bilat max sinuses extending towards pterygoid plates & cranial base, sphenoid sinus	3–4 mths: bilat swelling in malar eminence & ant sinus wall	Thin ant max wall, incidental retention cyst in L max sinus
Kiroglu <i>et al.</i> ⁴	21/M	Pneumosinus dilatans multiplex	All paranasal sinuses & mastoid air cells (bilat)	Seizure, facial deformity	All paranasal enlargement without bony destruction, hyperostosis or mucous membrane thickening
Nayak <i>et al.</i> ³⁵	20/M	Pneumosinus dilatans	Ant-med aspect of R max sinus	4 mths: R facial pain	Ant-med pneumatization without bone thickening or thinning
Vlckova & White ³⁶	33/M	Pneumosinus dilatans progressing to pneumocoele	L max sinus	12 mths: L facial paraesthesia & intermittent toothache 6 mths: facial asymmetry, L nasal obstruction, L exophthalmos	Initial imaging showed slightly enlarged max sinus with no bony erosion; later progression to significant enlargement with bony erosion
Lombardi <i>et al.</i> ³⁷	28/M	Pneumosinus dilatans	All paranasal sinuses	L max pain	Progressive rapid enlargement of max sinus, with discrete signs of orbital floor ‘blowing-up’
Braverman ³⁸	14/F	Pneumocoele	R max sinus	6 mths: R hemifacial pressure, numbness, rhinitis, exophthalmos	Extremely large max sinus, causing ballooning effect on med wall & obstruction of nasal passage
Present case	18/M	Pneumosinus dilatans	R max sinus	3 mths: R facial pain	Erosion of R middle & post ethmoid air cells, middle & inf turbinates, floor of orbit, & lat max wall

*Published in English. Pt = patient; yrs = years; M = male; R = right; max = maxillary; mths = months; inf = inferior; lat = lateral; F = female; med = medial; L = left; ant = anterior; bilat = bilateral; post = posterior; sup = superior; CT = computed tomography

The aetiology of paranasal sinus air cysts remains something of an enigma. Generally, they are described as primary or secondary. The proposed aetiologies of primary (idiopathic) cases include a one-way valve effect, gas-forming organisms, spontaneously discharged mucocele and dysregulation of hormones.

Many authors have suggested a one-way valve mechanism. This is supported by the presence of polypoid mucosa in the drainage pathway of the affected sinus in some cases.³⁹ The concept may also explain increasing symptoms associated with air travel and atmospheric pressure changes, or during sneezing. Previous authors have documented elevation of sinus pressures;^{16,38} the bubbling of gas through the extracted tooth socket in our patient's case would support this. However, most cases do not demonstrate sinus outflow obstruction, and the question as to why the sinus does not fill with mucus is still debated. The theory is also contradicted by the presence of focal expansions within the sinus in some cases.³⁵ The raised intra-sinus pressure may represent an end-stage effect of the process when the sinus has expanded to such an extent that it has occluded its own ostium, rather than representing a true causative phenomenon.

Other authors have suggested infection with a gas-forming organism as a possible aetiological factor; however, such an organism is yet to be identified.⁴⁰ It is a feature of these abnormalities that the mucosa has a normal appearance, with functioning cilia.^{2,37}

Some authors have described symptoms of sudden, spontaneous nasal discharge, indicating the possibility of an air cyst forming from a discharged mucocele.^{41,42}

Further theories relate to the dysregulation of sex and growth hormones, resulting in the stimulation of osteoblastic and osteoclastic activities, producing abnormal expansion.⁴³ However, no hormonal alteration has thus far been recorded.⁴⁴

- **Pathological air expansion of the maxillary sinus is relatively rare, with only 36 previously reported cases**
- **The nomenclature is confusing; the commonest terms are pneumosinus dilatans and pneumocele**
- **The main difference between these two conditions is radiological, and does not alter management**
- **Use of the single term 'air cyst' to describe these lesions is supported**

In syndromic (secondary) cases, observations particularly relating to the frontal sinus have noted over-development of the sinus occurring in association with under-development of the brain (e.g. in cranio-cerebral hemiatrophy),⁴⁵ or due to long-term shunting resulting in decreased intracranial pressure, hence allowing for expansion of the sinuses.⁴⁶ Air cysts have also been described in association with Melnick–Needles syndrome,¹⁹ Klippel–Trenaunay–Weber syndrome⁴⁷ and arachnoid cysts.^{48,49} Two of the five reported cases of pneumosinus dilatans multiplex were associated with mental retardation and facial deformity.^{3,4}

The question remains as to whether pneumocele and pneumosinus dilatans represent a progression of a single pathological process, or whether they have separate aetiologies. Hitherto, there have only been two documented cases

of pneumosinus dilatans progressing to pneumocele: one in the frontal sinus⁵⁰ and the other in the maxillary sinus.³⁶ In general, the onset of pneumosinus dilatans tends to be chronic (developing over years), as opposed to pneumocele, the symptoms of which develop quickly over a number of months. However, this may simply be due to pneumocele expansion reaching a critical size at which symptoms rapidly progress.

Computed tomography imaging is the most important modality in the diagnosis of these lesions. Magnetic resonance imaging should be used to exclude concomitant intracranial lesions in sphenoid and frontal lesions. Noyek *et al.*⁵¹ studied the radiological signs in four cases of pneumocele; all showed an enlarged sinus, thinning of the sinus wall, hyperaeration, hyperlucency, bony dehiscence, expansion of recesses, effacement of the ethmoidmaxillary angle and displacement of the middle meatus. Three of the four cases demonstrated profiling of dental roots and displacement of the orbital floor. In our literature review, 12 cases showed thinning of the sinus wall, most commonly the anterior and orbital wall.^{5,10,13,16–18,20,21,23,25,32,34} Only one case described bone thickening instead of thinning.¹⁴

Symptomatic lesions require surgical intervention. The commonest procedure performed is creation of a naso-antral window, aiming to equilibrate the intrasinus pressure. Thus far, no recurrence has been reported in treated lesions.

Conclusion

This report describes a case of pathological paranasal sinus expansion secondary to air, a rare but clinically significant type of sinus pathology. It also highlights the confusion regarding the nomenclature of such lesions. While it is possible that pneumosinus dilatans and pneumocele represent distinct pathological entities, the distinction is based more on radiological than clinical features. Furthermore, the distinction does not alter the management in the event of symptomatic cases, which comprises surgical decompression usually through enlargement of the anatomical ostium. We therefore support the use of a single term, such as 'aircyst', to describe these lesions.⁵

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Address for correspondence:

Dr Bing Mei Teh,
School of Surgery,
UWA,
M507, 35 Stirling Highway,
Crawley, Western Australia,
Australia 6009

Fax: +61 (0)893 464374
E-mail: bm_teh@yahoo.co.uk

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