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Nasal Speech Associated with Hyperaeration of the Sphenoid Sinus

MEIKE FRAEDERICH[1*], MOHSEN FAWZY ABOELHASAN[2*], JILL KNIPS[1], MAX HEILAND[1] and REINHARD E. FRIEDRICH[1]

[1]*Department of Oral and Maxillofacial Surgery, University Medical Center of Hamburg-Eppendorf, University of Hamburg, Hamburg, Germany;*
[2]*Saudi German Hospital Group KSA, Faculty of Dentistry, Al-Azhar University, Cairo, Egypt*

Abstract. *The sinus sphenoidalis is the paranasal sinus most centrally located in the skull base and known to vary extremely in dimensions. A very large volume of the sinus visible on skull radiographs is called sinus 'hyperaeration'. This finding has been described many times in the literature, but is generally rare. The term 'pneumosinus dilatans' is synonymously used to address an unexpectedly large paranasal sinus. In the majority of cases, sphenoid sinus hyperaeration is not associated with any clinical symptoms. On the other hand, different complaints have been described in cases of extensive sphenoid sinus volume, such as headache, inflammation or neoplasia. This is a report of an adult patient with nasal speech who was thoroughly investigated for alterations of the upper airway in order to clarify the reason for the speech anomaly. After extensive diagnostics, the patient was found to have normal anatomy and no functional alteration of the velo-pharyngeal complex. However, an impressively large sphenoid sinus was shown on computed tomograms. This is likely the first report detailing a patient with nasal speech and hyperaeration of the sinus.*

The sphenoid sinus is a normally present anatomical entity situated inside the sphenoid bone (1). This sinus is the most variable in extension and shape of all paranasal sinuses (2-4). Aplasia of this sinus is rare (5-7). Aeration of the sphenoid bone by the sinus is usually classified with reference to the *sella turcica* in an anterior-posterior direction (3, 7, 8). With reference to the medio-sagittal plane, the development of the sphenoid sinus directed to both lateral sides is independent of each other (9). Therefore, the sides of the normal sinus may differ markedly in symmetry using the medio-sagittal plane as the spatial reference level (1, 9). Furthermore, additional isolated air cells independent of the sphenoid sinus may develop in the sphenoid bone (10). The sinus does not normally extend beyond the spheno-occipital sutures (8). However, the sinus can extend into the adjacent bones, *e.g.* frontal or occipital bone, vomer, palatine or ethmoid (1-4). These extensions do not necessarily dissolve the osseous borders of the sinus (9). However, reports have detailed palpable fluctuations of the scalp in cases with extensive *pneumosinus dilatans* extending below the temporal soft tissues (11). On the other hand, a continuous anterior-posterior passage between the sphenoid sinus and another sinus was shown on cross-sectional images without loss of the cranial, dorsal and lateral osseous borders of the unified cavities (12).

This wide range of findings indicates the high variability of sinus morphology based on a floor plan of skull base pneumatisation (2). These differences in spatial expansion of the sphenoid sinus can have important surgical implications (9, 13, 14). Not all sphenoid sinus anomalies necessarily accompany a distinct pathology (15, 16). However, the so-called *pneumosinus dilatans* can cause severe health impairment of the affected individual, *e.g.* headache (15, 16), inflammation (16), blindness (17-19), or may even indicate a neoplasm of the region (20, 21). In the latter situation, the *pneumosinus dilatans* can be a consequence of slowly growing tumors causing a blockade of the physiological air flow of the sinus. Differential diagnosis to or identification of co-existence of other space occupying lesions is mandatory and unavoidable is the need for adequate imaging (22).

Sphenoid sinus development and volume. The pre- and postnatal development of the sphenoid sinus has been well described (2-4, 23-29). At the time of birth, the sphenoid sinus is only a blind sack bulging from the nasal cavity which does not reach the sphenoid cartilage or bone (2). Based on a

*These Authors contributed equally to this article.

Correspondence to: Professor Reinhard E. Friedrich, MD, DMD, Ph.D., FEBOMFS, Department of Oral and Maxillofacial Surgery, University Medical Center of Hamburg-Eppendorf, Martinistraße 52, 20246 Hamburg, Germany.Tel: +49 40741053259, e-mail: rfriedrich@uke.de

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cadaver study, Vidic described the sinus extending into the pre-sellar region before the fourth year of age (23). Further penetration into the body of the sphenoid bone can be found frequently in later years. The sinus reaches its maximum extension at an age of 13 to 20 years, with at least partial pneumatisation of the *sella* and sometimes even total pneumatisation of the *dorsum sellae*. Krmpotic-Nemanic *et al*. also stated the sphenoid sinus to have reached its final shape and dimensions around the age of 12 to 14 years, based on linear measurements of the anterior borders of the sphenoid sinus on an axial computed-tomographic (CT) scan study of the skull base (27). However, a more recent study of Yonetsu *et al*. provided evidence for aeration of the sphenoid bone not reaching its maximum extension until the third decade of life (8). They determined the sphenoid sinus volume in 214 patients and even developed a formula to estimate the volume depending on the patient's age. These authors measured sphenoid sinus volumes ranging from about 4.0 to 10.0 cm^3 in the 30- to 39-year-old age group. They were probably the first to describe the reduction of the sphenoid sinus volume after an age of about 50 years. They also addressed the finding that the sinus may only have two-thirds of its maximum volume at ages above 70 years. Data on gender-specific differences of sphenoid sinus volume are inconclusive, probably due to the large variations of size and study design (Table I). Elwany *et al*. described the average size of the sphenoid sinus between 2.5 ml in pre-sellar to 8.5 ml in post-sellar extended sinuses (30). They also compiled data of previous studies on sphenoid sinus volume. According to these authors' analysis of the literature, the average capacity of the sphenoid sinus in adults was estimated to be about 6 to 7.5 ml.

Hyperaeration of the sphenoid sinus was described by several authors (16) and is also known for other paranasal sinuses (16, 31-33). In most cases, this finding, *e.g*. the pneumatisation of the pterygoid process of the sphenoid bone, was asymptomatic and incidental due to routine radiographic diagnostics in single cases [orthopantomogram (10, 34)] or the report was found noteworthy in anatomical or radiographic studies examining the sphenoid sinus (25, 35). Hyperaeration of a sinus is defined by both the volume (16), where no limit is known, and the pneumatisation of skull bones in continuity to the sinus that are normally not aerated (16) or the confluence of sinuses to a large cavity (12). No impact on speech quality of a large sphenoid sinus is evident from the literature (16). This report presents a large sphenoid sinus with obvious consequences on speech quality.

Case Report

History and physical findings. A 32-year-old female presented at the Outpatient Clinic of the Department of Oral and Maxillofacial Surgery at the Saudi German Hospital Aseer, Saudi Arabia. The patient complained of nasal speech. She reported having suffered from nasality ever since childhood without improvement or response to medical treatment. She had no history of physical trauma or surgical procedures in the head and neck region. She described having had logopaedic speech therapy for her nasality, without any effect. Otherwise the physical examination was without any pathological findings. In particular, there was no velopharyngeal insufficiency or (submucosal) cleft. The patient had no clinical signs of inflammation. For diagnostic clarification a computed tomographic scan of the paranasal sinuses was performed.

Computed tomography. In the CT images, the naso-pharyngeal anatomy appeared to be normally developed, except for a marked hyperaeration of the sphenoidal sinus. The sinus bordered on the following structures (Figure 1): occipital bone, left anterior clinoid process and lesser wing, left greater wing, and pterygoid process on both sides. There was only discrete mucosal swelling at the base of both maxillary sinuses, otherwise there was an undisturbed aeration of all sinuses.

Volumetric analysis of the sinus was performed with OsiriX®-DICOM-Viewer open-source software (OsiriX® Imaging Software; Pixmeo, Geneva, Switzerland). The volume of the sphenoid sinus was measured as 15.55 cm^3. The three-dimensional imaging of the sphenoid sinus as shown in Figure 1 C-E was performed with BrainLab® (Brainlab AG, Feldkirchen, Germany).

Discussion

This is a report describing a *pneumosinus dilatans* of the sphenoid sinus associated with a permanent hypernasality of speech in an otherwise healthy and speech-competent individual. Hyperaeration of the sphenoid sinus has been described by several authors. In most cases, this finding was asymptomatic and incidentally depicted due to routine radiographic diagnostics of the skull (10), or in anatomic or radiographic studies of the sphenoid sinus (16).

Terminology. In recent literature on the health implications of enlarged paranasal sinuses, there is a trend identified for the use of the the term 'hyperaeration' in the radiological diagnosis of a very large sinus without any complaints of the individual related to this region (16). On the other hand, the term '*pneumosinus dilatans*' designates an enlarged paranasal sinus with health-related problems in a wide sense, varying from aesthetic disfigurement, *e.g*. extensive frontal bossing of frontal sinus with thinning of the cortical bone (16), to infection (36) or even skull base tumor, *e.g*. sphenoid sinus expansion associated with meningioma (20, 21). This distinction to reserve hyperaeration for defining a large sinus extending beyond the accepted osseous borders but without

Table I. *A selection of studies dedicated to measurements of the sphenoid sinus volume (SSV).*

Authors, year (reference no.)	No.	Age range (years)	Country	Mean±SD SSV (cm^3 or ml)		Min-max SSV (cm^3 or ml)	Imaging technique	Remarks
				Female	Male			
Elwany *et al*., 1983 (30)	100/100/50	20-50	Egypt	2.2±7.9	2.7±9.1	n.d.	Plain radiographs, manual measurements	Healthy subjects (n=100) and cadaver (n=100/50) studies, remains not further specified
Kawarai *et al*., 1999 (43)	20	21-36	Japan	13.7±6.2	17.1±7.4	Female: 4.9-25.1, male: 7.8-19.4	CT	Healthy volunteers
Yonetsu *et al*., 2000 (8)[a]	214	30-39	Japan	~4.0-11.0		Female: ~4-10, male: ~5-11	CT	Age-related volume measurements (decades), noticed expanding volume up to third decade of life
Sareen *et al*., 2005 (45)	22	n.d.	India	5.8		3.0-10.0	Manual measurements	Human cadaver study, remains not further specified
Karakas and Kavakli, 2005 (44)	91	5-55	Turkey	6.00±3.02	6.83±3.73		CT	No sexual dimorphism noted
Emirzeoglu *et al*. 2007 (49)	77	18-72	Turkey	13.6±0.7		Female: 0.3-20.3, male: 1.5-18.6	CT	No sexual dimorphism noted
Oliveira *et al*., 2007 (46)	50	n. d.	USA	10.88±3.83	15.40±5.59	Female: 3.30-20.28, male: 4.30-24.25	CT	Sexual dimorphism noted
Park *et al*., 2010 (50)[b]	260	0-25	Korea	2×3.418±1.263		n.d.	CT	Age-related volume measurements (years), no sexual dimorphism of volume noted, adult dimensions reached by 15 years
Kim *et al*., 2010 (51)	60	18-63	Korea	10.671	14.708	n.d.	CT	Sexual dimorphism noted, individual sphenoid sinus volume correlated to volume of mastoid cells
Lee *et al*., 2012 (47)[c]	62	<5-18	Korea	About 16[c]		About 5-19	CT	Age-related volume measurements (5 years group), adult dimensions reached by >20 years
Amusa *et al*., 2013 (48)	24	Adult	Nigeria	5.38±5.64 (right) and 4.23±1.99 (left)		n.d.	Water displacement measurements	Human cadaver study on dry skulls, remains not further specified

[a]Volumes refer to age group 30-39 years (n=17); [b]mean SSV (each side) >3.0 cm^3 in all groups per year (n=10 each) older than 9 years of age (number of investigated individuals and correlating volume for oldest age group); [c]volumes are derived from graphic representation of linear regression model; n.d.: not detailed.

any pathology on the one hand, and on the other hand, to restrict the term '*pneumosinus dilatans*' for a similar radiographic picture, but with evidence of clinical findings and symptoms, is convincing at first glance only. In some cases, *e.g.* excessive aeration of the frontal sinus, the indication for surgery may be solely for aesthetic reasons. Therefore, this distinction appears to be somewhat arbitrarily made. Indeed, a recent review on the literature addresses the different pathologies associated with an enlarged paranasal sinus under the umbrella term '*pneumosinus dilatans*' in order to sensitize the reader to the need to carefully investigate any enlarged paranasal sinus for associated pathologies (16). These authors avoid the term 'hyperaerated' sinus. The usage of *pneumosinus dilatans* in this wide sense was proposed by Reicher *et al*. (37), but the introduction of the term can be traced back to reports published at the beginning of the last century (38). These authors had already suggested to refer to '*pneumosinus dilatans*' as a case of enlarged paranasal sinus without obvious pathology (37).

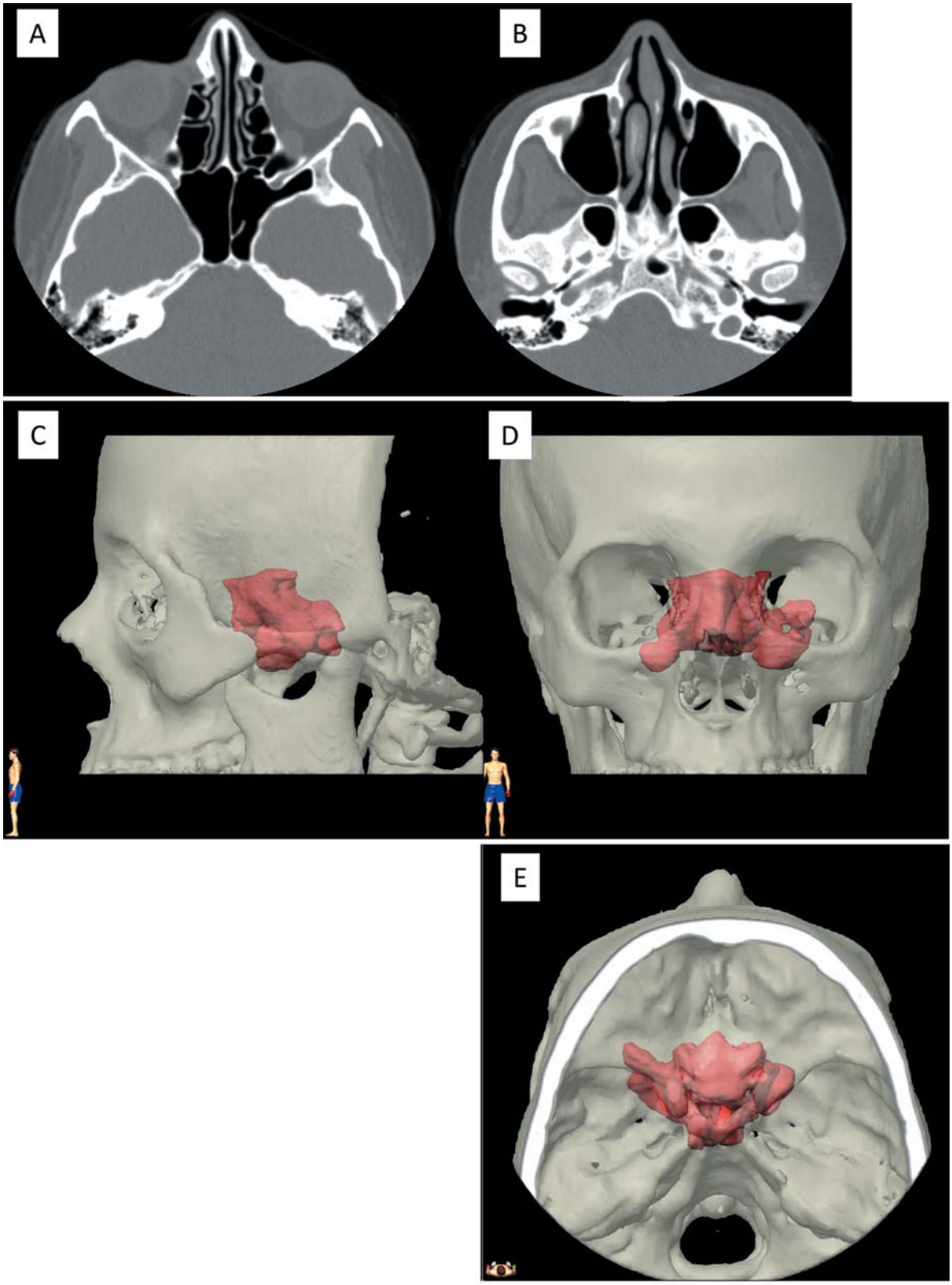

Figure 1. *Selected images of enlarged sphenoid sinus derived from computed tomographic (CT) scans. The axial CT slices show aeration of the left greater wing of the sphenoid bone (A) and the pterygoid process on both sides and clivus (B). Volume reconstruction with BrainLab®-software: lateral view (C), anterior view (D) and cranial view (E).*

Indeed, the hazards of a large sinus to the affected individual do not necessarily arise from the anatomy of the paranasal sinus alone, *e.g.* giving rise to ascending infections, but also from planned surgical interventions in this region, in particular in the case of performing a procedure based on inadequate imaging (39). Furthermore, it is not clear whether such an enlarged sinus will retain a constant volume over time (8, 40). Therefore, both terms appear to address the same phenomenon.

Radiological characteristics and associated pathologies. Morton published reported the case of a 16-year-old school boy who presented with swelling of the left temporal region and slight proptosis of the left eye with undisturbed eye movements (11). In this case, excessive sphenoid pneumatisation with a bony defect of the greater wing of the left side was found and the *sinus mucosa* had ballooned into the temporal fossa and the left orbit. This case report is a rare exception because the physical appearance of the individual is usually not affected by an expanding sphenoid sinus. However, there are no standards reported when diagnosing a thinned cortical border of a paranasal sinus on cross-sectional images. Anatomical data for the wall thickness of the sphenoid sinus are derived from cadaver studies (2). The definition of cortical thinning, erosion and expansion strongly rely on the resolution of the applied imaging technique (9).

Thinning of the sphenoid sinus was most frequently found to occur in the wall adjacent to the groove for the internal carotid artery (9). Expansion of the sphenoid sinus can also affect the sinus floor (9). However, bone remodelling in the enlarged sphenoid sinus is not an unavoidable consequence of a sinus enlargement (15). Due to hyperaeration of the sphenoid, vital structures such as the internal carotid artery or the optic nerve can bulge into or even run through the sinus. These anatomical findings may be important for endoscopic surgery (18). Furthermore, an enlarged sphenoid sinus with optic nerves running through the cavity can be associated with loss of vision, regardless of intervention (17, 18) or pathology (19).

Nasal speech and alteration of the paranasal sinus volume and geometry. Our search of the literature offered no case of patients complaining about nasal speech associated with hyperaeration of the sphenoid sinus. Furthermore, a recent review on the impact of *pneumosinus dilatans* of all paranasal sinuses on health, revealed no case causing nasal obstruction associated with *pneumosinus dilatans* of the sphenoid sinus (16). Therefore, the presumed association of the extensive aeration of the sphenoid bone and the obvious speech anomaly in the present case should be concluded with caution. However, excessive pneumatisation of paranasal sinuses can be associated with physical impairment, *e.g.* chronic headache (41). The variability of sphenoid sinus volume has been reported in several studies (42-49) based on different imaging techniques and measuring bases (Table I). In certain studies, extreme values of sphenoid sinus volume exceeded that of the present case (Table I).

The paranasal sinuses play an important role in speech performance (52, 53). Functional magnetic resonance imaging studies suggested a substantial effect of the paranasal sinuses on the nasality of speaking (52). Functional endoscopic surgery of the paranasal sinuses should respect possible changes in nasalance (54) and even conventional paranasal sinus surgery can cause measurable alterations of speech (55). Indeed, from a maxillofacial surgical point of view, the impact of the paranasal sinuses on speech is evident. In particular, consecutive alterations of speaking after extensive orthognathic procedures always need to be considered, predominantly in patients with developmental deficiencies of the midface and adjacent skull base (56, 57). Indeed, it was clearly demonstrated that sinus or orthognathic surgery with consequent change of sinus anatomy can lead to changes in speech. We, therefore, suggest that even simple growth of sinuses can lead to nasal speech (55, 57).

The impact of surgery on speech is discussed in detail in the fields of craniomaxillofacial surgery and phoniatrics (53, 58). LeFort-I osteotomy can cause unpredictable effects on the velopharyngeal complex in patients with complex craniofacial deformities (59). However, in orthognathic surgery, the impact of bone movement on the velopharyngeal competence is a crucial factor in treatment planning and performance. Therefore, surgically-induced alteration of speech in patients with subsequently modified shape and volume of the paranasal sinuses, in particular the maxillary sinus, does not truly mimic the resonating space provided by sphenoid sinus hyperaeration in an intact velopharyngeal complex. In fact, Hassan *et al.* were unable to find clear evidence directly relating the effect of orthognathic surgery to speech alterations (60). Alterations of nasalance showed no impact on speech reception (59).

Conclusion

Hyperaeration of the sphenoid sinus is easily diagnosed on CT scans and in many cases is an incidental finding without clinical symptoms. In the present case, we show how hyperaeration of the sphenoid sinus (*pneumosinuis dilatans*) can be associated with clinically remarkable alteration of speech, *i.e.* a hypernasality. We, therefore, recommend radiographic 3D imaging of the sinuses in cases of otherwise unexplained nasal speech.

Consent

The Authors0 declare that the patient gave their written informed consent for this case report to be published.

Competing Interests

The Authors declare that they have no competing interests in regard to this report.

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