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Air cyst of the maxillary sinus (pneumosinus dilatans, pneumocoele)

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Abstract
A cystic dilatation of a paranasal sinus by an entrapped air mass has been termed pneumosinus dilatans or a pneumocoele. The occurrence of this condition in the maxillary sinus is quite rare. In the present case, a mid-face mass mimicking an osteoma was the presenting sign of the disease. The pathogenesis, clinical features and radiological aspects of this entity are reviewed. In order to avoid confusion with other conditions of different aetiopathogenesis, we propose to label the lesion as 'air cyst' of the paranasal sinus, a term that best describes the nature of the lesion.

Introduction

A tumoural mass in the mid-face usually represents a cutaneous or mesenchymal tissue neoplasm, a developmental or dentigerous cyst or an expansile inflammatory or neoplastic process of the paranasal sinuses. A cystic dilatation of the maxillary sinus by an air mass, presenting as a solid tumour in the mid-face, should be a very unusual occurrence. In this article, in addition to the following case report, previous descriptions of this rare entity will be reviewed.

Case report

A 20-year-old male was referred for evaluation of a slowly developing mass in the left side of the face. He had no history of trauma, sinus headaches or allergy. He was aware of the growth six years before presentation.

Physical examination revealed a fixed, hard, asymptomatic tumoural mass of 2.5 × 1.5 cm in the left cheek (Fig. 1). Rhinoscopy showed no abnormality, and the rest of the head and neck examination was normal. Results of routine blood count, blood chemistry and urine analysis were normal. On X-ray examination, the paranasal sinuses appeared clear and well developed. Computed tomography (CT) showed anterior expansion of the left maxillary sinus displaying thinned bony walls. No mucosal abnormality was detected (Fig. 2).

Exploration of the left maxillary sinus was performed by the Caldwell-Luc approach. The anterior sinus wall was thinned and outwardly bulging. The sinus itself appeared remarkably large and lined with an apparently normal mucosa. No abnormality was detected in the sinus ostium area. The bulging of the anterior bony wall was removed. A large naso-antral window was created in the area of the left inferior meatus. The patient remained with a normal facial configuration. The histopathological examination of the removed bony and mucosal sinus wall disclosed no abnormality.

Discussion

The notion of normality as applied to the size of paranasal sinuses is a highly elastic one (Lombardi, *et al.*, 1968). Variations in shape and size may be frequently noted in the maxillary sinuses. Sinus size depends on developmental factors and may be influenced by acquired conditions. It may also become modified by local enlargements such as palatine, zygomatic,

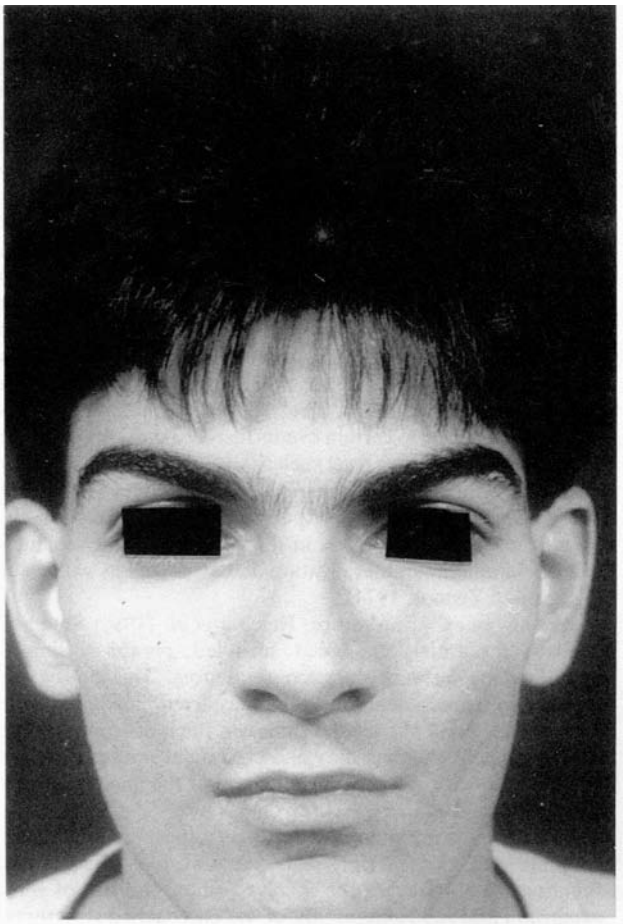

FIG. 1
Note the left paranasal tumoural mass.

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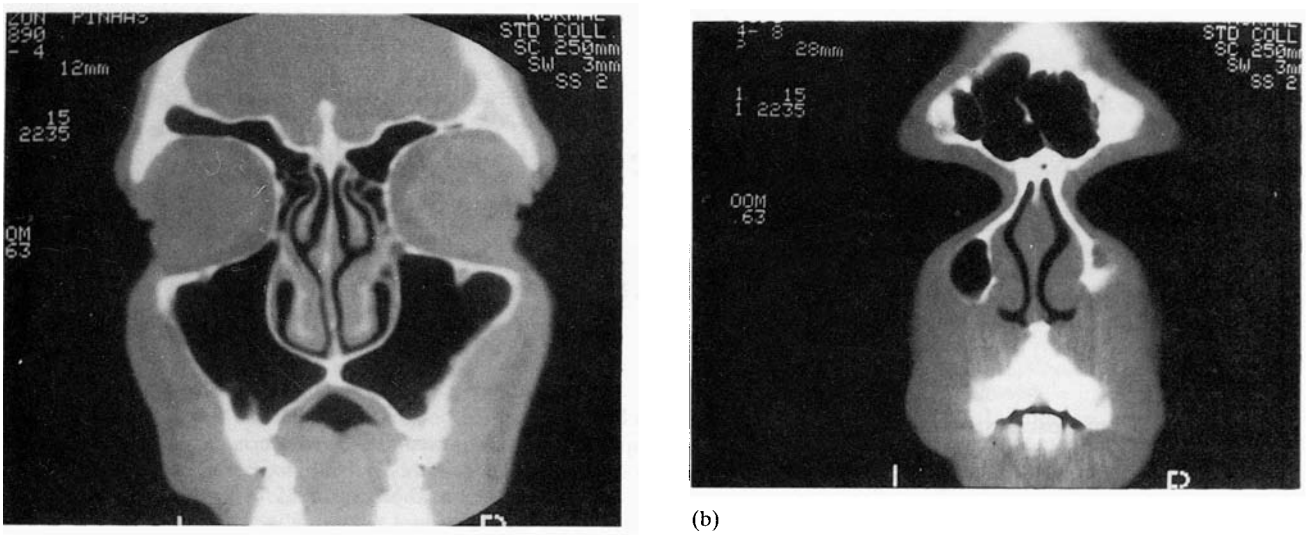

(a) (b)

FIG. 2

Coronal CT of the paranasal sinuses. (a) The left maxillary sinus displays a thinned and outward bulging of the lateral bony wall. (b) A superficial slice shows the anterior expansion of the left maxillary antrum

orbital and alveolar process extensions. An enlarged maxillary sinus is considered as a variant of the normal unless it is well modelled into the surrounding structures (Alberti, 1974). Abnormal expansion of an air-filled paranasal sinus, without overt mucosal changes, encroaching the adjacent tissues has been termed as pneumosinus dilatans (Benjamin, 1918; Lombardi, *et al.*, 1968) or pneumocoele (Noyek and Zismor, 1974; Zismor, *et al.*, 1975). The origin of this entity remains poorly understood. A one-way check valve mechanism at the sinus ostium area, probably due to redundant mucosa (Noyek and Zismor, 1974; Wolfenberger and Herrmann, 1987) or to a minor inflammatory process (Vines *et al.*, 1976), has been proposed for the aetiopathogenesis of this condition. In only one case, a stricture of the ostium has been identified (Meyers and Burtschi, 1980). As a result of such a block, the affected sinus will fail to equilibrate its air pressure through the ostium (Noyek and Zismor, 1974). The repeated air trapping over a period of time, due to certain events increasing the air pressure in the upper respiratory tract such as sneezing, autoinflation or coughing, will result in progressive dilatation of the paranasal sinus, comparable to the expansion of a cyst elsewhere. Because of pressure necrosis in the sinus wall, bony changes such as thinning, demineralization and even defects will occur (Noyek and Zismor, 1974).

In their review of literature, Reicher *et al.* (1986) found 60 cases of abnormally expanded, air-filled, paranasal sinuses with differing incidence and consequences. This entity was confined to one or a few sinuses and occurred mostly in young adult males. While the frontal sinus was the most common site of involvement, the maxillary and ethmoidal sinuses were the least affected. Until today, involvement of the maxillary sinus has been reported in eight cases.

Whatever the terminology used, pneumosinus dilatans or pneumocoele, these lesions manifest a variety of signs and symptoms, according to the involved sinus, the site of sinus expansion and the vulnerability of the encroached structures. Facial swelling, medial deviation of the naso-antral walls, middle ear effusions, herniation of a cystic mass through the defective sinus wall and proptosis were reported as signs of maxillary sinus pneumocoele (Morrison *et al.*, 1975). Facial pain and otalgia exacerbated by rapid changes in altitude, by sneezing or by autoinflation, were experienced by some of the patients. Others complained of nasal obstruction or fullness of the ear. A tumoural mass mimicking an osteoma, as in our case, should be an uncommon feature of the entity. A similar presentation of the anomaly has been mentioned in the German literature (Lombardi, *et al.*, 1968).

Radiographically, pneumosinus dilatans and pneumocoele share the same features (Reicher, *et al.*, 1986). They are characterized by an expansile, hyperlucent paranasal sinus with thinned body walls exhibiting outward ballooning and demineralization. The sinus recesses are expanded. The mucoperiosteum is usually intact. Som *et al.* (1983) tried to differentiate between pneumosinus dilatans and pneumocoele by the finding of bone erosion as occurring in the latter. This view was recently contradicted by Reicher *et al.* (1986) which described bone defects in cases of pneumosinus dilatans. Large sinuses may be confused with cystic dilatation of the sinus, but they do not present with thinning and erosion in their walls. Furthermore, they are asymptomatic.

Maxillary sinus pneumocoeles benefit from the creation of the nasoantral window. Cases described in the literature remained symptom-free following this procedure. Deformities resulting from sinus wall displacement can be corrected by osteotomy (Zismor *et al.*, 1975), as was done in our case.

It is worthwhile to note that the terms pneumocoele and pneumosinus dilatans, which apply to the description of a cystic dilatation of a paranasal sinus by an entrapped air mass, have also been applied to some other different conditions. Intracranial or extracranial air masses stemming from an acquired or congenital bone defect in the pneumatic spaces of the skull, have been termed as pneumatocoele or aerocoele (Dandy, 1926; Levenson *et al.*, 1989). Pneumosinus dilatans have been used in cases of paranasal sinus enlargement secondary to acromegaly, cerebral hemiatrophy, optic chiasm meningioma and orbital exenteration (Lombardi *et al.*, 1968; Reicher *et al.*, 1986; Levinson *et al.*, 1989).

The term 'pneumocoele' refers to a morphological state characterized by the presence of an air mass, not necessarily in the form of a cyst. Furthermore, this term does not reflect the dynamism of a cystic process of the type we have described. On the other hand, since the enlargement of a cyst is a natural occurrence, it is unnecessary to re-emphasize this behaviour as in the term of 'pneumosinus dilatans'. Due to the fact that pneumocoele and pneumosinus dilatans are considered as similar or at least variants of the same entity (Reicher *et al.*, 1986), we propose to label both conditions as 'air cyst' of a paranasal sinus. This term best describes the nature of the

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disease and avoids confusion with the above cited lesions of different aetiopathogenesis.

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