Pneumosinus Dilatans: Rare Cause of Slowly Changing Frontal Contours

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Case Report

Pneumosinus Dilatans: Rare Cause of Slowly Changing Frontal Contours

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Abstract
Frontal pneumosinus dilatans is a rare entity that is documented only by a few reports. It is an abnormal expansion of the aerated frontal sinus, with sinus walls of normal thickness. The expansion of the bone may be generalized or focal. It is not only an aesthetic problem but may also have functional consequences. We herein present the case of a 23-year-old woman who had been referred to the maxillofacial and plastic unit in Sousse (Tunisia) complaining of a prominence on her right supraorbital region and forehead. The cause was unknown and there were no functional problems. Computed tomography showed a large pneumatized frontal sinus and confirmed the resulting bony prominence. The patient was taken to the operating room, where the frontal and supraorbital regions were exposed through a bicoronal incision. The anterior wall of the right frontal sinus was removed, and was divided into segments that were then fixed in the desired position using a titanium mesh plate. Excellent results were obtained, and the patient had no complaints and was satisfied with her appearance. She remained well two years postoperatively. Pneumosinus dilatans is an entity every plastic surgeon should be aware of because its treatment falls within the realm of craniofacial surgery. The etiology remains unclear, and surgical management is directed toward surgical exploration of the sinus to ensure recontouring of the anterior table of the frontal sinus to correct any cosmetic deformity. Several authors have published different surgical techniques.

Level of Evidence: 5

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Pneumosinus dilatans (PD) is a rare, usually benign and asymptomatic disease of unknown etiology characterized by expansion of one or more of the paranasal sinuses, with no evidence of bone destruction or pathological changes of the underlying mucosa. The first description of the disease was given by Meyes in 1898, and Benjamins1,2 first assigned the name pneumosinus dilatans in 1918.

We report one new case of frontal PD and discuss the etiology and clinical and radiological features, giving special attention to the treatment.

CASE REPORT
This 23-year-old woman was referred to the maxillofacial and plastic unit in Sahloul Hospital (Sousse, Tunisia), complaining of a prominence on her right supraorbital region and forehead, with a mass that enlarged slowly over the previous seven years. The medical history was unremarkable, and no history of rhinitis, sinus symptoms, or allergies was found.

Clinical examination revealed a fixed, hard, nontender 3 × 2.5 cm swelling in the right supraorbital region. The overlying skin was normal. The left side of the face was normal, and no other signs were present (Figure 1A, C, E).
Figure 1. Preoperative frontal (A), lower (C), and lateral (E) views of this 23-year-old woman with right frontal swelling before craniofacial surgery. Postoperative frontal (B), lower (D), and lateral (F) views demonstrate an improvement in contour 2 years after craniofacial surgery.
The cause was unknown, and there were no functional problems. Laboratory results were within normal limits. Computed tomography (CT) showed a large pneumatized frontal sinus and confirmed the resulting bony prominence with no evidence of bony erosion, thinning of the walls, or mucosal abnormalities (Figure 2).

In view of the cosmetic appearance and the increased size of the swelling on the right side of the face, the patient was operated using a bicoronal approach, exposing the frontal and supraorbital regions. The supraorbital neurovascular bundle was identified and preserved. The anterior wall of the right frontal sinus was removed and divided into segments, which were then contoured and fixed in a new position using titanium mesh plate with sufficient size for a harmonious reconstruction (Figures 3 and 4). Mucosa of the frontal sinus was found to be normal. The ostium was inspected and found to be macroscopically normal. The wound was closed in layers, and sutures were removed on the tenth postoperative day.

Excellent results were obtained, the patient had no complaints and no sinus symptoms, and was satisfied with her appearance (Figure 1B, D, F). She remained well two years postoperatively. After 24 months, no recurrence has been detected.

**DISCUSSION**

**Definition and Terminology**

PD is characterized by idiopathic progressive expansion of one or more paranasal sinuses beyond the normal margins, without evidence of mucous membrane changes. The expansion may involve the complete sinus or a part of it.3

The medical literature offers various labels to describe enlargement of the sinus by air, including frontal sinus hypertrophy, PD, pneumosinus frontalis, aerocele, pneuomocele, sinus ectasia, hyperpneumatization, pneumatocele, air cyst, and others.1,4,5

The varying terminology used to describe abnormal expansion of the frontal sinus has caused some confusion about the etiology and diagnosis of the condition.1 Urken et al4 classified the deformity into three groups—hypersinus, pneuomocele, and PD—as follows:

- Hypersinus or hyperpneumatization was defined as an enlarged frontal sinus that has developed beyond the upper limits of normal. The walls are normal, and the hyperaerated sinus does not extend over the normal...
limits of the frontal bone. The patient is asymptomatic, and the condition requires no intervention.1

- Pneumocele refers to an aerated sinus with variable thinning of the sinus walls. The thinning, focal or generalized, differentiates pneumocele from PD. It is a pathological abnormality.1

- PD is a condition where the sinus abnormally expands beyond the normal limits of the frontal bone. The bony walls of the sinus are of normal thickness, but are displaced, causing frontal bossing. There is no evidence of erosion, and the mucosa is of normal appearance. The frontal sinus is most commonly affected, and the ethmoidal, sphenoidal, or unilateral maxillary sinus may be involved.6,7

Etiology

The etiology of primary PD has been the source of great debate for many years. It is still unknown, but eight possible mechanisms have been proposed as follows: a spontaneously draining mucocele, the presence of a gas-forming microorganism, the presence of a one-way valve, congenital abnormality, hormonal change, local growth disturbances, osteoclastic and osteoblastic activity, and trauma.1,7-9

Generally, frontonasal duct obstruction of any cause and the subsequent increase in sinus pressure seem to be the most important factors in the pathogenesis of PD.1,3,4 In this case, the ostium was inspected and found to be macroscopically normal, and we did not find a clear etiology.
Review of the literature reveals that age at presentation varies from puberty to the elderly, but PD has not been reported in children. This may be due to the age at which the normal paranasal sinus develops, as well as the gradual onset of PD.5,6

Diagnosis
Diagnosis is made by clinical examination, and confirmation by radiography (plain film or CT), when the characteristic enlargement of the sinus is seen.7,8

Clinical symptoms are typically related to the displaced structures. In the case of outward expansion, the typical signs are frontal bossing and prominence of the supraorbital ridge. The expansion may be directed intracranially or toward the orbit, nose, and other sinuses. Related symptoms include sinus pressure, diplopia, ocular alterations, anosmia, and headache.1,4,5,9,10

PD has also been associated with other disorders, such as meningioma, fibro-osseous disease, cerebral hemiatrophy, Klippel-Trenaunay-Weber syndrome, port-wine stains, and arachnoid cysts.1,9-12

CT scanning provides the most expedient differentiation from a host of other causes of frontal swelling, in particular solid mass lesions. Cyst wall thinning may even progress to areas of dehiscence, in which case a pneumatocele is produced.

The differential diagnosis of a frontal mass may include benign and malignant cutaneous or mesenchymal tissue neoplasms, a developmental cysts, expansive inflammatory processes, mucoceles, and fibrous dysplasia.2,13,14

Treatment
The aim of treatment is to restore both form and function to normal, although it is difficult to draw a boundary between them.1

To ensure low risk of intracranial injury, Galiè et al placed a small window inferiorly near the periphery of the sinus in a safe position, and, after they passed a fiber-optic light through the window, the frontal sinus was transilluminated, and the limits of the enlarged sinus were outlined and marked on the bone. The surgery may also be performed using stereotactic instruments.

Different therapeutic options have been proposed for treating the deformity associated with PD,15 and various effective open procedures have been described16 as follows:

- A craniofacial approach with reshaping of the fronto-orbital bossing and repair with bone cement.5 The use of calcium phosphate cement to obliterate the osteotomizing frontal sinus was described in the literature1,9; it has anecdotally led to a high incidence of problems. Utilizing alloplastic material in a potentially contaminated space will very likely elicit problems.1
- Reduction of the anterior wall of the frontal sinus, performed without filling the resultant cavity with bone or hydroxyapatite.15 With this technique, the residual convexity of the bone is not treated, and the cosmetic result is not satisfactory.
- Removal of the anterior wall of the frontal sinus. An appropriate amount of bone is sliced off from this anterior bone plate, and the remainder of the plate is then replaced and wired into position.17 This gives an unstable fixation, while in the frontal area a solid osteosynthesis is needed. The main difference between this and our technique is stability.
- Removal of the anterior wall of the frontal sinus and dividing this into segments, which are then fixed in the desired position using miniplates 18 or a titanium mesh plate, such as in our case. From the original anterior wall of frontal sinus that was removed, we used an appropriate amount of bone equal to the surface of the frontal defect, which allowed maximum coverage of the
bone defect below the titanium mesh. This method, described in this article, is a good option for treatment of PD, and it provides an excellent cosmetic result without additional bone grafts. Because the duct is open, and the sinus mucosa is normal with no sinus pathology, treating the anterior wall of the frontal sinus should be adequate and would simplify the procedure. This would be analogous to treating an anterior wall of a frontal sinus fracture. Solid fixation and harmonious reconstruction were obtained with a titanium mesh plate. The patient had no complaints and was satisfied with her appearance.

- Surgical treatment of PD frontalis is not only directed at correcting the bony deformity but also at reestablishing an appropriate drainage when the nasofrontal duct is involved. In the case presented here, the sinus was not obliterated, and after intraoperative evaluation of the ostium showed a widely patent nasofrontal recess and normal mucosal lining.

CONCLUSIONS

Frontal PD is a rare entity, with an etiology that remains unclear. It is diagnosed with standard radiography; ie, a CT scan. The diagnostic criteria include enlargement of an air cell or of the whole sinus and the presence of only air within the walls of the sinus. Different therapeutic options have been proposed for treating the deformity associated with PD. We treated our patient by removing the anterior wall of the frontal sinus and carried out the reconstruction with titanium mesh plate of a sufficient size for a harmonious and solid reconstruction.

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