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CASE STUDY

Continuous positive airway pressure is effective in treating upper airway oedema

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Continuous positive airway pressure is effective in treating upper airway oedema. F. Barbé, S. Pons, B. Togores, J. Sauleda, R. Soler, A.G.N. Agustí. ©ERS Journals Ltd. 1996.

ABSTRACT: The case of a patient with diffuse idiopathic skeletal hyperostosis (DISH) and upper airway oedema, is described. The patient presented with alveolar hypoventilation and obstructive apnoeas during sleep.

Intravenous steroids (methylprednisolone, 160 mg·day^{-1}) for 5 days did not reduce the oedema. However, it was rapidly reversed by the use of nasal continuous positive airway pressure (nCPAP). In addition, daytime pulmonary gas exchange was improved and sleep apnoea abolished. This beneficial effect made tracheostomy unnecessary.

This case report suggests that CPAP can be a potentially useful therapeutic alternative to tracheostomy in the clinical management of upper airway oedema.
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Treatment of severe upper airway oedema often requires a tracheostomy. However, this is an invasive procedure that is associated with significant morbidity. We report the case of a patient with diffuse idiopathic skeletal hyperostosis (DISH) [1] and cervical involvement, who presented to the emergency room of our institution because of severe upper airway oedema. The use of nasal continuous positive airway pressure (nCPAP) reduced the oedema and improved the clinical condition of the patient, making tracheostomy unnecessary. This case report suggests that nCPAP may be a valid, noninvasive alternative to tracheostomy in the clinical management of life-threatening upper airway oedema.

Case report

A 73 year old patient was hospitalized because of severe upper airway oedema. The patient had a history of progressive limitation of cervical motility and dysphagia for solid foods over the last few years. During the last weeks, dysphagia had worsened and was complicated by retropharyngeal pain, dyspnoea, inspiratory stridor and dysphonia. Physical examination revealed an obese man (body mass index 35 kg·m^{-2}), with a short neck of limited motility. A nasopharyngoscopy showed significant oedema of the supraglottic area with redundant soft tissue, which occluded the glottic area intermittently. A computed tomography (CT) scan showed ossification of the anterolateral cervical and dorsal spine, with spurs of considerable size that compressed and displaced the hypopharynx towards the supraglottic area (fig. 1).

Following a diagnosis of DISH and upper airway oedema, intravenous therapy with methylprednisolone, 160 mg·day^{-1}), was initiated. However, despite maintaining this therapy for five consecutive days, the clinical condition of the patient worsened, and episodes of upper airway airflow limitation leading to obstructive apnoea appeared during the daytime. Awake arterial blood gases breathing room air revealed: pH 7.42, arterial oxygen tension (P_{a,O_2}) 8.26 kPa, arterial carbon dioxide tension (P_{a,CO_2}) 7.33 kPa. A nocturnal polysomnographic study (Ultrasom, Nicolet, USA) showed frequent obstructive apnoeas with an apnoea-hypopnoea index of 45 event·h^{-1}. Titration of nCPAP demonstrated that a positive pressure of 12 cmH_2O abolished these respiratory events. Interestingly, because the patient was overweight severe snorer, an overnight domiciliary pulse oximetry had been recorded 6 months before hospitalization. The results from this did not suggest the presence of obstructive sleep apnoea at that time.

Twenty four hours after starting ventilatory support with nCPAP, the clinical status of the patient had improved

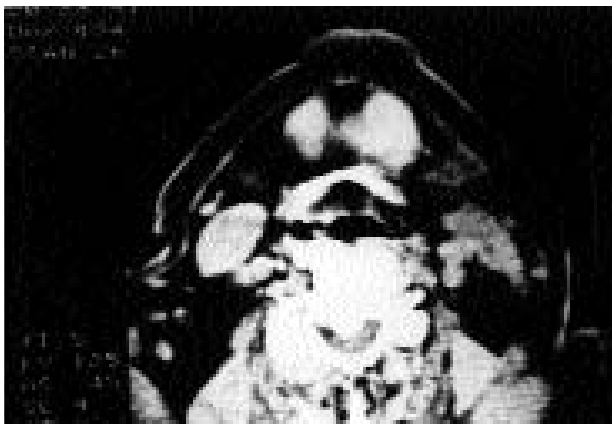

Fig. 1. – Cervical computed tomography (CT) scan showing a great anterior osteophyte compressing the hypopharynx and displacing the supraglottic area.

significantly, and a second nasopharyngoscopy (performed 48 h after starting nCPAP) showed a considerable decrease of the hypopharyngeal oedema. Nasal CPAP was continued at night for 8 days. At discharge (awake and breathing room air), pH was 7.46, P_{a,O_2} 11.1 kPa, and P_{a,CO_2} 5.60 kPa. The patient used nCPAP at home every night. Two months later, an ambulatory nasopharyngoscopy showed complete resolution of the upper airway oedema. A second sleep recording (without nCPAP) demonstrated an apnoea-hypopnoea index of 15 events·h^{-1}.

Discussion

DISH is a disease characterized by the ossification of the anterolateral spine, creating osteophytes of considerable size [1]. Recently, it has been shown that DISH can cause obstructive sleep apnoea (OSA) [2]. The present case report supports such a potential association. However, two aspects deserve further comment. Firstly, the potential role of upper airway oedema in the pathogenesis of OSA in our patient and, secondly, the usefulness of nCPAP as a noninvasive therapeutic alternative in the clinical management of life-threatening upper airway oedema.

The pathogenesis of the oedema associated to DISH is not well understood. It has been suggested that the cycle inflammation-oedema-muscle spasm-pain due to bone growth might explain it [3, 4]. Therefore, it is theoretically possible that the progression of DISH caused severe upper airway oedema and that the latter, in turn, was the cause of the OSA in our patient. The subacute clinical course of our patient before attending the emergency room, and the fact that pulse oximetry 6 months before hospitalization did not suggest OSA, support this view. Nonetheless, because the patient was overweight and a severe snorer, we cannot exclude the possibility that he originally had DISH and OSA (not detected by pulse oximetry), and that the repeated ineffective efforts of OSA, which sucked the upper airway mucosa into the airway obstructed by DISH, were in fact the cause of the oedema.

Nasal CPAP is widely used in the treatment of OSA [5], but its mechanism of action is debated. It is postulated that it may act as a pneumatic splint in the oropharyngeal area [5, 6], may stimulate local receptors enhancing the patency of the upper airway [7, 8], and/or may reduce local oedema [5, 9, 10]. The mechanism by which the latter can occur is not well understood either, but it may be related to the effects of nCPAP upon upper airway circulation and/or inspiratory work [5]. In our patient, upper airway oedema did not improve with *i.v.* steroids. By contrast, soon after starting nCPAP, a dramatic improvement in the clinical status of our patient was observed and pharyngoscopy showed a marked decrease of local oedema with a parallel increase in upper airway calibre. Furthermore, 2 months later, the apnoea-hypopnoea index was significantly reduced. Therefore, we postulate that the clinical and physiological improvement of our patient might have been directly related to administration of nCPAP.

In summary, this case report shows that nCPAP is effective in the clinical management of upper airway oedema. We suggest that nCPAP might be useful in the clinical management of upper airway oedema as a noninvasive therapeutic alternative to tracheostomy.

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