

An extensive maxillary pneumosinus dilatans*

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SUMMARY

Pneumosinus dilatans is a very rare lesion. An aggressive case of a maxillary pneumosinus dilatans is presented. CT and MRI showed that the lesion involved the orbit, cheek and nasal cavity. A reopening to the affected maxillary sinus as a FESS procedure was performed with success.

Key words: pneumosinus dilatans, CT, MRI, FESS

INTRODUCTION

Pneumosinus dilatans was first defined by C.E. Benjamins (1918), as an abnormal enlargement of the paranasal sinuses containing air only. The sinus wall with its mucosa is intact, completely surrounding the lesion. Pneumosinus dilatans can involve all paranasal sinuses. The frontal sinus is most commonly affected, followed by the ethmoids and the sphenoid sinus (Wolfensberger, 1984). In some cases of pneumosinus dilatans in the sphenoid sinus, an association with optic nerve sheath meningiomas has been reported and it has been suggested that sphenoid pneumosinus dilatans may be a radiological sign of an occult optic nerve sheath meningioma (Miller et al., 1996). Pneumosinus dilatans in the maxillary sinus is very rare. Until today, 11 cases of maxillary pneumosinus dilatans have been reported (Breidahl et al., 1997; Komori & Sugisaki, 1988; Mauri et al., 2000; Meyers & Burtschi, 1980; Morrison et al., 1976; Noyek & Zizmor, 1974; Tovi et al., 1991; Vines et al., 1976; Wolfensberger, 1984; Zizmor et al., 1975). Most of these cases have confusingly been named pneumocele, but are actually pneumosinus dilatans according to Benjamin's definition (Breidahl et al., 1997). Thus pneumosinus dilatans differs from pneumocele, which is defined as a pocket of air in a soft tissue envelope, extending beyond the boundaries of the sinus with a defect in the sinus wall through which sinus mucosa herniates. The cause of pneumosinus dilatans remains unclear. We want to present a case of a 40-year-old male with an extensive pneumosinus dilatans in the maxillary sinus. Functional endoscopic sinus surgery (FESS) was performed with success.

CASE

A 40-year-old man was referred to an ophthalmologist because of a right-sided exophthalmus. The patient had observed a facial asymmetry and displacement of his right eye during several

months. The symptoms worsened by nose blowing. Four years prior to referral, the patient had noticed acute onset of temporal pain and paraesthesia in his right cheek and in his right eye during a long-distance flight. The patient was previously healthy and had never received nasal or sinus surgery. The ophthalmologic examination confirmed the right-sided exophthalmus and swelling of the right cheek. Our examination comprising rhinoscopy showed that the nasal wall including the middle turbinate was displaced medially towards the nasal septum obliterating the middle meatus. Compression of the zygoma-maxillary complex revealed a sound like the one produced when you press the bottom of a tin. CT and MRI scan were performed demonstrating abnormal enlargement of the right maxillary sinus involving the nasal cavity, the sphenoid sinus and the orbit (Figures 1 and 2). A FESS operation was indicated. The medial maxillary wall above the inferior turbinate was removed and connected to the area of the natural ostium, under general anaesthesia. When the right orbit was manipulated from the outside, a distinct mobility of the floor of the orbit was observed. Histology showed that the lining mucosa was oedematous and with a slight metaplasia. At the follow-up examination 9 months postoperatively the patient had no symptoms and there was no sign of exophthalmus or facial swelling. The CT scan showed decreasing volume of the right maxillary sinus (Figure 3).

DISCUSSION

The cause of pneumosinus dilatans has been discussed by several authors (Breidahl et al., 1997; Komori & Sugisaki, 1988; Mauri et al., 2000; Tovi et al., 1991). The cheek valve theory posits that repeated air trapping can produce chronic distension of the sinus as a result of positive intrasinus pressure. Repeated air trapping can be produced by sneezing or auto

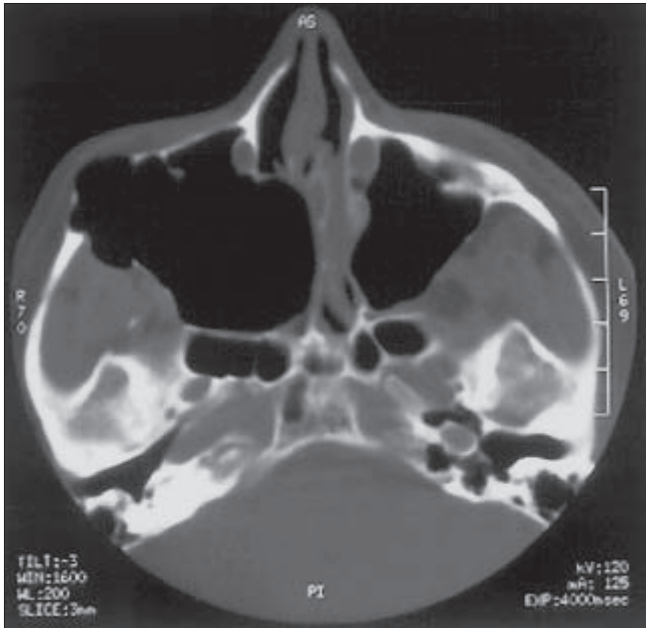


Figure 1. Preoperative axial CT scan demonstrating abnormal dilatation of the right maxillary sinus bulging into the nasal cavity and the sphenoid sinus. Note thinning of the anterior and medial wall of the maxillary sinus. The volume of the maxillary pneumosinus dilatans was measured to be approximately 47 ml. The corresponding volume of the left maxillary sinus was 19 ml.

inflation. This theory has been tested by Wolfensberger (1984), who measured the antral pressure before and after Valsalva's manoeuvre on a patient with right-sided maxillary pneumosinus dilatans. On the left side, the antral pressure returned to normal on completion of the manoeuvre. On the affected side



Figure 2. Preoperative coronal MR scan (STIR sequence) demonstrating the enlarged right maxillary sinus with expansion both into the orbit close to the optic nerve (arrow) and into the alveolar process.

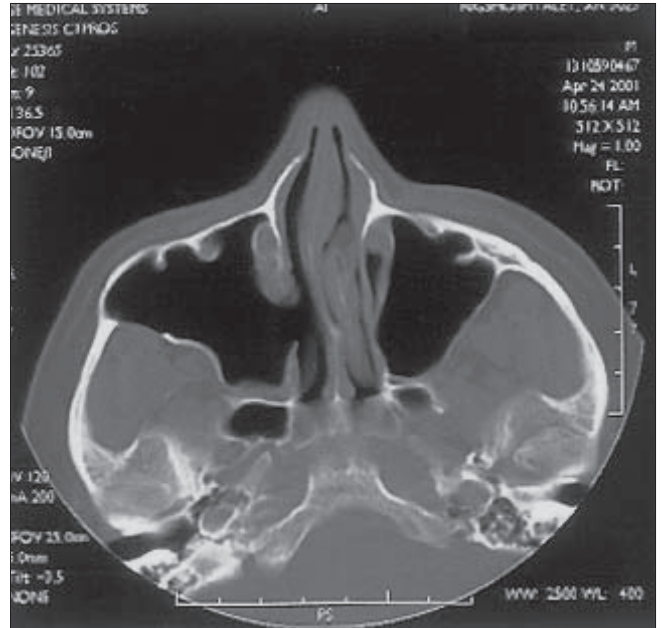


Figure 3. A CT scan 9 months postoperatively showed decreasing volume (29 ml) of the right maxillary sinus. Note remodelling of the anterior wall.

the antral pressure was 100 mm of water after one, and 350 mm after two Valsalva's manoeuvres. The increased pressure persisted for several minutes. Subsequently endoscopy of both maxillary ostiums was performed and on the right side no ostium could be found. A review of clinical data from the 11 published cases of maxillary pneumosinus dilatans shows that all patients were between 12 and 62 years old. There was no gender difference. None of the patients had a history of chronic sinusitis or previous sinus surgery. As in our case, the patients had presented with one or several of the following symptoms: unilateral pain and swelling of the cheek, exophthalmus and nasal obstruction which were worsened by nose blowing or sneezing. Clinical examination showed exophthalmus and a prominence of the zygoma-maxillary complex. Unilateral nasal stenosis caused by the displacement of the medial maxillary wall towards the nasal septum is common. The Caldwell-Luc operation has typically been preferred (Meyers & Burtschi, 1980; Noyek & Zizmor, 1974; Tovi et al., 1991; Vines et al., 1976; Zizmor et al., 1975) and there have been no reports of recurrence after surgery. We recommend CT and MRI for preoperative diagnosis as the findings in case of pneumosinus dilatans are very typical (Figures 1 and 2). CT and MRI can exclude meningioma and map the lesion. The treatment consists of a surgical opening to the affected sinus. The Caldwell-Luc procedure has hitherto been preferred for maxillary pneumosinus dilatans.

Application of the less invasive endoscopic surgery, FESS, was in our case obvious and effective. Thus we suggest FESS as a treatment of pneumosinus dilatans irrespective of the localisation of the lesion in the paranasal sinuses.

REFERENCES

- 1 Benjamins CE (1918) Pneumo-sinus frontalis dilatans. *Acta Laryngol* 1: 412-422.
- 2 Breidahl AF, Szwajkun P, Chen YR (1997) Pneumosinus dilatans of the maxillary sinus: a report of two cases. *Br J Plast Surg* 50 1: 33-39.
- 3 Komori E, Sugisaki M (1988) Ectopic pneumosinus maxillaris dilatans. A case report. *J Craniomaxillofac Surg* 16 5: 240-242.
- 4 Mauri M, de Oliveira CO, Franche G (2000) Pneumosinus dilatans of the maxillary sinus. Case report. *Ann Otol Rhinol Laryngol* 109: 278-280.
- 5 Meyers AD, Burtschi T (1980) Pneumocele of the maxillary sinus. *J Otolaryngol* 9: 361-363.
- 6 Miller NR, Golnik KC, Zeidman SM, North RB (1996) Pneumosinus dilatans: a sign of intracranial meningioma. *Surg Neurol* 46 5: 471-474.
- 7 Morrison MD, Tchang SP, Maber BR (1976) Pneumocele of the maxillary sinus. Report of a case. *Arch Otolaryngol* 102: 306-307.
- 8 Noyek AM, Zizmor J (1974) Pneumocele of the maxillary sinus. *Arch Otolaryngol* 100 2: 155-156.
- 9 Tovi F, Gatot A, Fliss DM (1991) Air cyst of the maxillary sinus (pneumosinus dilatans, pneumocele). *J Laryngol Otol* 105: 673-675.
- 10 Vines FS, Bonstelle CT, Floyd HL (1976) Proptosis secondary to pneumocele of the maxillary sinus. *Neuroradiology* 11: 57-59.
- 11 Wolfensberger M (1984) Pathogenesis of pneumosinus maxillaris dilatans. *HNO* 32: 518-520.
- 12 Zizmor J, Bryce M, Schaffer SL, Noyek AM (1975) Pneumocele of the maxillary sinus. A second case report. *Arch Otolaryngol* 101: 387-388.

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ANNOUNCEMENT

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