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PNEUMOSINUS DILATANS: A CASE REPORT

Mohamed Ali GLITI[1,*], Niema Benkhraba[2], Sophia Nitassi[3], Bencheikh Razika[4], Benbouzid Mohamed Anas[5], Abdelilah Oujilal[6], Leila Essakalli Houssyni[7]

ENT department, rabat specialty hospital, ibn sina university hospital, hafiane-cherkaoui avenue, 10100 rabat, morocco
*[*1]Resident physician in otorhinolaryngology, department of otorhinolaryngology, head and neck surgery, ibn sina university hospital, rabat, morocco.*
[23]Professor of otorhinolaryngology, department of otorhinolaryngology, head and neck surgery, ibn sina university hospital, rabat, morocco.
[4567]Faculty of medicine and pharmacy of rabat, Mohammed V University, rabat, morocco.

Corresponding Author:-

ABSTRACT:

Frontal *sinuses that present with abnormally large size are often clinically described as a deformity of the forehead, caused by Pneumosinus dilatans (PD). We report a rare case of Pneumosinus dilatans frontalis which was accidentally discovered by a paranasal sinus CT scan requested following frontal headaches, and thus revealing diffuse osteoporosis. Pneumosinus dilatans is often an unexpected radiological finding; however, in most cases, it is diagnosed by recognition of a frontal deformity*.

Keywords: *Pneumo sinus dilatans, sinusitis, osteoporosis.*

RESUME:

Les sinus frontaux qui se présentent d'une taille anormalement volumineuse sont souvent décrits cliniquement par une déformation du front , causé par Pneumosinus dilatans. Nous rapportons un cas rare de Pneumosinus dilatans frontalis qui a été découvert accidentellement par un scanner des sinus paranasaux demandé suite à des céphalées frontales, et révélant ainsi une ostéroporese diffuse. La Pneumosinus dilatans (PD) est souvent une découverte radiologique inattendue; cependant, dans la plupart des cas, elle est diagnostiquée par la reconnaissance d'une déformation frontale.

Mots clefs: *Pneumosinus dilatans, sinusite, ostéoporose.*

INTRODUCTION:

Pneumosinus dilatans (PD) is a very rare condition which most often results in a deformity of the face and is caused by abnormal dilation of the paranasal sinuses, especially the frontal sinuses, which contain only air and are lined by a mucous membrane normal. Meyes first described the disease now known as PD in 1898 [1]. However, in 1918, Benjamin first assigned the name Pneumosinus dilatans to this facial deformity [2]. A review of the literature by Borelli et al in 1979 revealed only 51 reported cases of PD. [3]. In recent years, a few more cases have been reported. Most involved the frontal sinus, although the maxillary sinus, ethmoid cells, and sphenoid sinus may also be affected [4-6].

Abnormal expansions in the dimensions of the frontal sinuses have also been diagnosed as pneumoceles, pneumosinus frontalis, sinus enlargement, aerocele, hyperpneumatization, and sinus ectasia. These terms may describe the same clinical condition. Urken et al. [7] classified pneumatic dilations of the paranasal sinuses into three categories based on clinical symptoms, radiological widening, and thinning of the sinus walls. In order of increasing severity, the classifications were hypersinus, pneumosinus dilatans, and pneumocele. However, it is not known how these diseases are linked.

The etiology and pathogenesis of this unusual process are not yet determined. PD is often an accidental radiologic discovery; however, in most cases, it is diagnosed by recognition of a frontal deformity. Common complaints in patients include local pain caused by tissue expansion or frontal nerve compression, headache, frontal paresthesia or sensory deficit, olfactory loss, and ophthalmic disturbances. Stretch and Poole also reported a case of blindness associated with PD caused by osteodysplasia of the optic canals and compression of the optic nerves [8].

X-rays, clinical findings, and hormonal tests can help differentiate PD from other acquired deformities of the frontal bones, such as acromegaly, exostosis, and frontal mucocele. PD has been described both in an isolated form and in association with other diseases, such as arachnoid cyst, [9] cerebral hemiatrophy, [10] meningioma, [5, 11, 12] and KlippelTrenaunay-Weber syndrome [13].

CASE REPORT:

This is a 63-year-old patient with no notable pathological history who is followed in the consultation ward of the otorhinolaryngology department for management of a frontal headache estimated at 5 by the visual analog scale, which dates back to 3 years, with no notion of craniofacial trauma or surgery. The clinical examination found no deformation of the face, particularly the frontal one, no notion of rhinorrhea or epistaxis was reported and the rhinological and oropharyngeal, clinical, and endoscopic examinations were unremarkable. The patient was put on analgesics containing paracetamol at a dose of one gram every six hours for 15 days and corticosteroid therapy by nasal spray containing Fluticasone for 2 months.

Given the non-improvement in symptoms with the prescribed treatment and the normality of clinical and endoscopic examinations, a nasosinusal CT scan was requested showing an aspect of dilation of bilateral frontal sinuses with diffuse bone demineralization (Figure1).

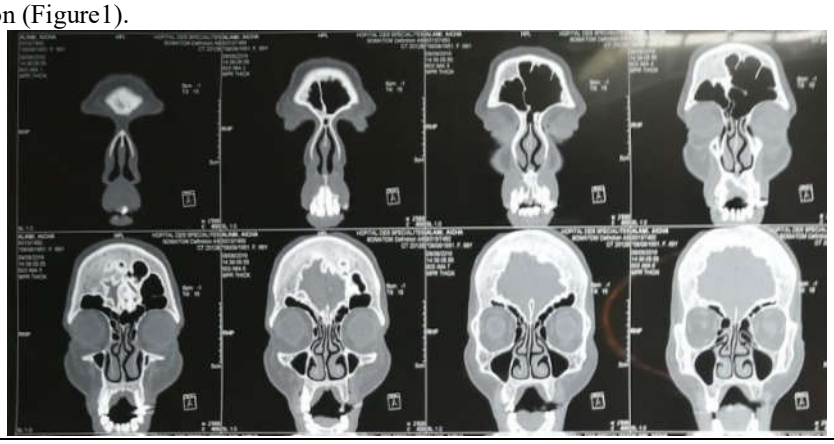

Figure 1:CT image in axial section through a bone window showing the broad dimensions of the two frontal sinuses

The patient was referred and taken care of in the rheumatology department where the diagnosis of severe osteoporosis was retained after performing bone densitometry with a T score of less than minus 2.5. The patient was reviewed six months later with marked improvement in pain (2 on the visual analog scale) after initiation of treatment.

DISCUSSION:

The etiology and pathogenesis of PD are still not well established. Various hypotheses have been proposed to explain its pathogenesis. Benedikt et al have proposed the spontaneous drainage of a frontal mucocele as a pathogenic mechanism [14]. Other authors have described cases in which patients report a spontaneous and profuse nasal discharge, supporting this mechanism [7]. However, most patients do not describe such symptomatology in their pathological history.

Alterations and changes in the levels of growth hormones and sex hormones have also been implicated in PD [15]. Such alterations lead to disruption of the control of osteoblastic activity, leading to excessive bone growth and abnormal pneumatization and expansion of the sinuses. Unfortunately, there is no evidence to support this theory, as stated by Smith et al. [16] who did not record any hormonal alteration in the case they reported. Prott, who also found no hormonal alterations in his patients, suggested that sinus enlargement may be secondary to increased intrasinus pressure [17]. This increase in pressure could be caused by an efferent obstruction of the duct with a valve mechanism; Harrison and Young described this theory in 1955 [18]. However, few simple and anatomically demonstrable valves have been reported by

other authors. Candan et al. demonstrated an increase in intrasinus pressure caused by a non-return valve mechanism [19]. Carta et al. reported a case of PD with diplopia, proptosis, ophthalmoplegia, and compressive optic neuropathy [20]. The authors attributed amaurosis to changes in atmospheric pressure. These changes, caused by probable valve obstruction of the frontal sinus, resulted in compression of the optic nerve. Endoscopic permeabilization of the nasal-antral communication resulted in a complete recovery.

Urken et al. have also discussed the theory of the valve mechanism [7]. However, they were unable to formulate a clear and possible etiology and pathogenesis of PD, despite reviewing a large number of asymptomatic patients and six reported cases of PD.

Despite the confusion regarding the pathogenesis and classification of PD, the treatment options for treating PD are quite clear. In hypertensive patients with symptomatic PD but without significant frontal lumps, the treatment of choice appears to be functional endoscopic sinus surgery aimed at treating sinus duct obstruction. When the frontal boss is the main complaint, endoscopic surgery is not effective; in such cases, open surgery with an osteoplastic flap is indicated.

Various open procedures have been described and most of them are effective. However, increased attention to aesthetic results requires technical improvement. Prott described the fracturing of the frontal bone, but the aesthetic results are unpleasant [17]. Urken et al. adopted frontal bossing treatment by creating an osteoplastic flap, with a recession of the anterior table into the sinus, followed by fatty obliteration of the remaining cavity [7]. Cosmetic results have not been reported. Pospisil and Balmer have proposed a frontal osteotomy with osteotomy [21]. The space created by the size difference between the remaining reduced anterior frontal bone and the original sinus margins was filled with bone chips and Surgicel. However, a large deformity results in a difference in curvature, which may require excessive fragmentation of the anterior plate to accommodate the frontal bone. We believe that a radial or 'alternative' osteotomy (a series of alternating osteotomies on opposite sides of a very convex piece of bone allowing the bone to zigzag apart) of the wall of the anterior sinus allows its compression, training for better control of frontal projection and contour. The exclusion of the sinuses and the obliteration of fat or bones help to minimize the possibility of recurrence.

Usually, when endoscopic sinus surgery is not suitable for correcting a clear frontal valve obstruction, and when a frontal deformity needs to be corrected, open therapy is the first choice of treatment for PD. In our case, no surgery was possible due to the diffuse bone demineralization.

CONCLUSION:

Frontal PD is a rare entity, with an etiology that remains uncertain. It is diagnosed with a standard x-ray or a CT scan. Diagnostic criteria include enlargement of an air cell or the entire sinus and the presence of air only in the walls of the sinus. Different therapeutic options have been proposed to treat the deformity associated with PD while respecting the functional and aesthetic result.

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