The Journal of Laryngology and Otology
August 1987. Vol. **101.** pp. 828–832

Clinical Records

Pneumosinus dilatans

by

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Introduction

Pneumosinus dilatans is an abormal dilatation of the paranasal sinuses containing only air and lined by normal mucosa. The presenting symptoms are usually ocular. The association with fibro-osseous disease and meningiomata of the optic chiasmal region is discussed.

Case report

PH, a 16-year-old schoolgirl, presented with an eighteen-month history of right-sided proptosis with some diminution of vision. Clinically there was an obvious proptosis (Fig. 1) with slight fullness of the infra-orbital region. Ophthalmologically she had early papilloedema and an enlarged blind spot and a reduction in visual acuity.

Plain radiology revealed general expansion of both maxillary antra with new bone formation. On the right side a soft-tissue mass involving the orbital floor was clearly appreciated but a smaller less apparent one on the left was missed (Figs. 2 and 3).

A right Caldwell-Luc procedure was used to approach the lesion which was found to involve the anterior wall of the maxilla. A second incision was made in the infra-orbital margin to approach the orbital floor. The orbital contents were markedly displaced but the orbital periosteum was not breached. Piecemeal removal was carried out and material sent for histology. Histological examination of the lesion showed irregular foci of woven bone with scanty adjacent oesteoblasts set in a cellular fibrous stroma containing a sprinkling of inflammatory cells and numerous giant cells. The features were interpreted as fibrous dysplasia of bone.

Two weeks post-operatively her proptosis had completely subsided and the visual acuity and blind spot had returned to normal. There was some residual scarring on the nasal border of the retina but the papilloedema had resolved.

Twenty months later she had a further complaint of right cheek fullness. A CT scan (Fig. 4) showed the right-sided surgical defect but additionally a soft-tissue mass along the inferolateral area of the left orbit. The original plain X-rays were reviewed and the small mass was now appreciated.

It was felt that the left-sided lesion was similar to the right and as the patient was asymptomatic surgery was deferred.

Discussion

Pneumosinus dilatans is described as an abnormal dilatation of the paranasal sinuses which contain only air and are lined by normal mucosa. The first recorded case was described by Benjamins (1918), from Utrecht, involving the frontal sinus.

Lombardi (1967) reported 51 cases. He showed that the condition had a predilection for certain sites: the lateral recesses of the frontal sinuses, the superior recess of the maxillary antrum, the sellar region of the spheno-ethmoidal complex and the orbit in the anterior ethmoidal complex.

The association of pneumosinus dilatans and meningiomata of the anterior chiasmatic angle, tuberculum sellae and planum sphenoidale is well recognized. Lloyd (1985) has recently reported 6 cases of which three were associated with meningoma, presenting with either unilateral or bilateral exophthalmos. Wiggli and Oberson (1975) had 7 patients with anterior chiasmatic angle meningiomas all with ocular symptoms and radiological changes in the planum sphenoidale. Spoor *et al.* (1981) had a case of pneumosinus dilatans in combination with the 'Klippel-Trenaunay-Weber syndrome' (port-wine haemangiomas, varicose veins

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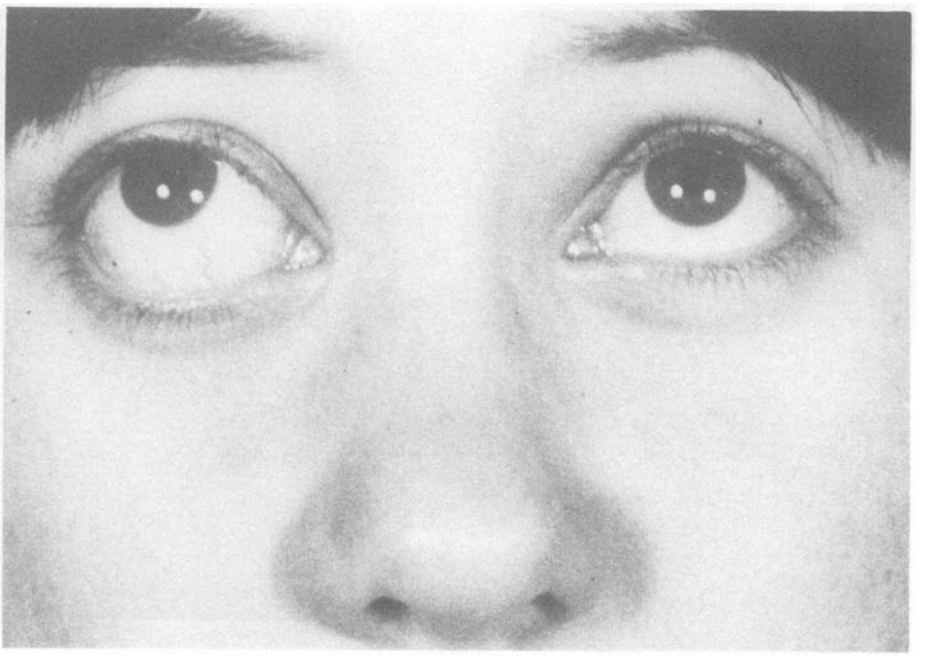


FIG. 1
Obvious right-sided proptosis.

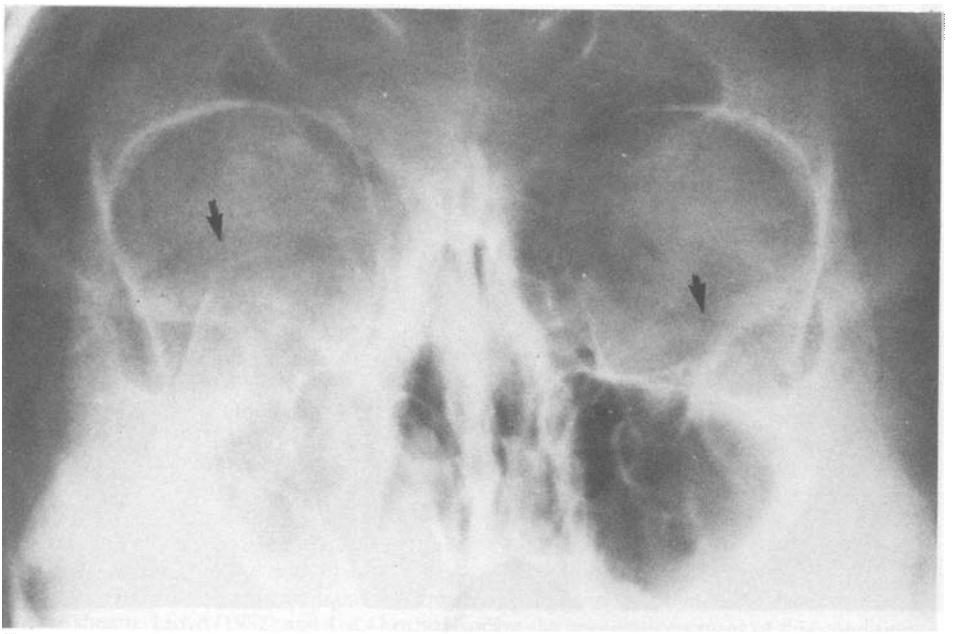


FIG. 2
Plain radiograph showing large maxillary antra. The arrow on the right shows the fibro-osseus lesion. A similar lesion on the left is not so clearly illustrated.

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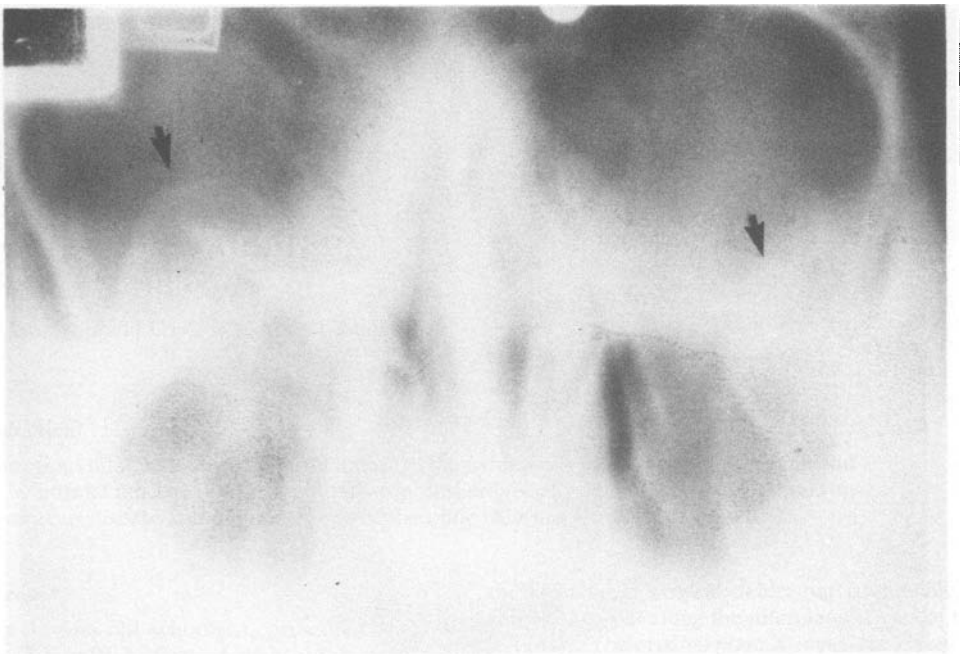


FIG. 3
Plain tomogram. The soft-tissue swellings of the orbital floors are more clearly visualized.

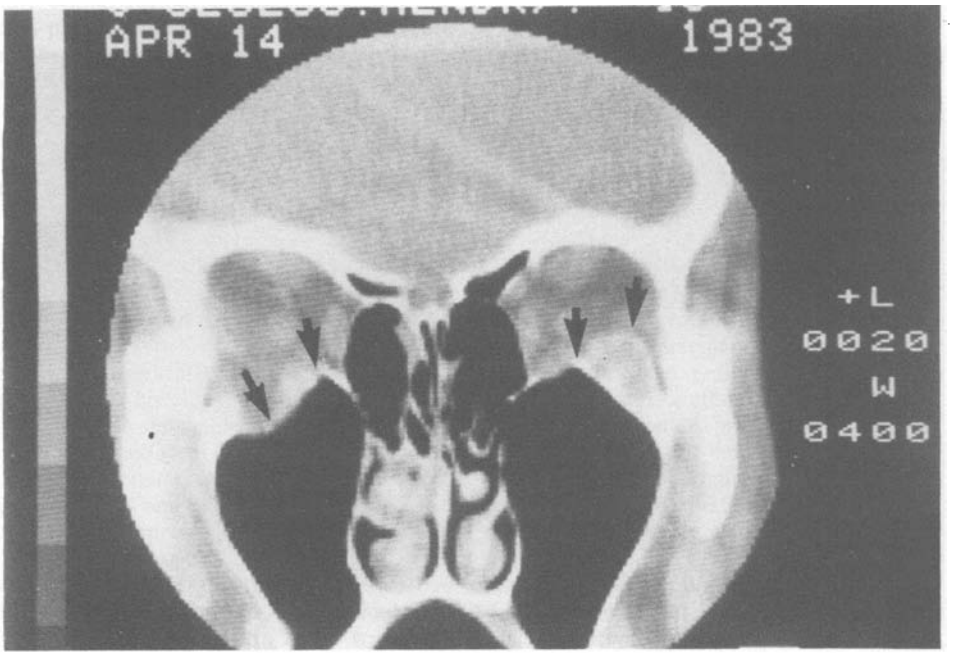


FIG. 4
Post-operative CT scan. The medial arrows reveal the superior expansion of both antra. The right lateral arrow shows the operative defect in the orbital floor. The left lateral arrow reveals the soft-tissue swelling in the orbital floor.

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FIG. 5

A lateral radiograph showing the blistering of the spheno-ethmoidal region (lower arrow) with hyperostosis, in a case of sellar meningioma.

and soft tissue and bony hypertrophy) and familial Adie's syndrome, who developed bilateral perioptic meningiomas.

Only Lloyd (1985) mentions the association of fibro-osseus disease in pneumosinus dilatans. This was noted in three of six cases, the maxillary antrum bilaterally in two and the fronto-ethmoidal in one. All three presented with proptosis. Lloyd states that the presence of fibro-osseous pathology is easily recognized by plain radiology.

The commonest radiological appearance encountered in cases of pneumosinus dilatans associated with optic meningiomata or fibro-osseous disease is 'blistering' or 'hyperostosis' of the tuberculum sellae or planum sphenoidale. This is best visualized on a true lateral plain X-ray and was first described by Wiggli and Oberson (1975). These features include a localized deformity of the spheno-ethmoidal line 'blistering', overlying which the bone is thickened ('hyperostotic') (Fig. 5).

Hirst *et al.* (1982) state simply that a neuroradiological finding of a dilated sphenoid sinus, even without hyperostosis, but with occular symptoms is highly suggestive of a meningioma of the optic nerve sheath. Lloyd (1982) had 4 of 41 orbital meningiomata arising from the optic chiasm in the region of the tuberculum sellae. All 4 cases showed hyperostosis of the planum sphenoidale.

It is important to appreciate that pneumatization of the sinuses varies greatly. The sphenoid sinus is the most variable; in some instances the whole bone may be pneumatized including the clinoid and ptyergoid processes, greater wings of the sphenoid and even the basilar process of the occipital bone. Focal enlargement of the sphenoid sinus can occur with or without bony thickening in cases of acromegaly and agenesis of a cerebral hemisphere. A relative overgrowth of the antra occurs in microcephaly and in congenital anophthalmos.

The pathophysiology of sinus dilatation is ill understood. An abnormality of bony growth and remodelling produced by the meningioma or fibro-osseus disease is likely. Wiggli and Oberson (1975) suggested that a protracted period of low pressure could produce a bone remodelling process leading to a predominant osteoblastic activity and sinus expansion at the site of hyperostosis (the area of least resistance). A one-way valve type obstruction of the sinus (Sugita *et al.*, 1977), a form of osseous reorganisation (Seutin *et al.*, 1979) and a stimulating effect of a local meningioma (Hirst *et al.*, 1982) are the speculative causes of this condition.

For the otolaryngologist this case serves to illustrate that pneumosinus dilatans usualy presents with ocular symptoms and has two major causes.

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Fibro-osseus disease is easily recognized but the changes associated with meningiomas may be more subtle and require the latest imaging techniques, such as CT or MRI.

Acknowledgements

The authors would like to thank Mr. G. W. Glover, Consultant ENT surgeon, Mount Vernon Hospital, for permission to report details of a patient under his care and Dr. G. Lloyd, Consultant Radiologist, Royal National Throat, Nose and Ear Hospital for his advice.

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