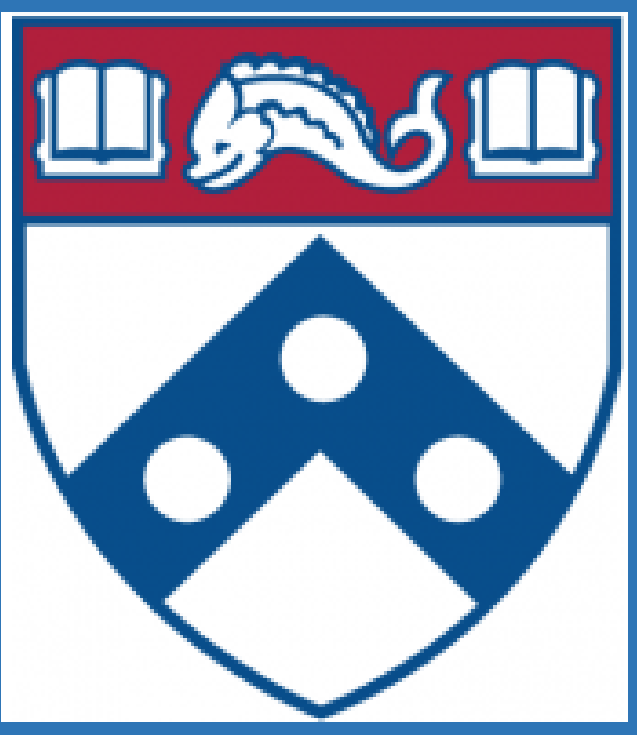


Progression of a Frontal Sinus Pneumocele in a Patient with Recurrent Nasal Polyps

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Abstract

Pneumoceles are a rare condition of abnormal expansion of one or more paranasal sinuses. Its etiology remains unclear although numerous hypothesis have been proposed in the literature. We present the case of a 34 year old male with longstanding history of chronic rhinosinusitis with nasal polyps and an enlarged left frontal sinus. The patient initially underwent endoscopic sinus surgery which improved his sinus and nasal symptoms and he was subsequently lost to follow up. He returned 5 years later with recurrent nasal polyps along with further expansion of his frontal sinus. Although the pathophysiology of pneumoceles remains a mystery, we present a case showing progression of frontal sinus growth despite surgical alterations of patient's sinus outflow tract and discuss the implications on the possible pathophysiology of this condition.

Introduction

Pneumoceles are a rare condition of abnormal expansion of one or more paranasal sinuses. Its etiology remains unclear although several hypothesis have been proposed ranging from gas forming bacteria to a "ball-valve" sinus outflow tract obstruction.(1) We present a case of a patient with a history of nasal polyps and an enlarged frontal sinus. He returned to our clinic several years after treatment of his polyps with recurrent nasal polyps and further expansion of his frontal sinus. To our knowledge, this is the first description in the literature of progression of a pneumocele in the setting of recurrent sinus disease. We describe his case and discuss the implications on the possible pathophysiology of this condition.

Case Report

A 34 year old male presents with history of chronic rhinosinusitis with nasal polyps and a left frontal sinus pneumocele. He initially presented to our clinic in 2011 with a history of prior sinus surgery in 2009 with little symptom improvement. Physical exam and computed tomography(CT) of the sinuses were obtained showing post-surgical changes from prior endoscopic sinus surgery as well as residual polyps and an enlarged left frontal sinus.(Figure 1) He underwent revision endoscopic sinus surgery and medical management with both oral and nasal steroids which improved his symptoms. He was subsequently lost to follow up but returned 5 years later with return of nasal symptoms after cessation of his medications. Physical exam at that time revealed bilateral nasal polyps. Repeat CT and magnetic resonance imaging of his sinuses revealed extensive recurrent nasal polyps as well as further expansion of his left frontal sinus.(Figure 2 and Figure 3) He was taken for revision endoscopic sinus surgery and was placed back on steroids for medical management. His symptoms improved and he continues on medical management at this time.

Discussion

Pneumoceles were first described by Benjamin in 1918 as benign expansions of one or more paranasal sinuses.(1) He coined the term pneumosinus dilatans to describe his findings. Since that time the nomenclature remains mixed with pneumoceles and pneumosinus dilatans being used interchangeably. He proposed three hypothesis: gas forming bacteria, spontaneous drainage of a frontal sinus mucocele, and frontal sinus outflow tract obstruction.(1) No gas forming bacteria have ever been found leaving that hypothesis unproven.(3) There are isolated reports of spontaneously draining mucoceles, however most patients do not describe symptoms of sudden mucocele drainage.(4-6) Increased frontal sinus pressure through a ball valve mechanisms has become a more widely accepted hypothesis with numerous reports showing evidence suggestive of this theory.(7-9) Nevertheless, not all patients have apparent sinus obstruction and up to 25% of pneumosinus dilatans are associated with other pathologies including intracranial lesions such as meningiomas and arachnoid cysts that do not obstruct the sinus outflow of these patients. (2, 10)

Alternative theories have been proposed, the most notable is that pneumosinus dilatans is a disorder of bone growth or metabolism.(2) Jankowski et al used positron emission tomography to show increased sodium fluoride uptake within the bony walls of the enlarged sinuses compared to the surrounding normal sinuses. Bone biopsy from their study also showed intense and diffuse remodeling of bone, with substitution of normal lamellar trabecular bone by osteoid trabeculae.(9) The results suggest that there is significant increase in bone metabolism within the dilated sinus.

Although there is evidence that bone metabolism is affected by pneumosinus dilatans, most patient do not have evidence of a metabolic or bone disorder. We propose to elaborate on Benjamin's original theory that pressure within the sinus triggers the abnormal bony metabolism that leads to abnormal dilation. Rather than being purely related to internal sinus pressure, the abnormal sinus growth may be dependent on a balance of both internal and external pressures acting on the sinus.

Internal pressure from within the sinus, such as with mucoceles and obstructing polyps can lead to activation of bone metabolic activity and enlargement.(5-9) Pressure by external structures such as the brain against the sinus may serve as a counter balance to signal to the sinus to arrest growth. Examples of this can be seen in congenital deformities of brain development and decreases in intracranial pressure by ventriculoperitoneal shunting which have been associated with pneumosinus dilatans.(11, 12) Parizel et al in their study of skull base meningiomas associated with pneumoceles noted that the dilation of the sinus only occurred along the wall adjacent to the tumor. He suggested that there may be local pressure/traction effects of the meningioma on the dura adjacent to the sinus leading to expansion.(10)

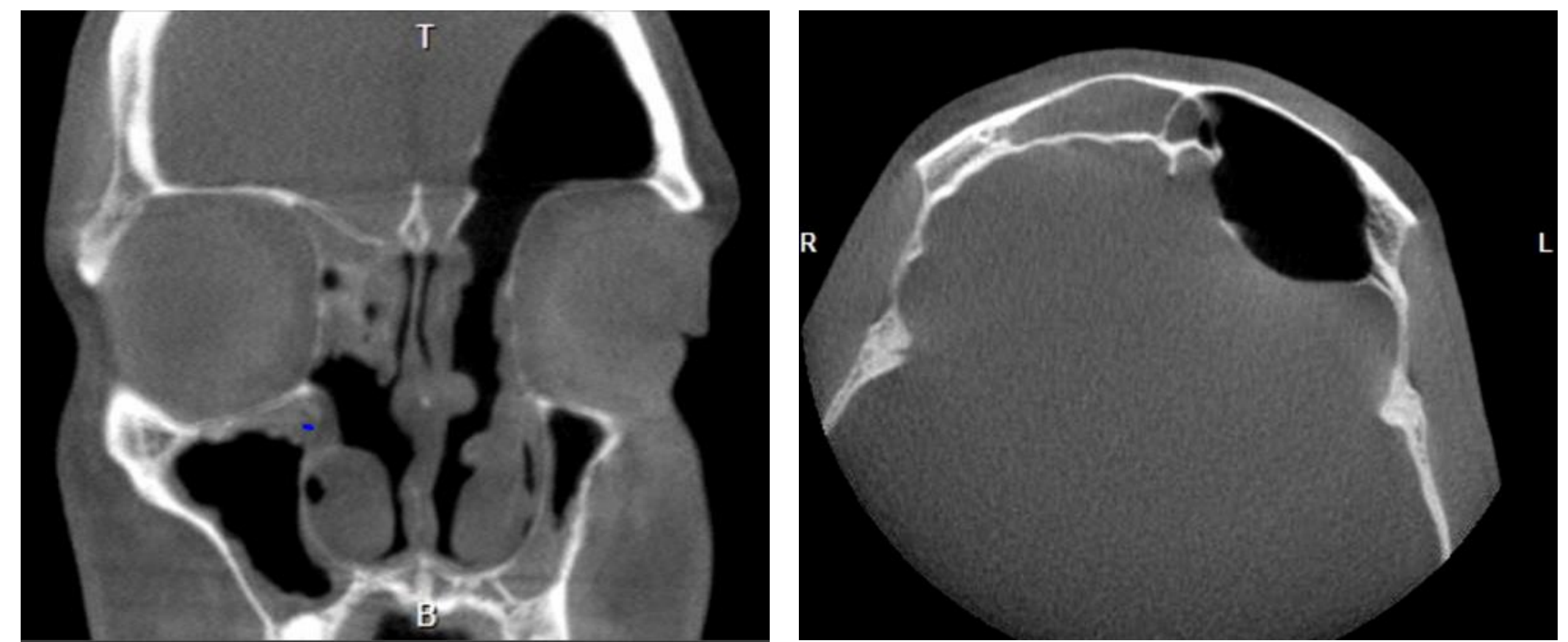


Figure 1: Coronal and axial computed tomography images from patient's initial presentation showing nasal polyps and an enlarged left frontal sinus.

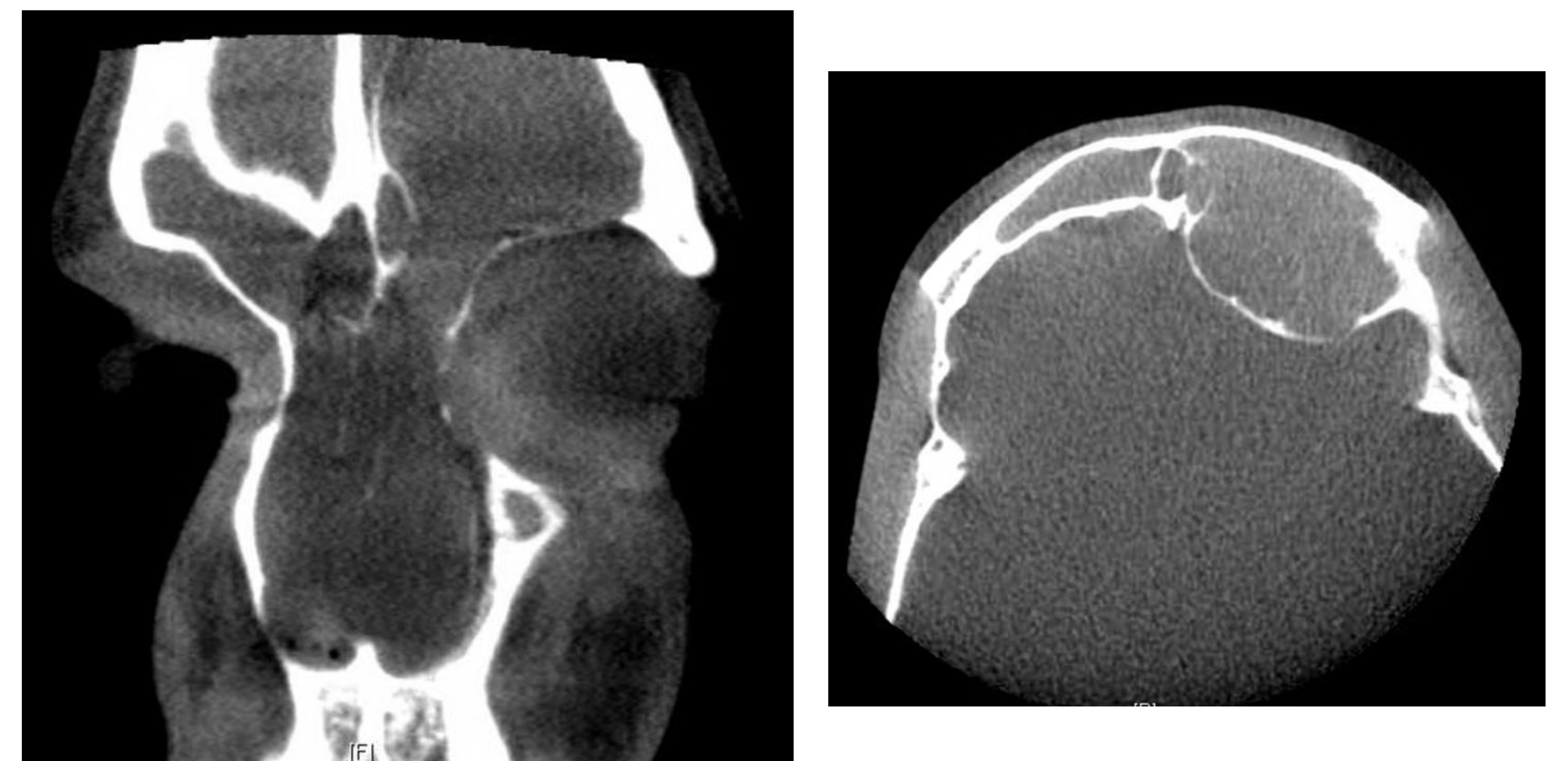


Figure 2: Coronal and axial computed tomography images from patient's follow up 5 years later presentation showing recurrent nasal polyps and an enlarged left frontal sinus.

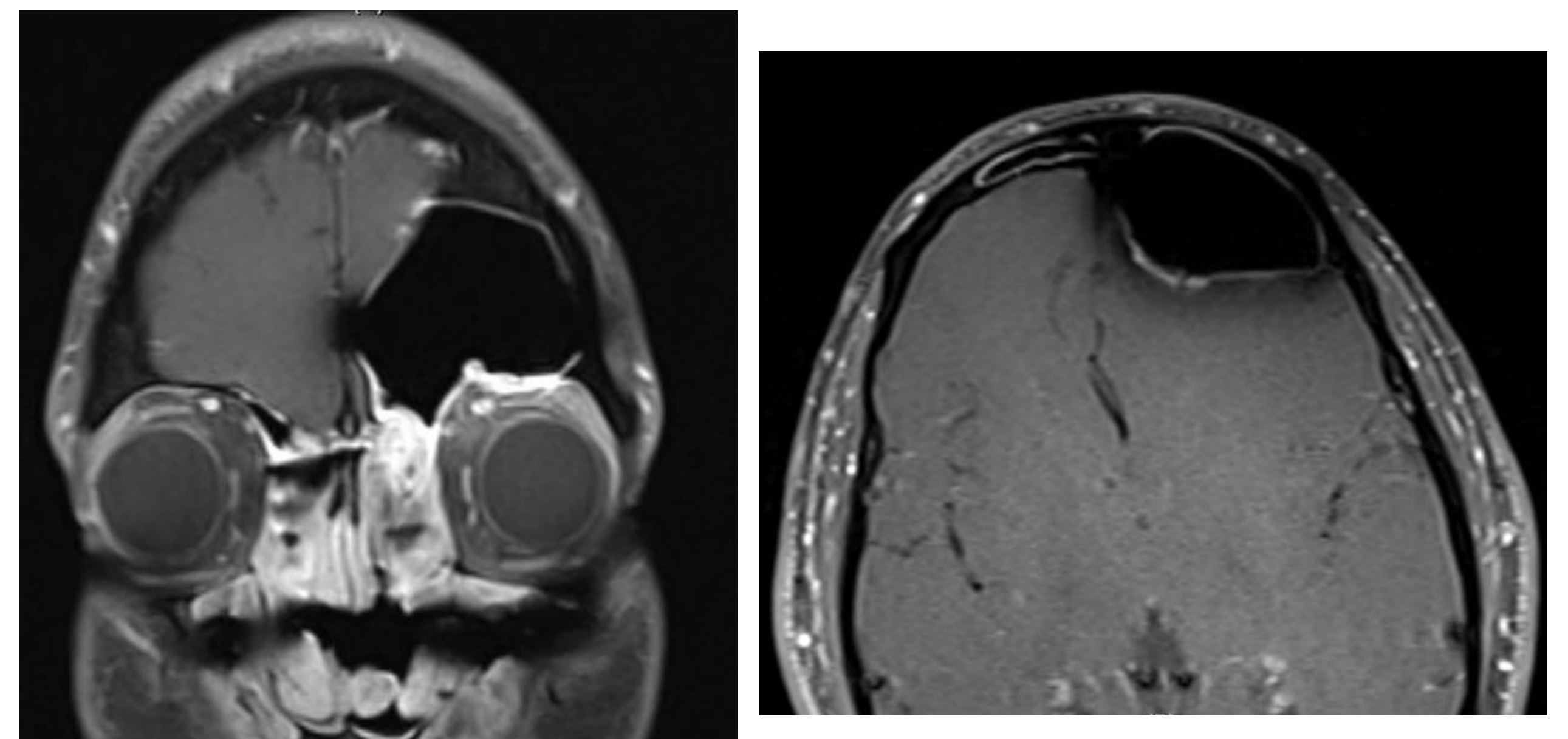


Figure 3: Coronal and axial T1 post contrast magnetic resonance imaging showing residual polyps obstructing the frontal sinus outflow tract with a further enlarged left frontal sinus when compared to five years earlier.

Conclusion

Given the current literature, we conclude that pneumosinus dilatans occurs due to abnormal bony remodeling that is triggered by disequilibrium in the pressure/traction forces placed on the sinuses. Increased pressure in the sinus; either through increased pressures from ball valve obstruction, expansion of mucoceles, or other etiologies can lead to further expansion. Furthermore, negative pressure from external sources, such as traction by meningiomas or absence of counter pressure from the brain by an arachnoid cyst can also prompt the same bony growth leading to hyperpneumatization. Our patient with a history of persistent nasal polyps likely experienced episodes of ball valve obstruction due to his recurrent nasal polyps leading to increased pressure and continued expansion of his frontal sinus.

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