European Annals of Otorhinolaryngology, Head and Neck diseases 136 (2019) 43–46

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EM|consulte
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Case report

Maxillary sinus pain with radiolucent sinuses due to agenesis of the membranous ostium

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ARTICLE INFO

Keywords:
Sinusitis
Sinus pain
Diagnosis
CT scan
Middle meatus antrostomy

ABSTRACT

Introduction: The absence of opacities on CT scan usually eliminates paranasal sinus disease as a cause of facial pain. The authors report a case, which constitutes an exception to this general rule, corresponding to a new aetiology of sinus pain.
Case report: A 16-year-old boy presented with very painful "recurrent acute sinusitis" triggered by pressure changes (altitude, diving, surfing), with no sinus opacity on CT scan. Surgical exploration demonstrated absence of a primary or accessory maxillary ostium. Middle meatus antrostomy relieved the patient's pain.
Discussion: The pathophysiology of this case of recurrent acute pseudo-sinusitis and the efficacy of antrostomy can be explained by the evo-devo theory of the origin and function of the paranasal sinuses. This case illustrates the absence of communication in the ethmoid of the membranous sac lining the maxillary sinus, formed by degeneration of the maxillary erythropoietic bone marrow. Under stable environmental conditions, the continuous production of nitric oxide by the sinus epithelium is eliminated by simple transmembrane diffusion, but is insufficiently eliminated in the case of rapid pressure changes, inducing sometimes very severe sinus pain, mimicking sinusitis. This case report paves the way for more detailed studies on the role of the paranasal sinuses in facial disease and respiratory physiology.

1. Introduction

The paranasal sinuses, discovered by the Renaissance anatomists, were only able to be explored clinically with the advent of X-rays, discovered by Roentgen at the end of the 19th century [1]. Since the end of the 19th century, the diagnosis of sinusitis is based on changes of sinus radiolucency.

The purpose of this article is to more clearly define the place of CT in the radiological diagnosis of sinusitis. We report the case of a boy with recurrent acute sinus pain, in whom the CT scan was initially interpreted as being normal in the absence of any paranasal sinus opacity. This sinus pain appeared to be due to constitutional absence of perforation of the maxillary sinus membrane in the ethmoidal middle meatus, and was cured by middle meatus antrostomy that restored elimination of the nitric oxide naturally produced by the maxillary mucosa.

2. Case report

A 16-year-old boy consulted following an episode of very painful "acute sinusitis", occurring several weeks previously during a skiing holiday. As soon as he reached an altitude of 1800 metres, he experienced very severe pain in the left lateral incisor, associated with a feeling of "mobile tooth that appeared to be pushed anteriorly". Over several hours, the pain spread to involve all of the left infraorbital region, prevented him from sleeping on the first night, and confined him to bed for three days despite antibiotic therapy started on the day following his arrival. The pain slowly improved from the fourth day onwards and had completely resolved after his return home three days later. He no longer remembered whether or not he had experienced any associated nasal symptoms, but he did not report any purulent nasal discharge and the tooth, examined by his dentist after his return home, was healthy.

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https://doi.org/10.1016/j.anorl.2018.10.001
1879-7296/

He had experienced a similar but less painful episode several months previously, following diving with breath holding during a rescue-training course, which led him to abandon this training.

He had also experienced a similar episode of pain two years previously while surfing, but this pain initially did not last more than half an hour.

He did not report any chronic nasal dysfunction between these episodes. The radiolucency of the left maxillary sinus (as well as the other sinuses and the ethmoid) was normal, with no signs of mucosal hypertrophy or retention, apparently excluding classical barotrauma.

However, the morphology of the left maxillary sinus suggested the possibility of pneumosinus dilatans (Fig. 1). Pneumosinus dilatans is a bone disease inducing remodelling of the walls of the paranasal sinuses due to osteoclastic and osteoblastic activity. The combination of increased uptake on sodium fluoride (NaF) PET-CT scan and no increased uptake on fluorodeoxyglucose (FDG) PET-CT has been shown to confirm the diagnosis of pneumosinus dilatans [2]. Both PET scans were normal in the present case.

Surgical exploration under general anaesthesia of the left middle meatus, after unciformectomy, demonstrated the absence of either a primary or accessory maxillary ostium. The maxillary sinus membrane was then perforated with the suction cannula at the expected position of the ostium and a 7 mm diameter antrostomy was created with cutting forceps. The middle meatus antrostomy remained patent at the follow-up visit at the first postoperative month.

One year later, the patient had resumed surfing, diving and alpine skiing without any pain.

3. Discussion

The sinus origin of the pain experienced by this patient was retrospectively confirmed by the resolution of this pain after middle meatus antrostomy. However, the absence of the left maxillary ostium observed under general anaesthesia was difficult to predict on CT scan (Fig. 1).

Pneumatization of the left maxillary sinus was not accompanied by any mucosal abnormality or retention opacity, but appeared to be more extensive on the right, leaving expansive impressions on the inferior wall of the orbit and in the alveolar recess. The hypothesis of left maxillary pneumosinus dilatans was proposed. FDG PET-CT scan investigating the mucosa and NaF PET-CT scan investigating bone were performed in order to confirm the diagnosis of pneumosinus dilatans and exclude a diagnosis of simple hyperpneumatization [2]. The absence of bone uptake on NaF PET-CT argued against the hypothesis of pneumosinus dilatans, but the efficacy of middle meatus antrostomy in the treatment of pneumosinus dilatans nevertheless justified surgery [2].

Surgical exploration under general anaesthesia (Fig. 2a) started with unciformectomy of the horizontal segment in order to expose the region of the ostium (Fig. 2b). No ostium, either open or closed, was identified (Fig. 2c, d) [3]. Palpation of the ostial region with the

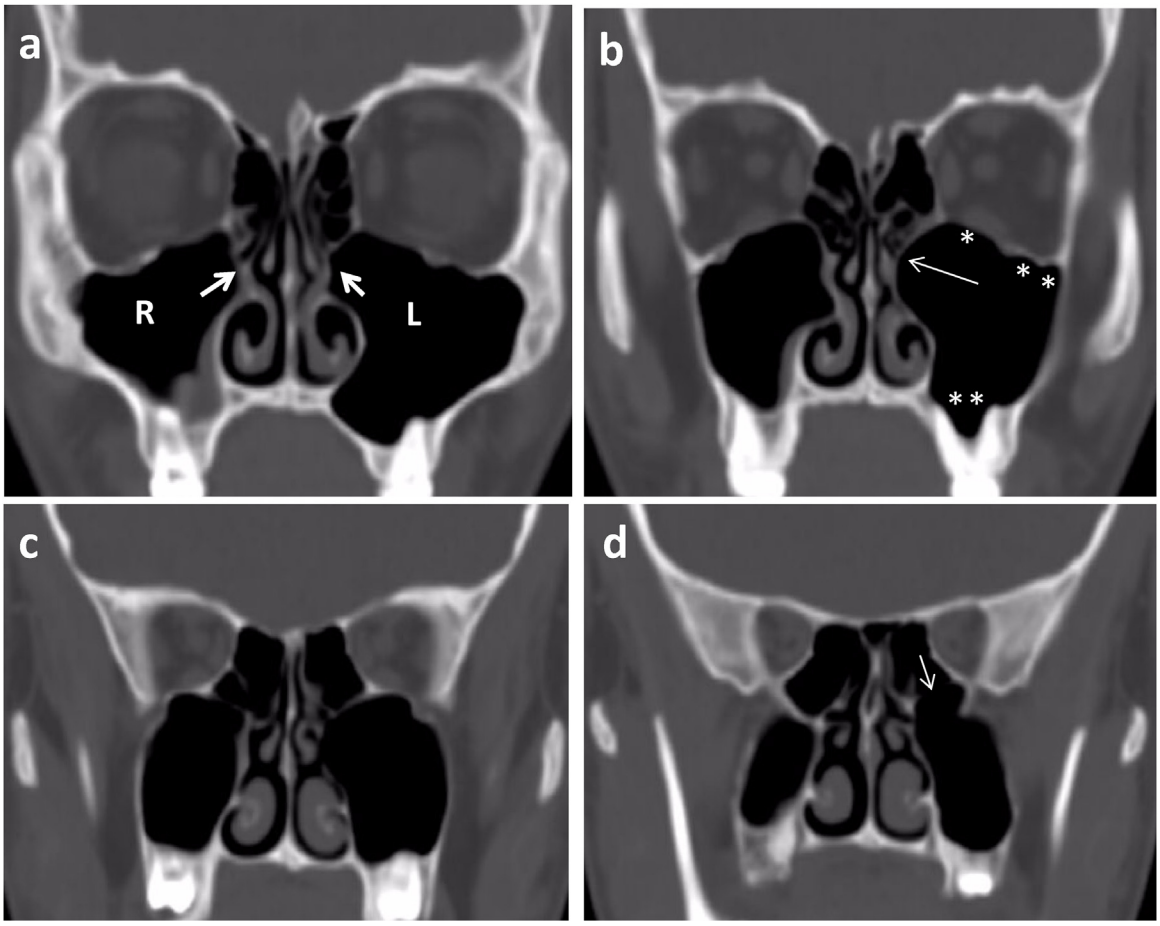

Fig. 1. CT scan of the nose and paranasal sinuses, coronal sections: a: section through the nasomaxillary canal (arrow), which is large on the right side (R) and narrow on the left side (L); the left maxillary sinus appears to be larger than the right maxillary sinus; b: the soft tissue wall of the fontanelle region, which forms the nasomaxillary septum posteriorly to the ostial region, protrudes medially (arrow), obstructing the left middle meatus; the left maxillary sinus appears to be larger than the right maxillary sinus and its polyhedral pneumatization leaves expansive impressions on the inferior wall of the left orbit (*) and in the alveolar recess (**); c: a posterior coronal section confirms the previous findings and the absence of mucosal hypertrophy or retention; d: pneumatization of the left maxillary sinus extends towards the posterior ethmoid, from which it remains separated by only the maxillary membrane (arrow).

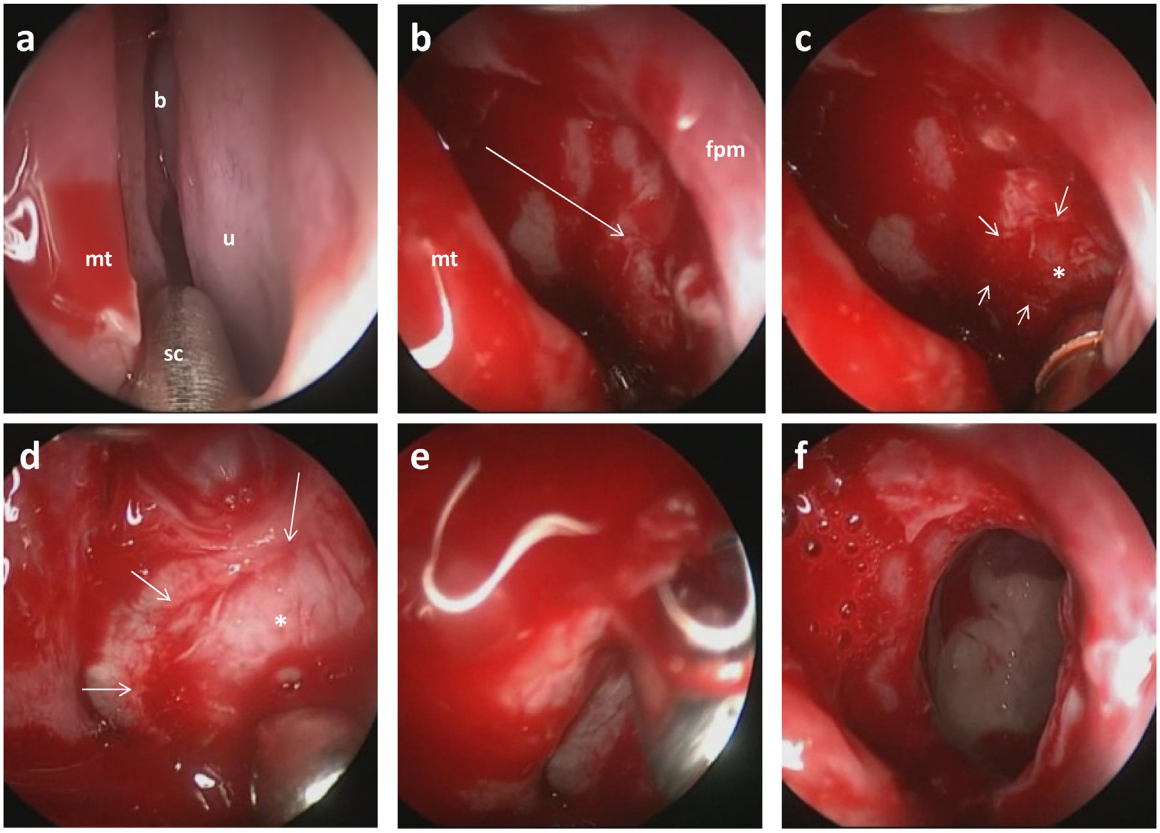

Fig. 2. Surgical exploration of the ostial region of the left maxillary sinus and piercing of the sinus membrane: a: the anatomy of the left middle meatus appears to be normal; b: unciformectomy revealed an imperforate continuous mucosa (arrow); c: palpation with a blunt suction cannula also failed to reveal a patent orifice communicating with the maxillary sinus; this palpation excluded the presence of a thin layer of mucosa (arrows) covering a fibrous membrane (*); d: exposure of the maxillary fibrous membrane (*) by retracting the thin mucosa (arrows); e: pressure on the fibrous membrane produced an air bubble on the surface of the membrane; f: the maxillary fibrous membrane was perforated by an orifice measuring about 7 mm in diameter. (mt: middle turbinate; sc: suction cannula; u: uncinate process; b: bubble; fpm: frontal process of the maxilla).

suction cannula induced the formation of air bubbles on the surface of the sinus membrane (Fig. 2e), which was simply perforated with forceps, creating a limited opening, about 7 millimetres in diameter (Fig. 2f), which was not subsequently enlarged by conventional middle meatus antrostomy extended to the fontanelle region.

The efficacy of middle meatus antrostomy and the pathophysiology of this case of recurrent acute pseudo-sinusitis, or more exactly recurrent acute sinus pain with radiolucent sinuses, can be explained by the evo-devo theory of the sinuses, according to which the sinuses are not formed from ethmoidal air cells but result from disintegration of the bone marrow of the maxillary, frontal and sphenoidal bones, resulting in cavities lined by NO-producing epithelium [4]. The sphincter function of the ostium [3] would regulate the release of boluses of NO [5]. Apart from the various hypotheses proposed to date [6], the real reason for the existence of paranasal sinuses appears to be their respiratory function of production, storage and release on demand of nitric oxide (NO), an aerocrine messenger that increases arterial blood oxygenation in the pulmonary alveoli on demand [7]. The finding of the possible absence of a maxillary ostium appears to be an additional argument in support of the evo-devo theory of the formation of the paranasal sinuses. What explanation can be proposed other than that the maxillary sinus in this patient was pneumatized from ethmoidal air cells in the absence of communication between the two structures? In this patient, regression of the bone marrow creating the sinus cavity lined by its specific membrane failed to create a communication with the ethmoid. Surgical penetration of this membrane certainly did not restore the sphincter function, but NO, which, under stable atmospheric pressure conditions, appeared to diffuse across the membrane before being evacuated (Fig. 2e), was now evacuated via this artificial orifice (Fig. 2f) in the case of sudden pressure variations (altitude, diving, surfing), eliminating the patient's sinus pain.

The role of the paranasal sinus ostium dysfunction in facial pain should now be taken into account and evaluated in the light of this case report. Agenesis of the membranous ostium should therefore be systematically investigated in cases of classical barotrauma with opacity of the paranasal sinuses. The clinical case presented here may correspond to a rare aetiology of facial pain, which should be suspected in the presence of pressure-induced pain. Complete absence of opacity in a closed sinus does not exclude pain due to sinus disease and questions the more conventional concepts concerning ventilation and drainage of the paranasal sinuses.

4. Conclusion

This case illustrates the evo-devo theory of pneumatization of the paranasal sinuses and facilitates interpretation of CT images of the nose and paranasal sinuses in the diagnosis of painful nasal and paranasal sinus dysfunction. This case report paves the way for more detailed studies on the role of the paranasal sinuses in facial disease and respiratory physiology.

Financial statement

No financial disclosures.

Disclosure of interest

The author declares that he has no competing interest.

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