Recurrent acute pulmonary oedema after aortic and mitral valve surgery due to trachea malacia and obstructive sleep apnoea syndrome

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In this report we describe a patient with recurrent episodes of acute pulmonary oedema after aortic and mitral valve surgery. The first episode of pulmonary oedema was caused by mitral valve dysfunction. The second episode of pulmonary oedema was not clearly associated with a mitral valve problem, but reoperation was performed in the absence of another explanation. After the third episode of acute pulmonary oedema occurred, the diagnosis of obstructive sleep apnoea syndrome (OSAS) was considered and confirmed. After starting treatment with continuous positive airway pressure (CPAP) during his sleep the patient had no further episodes of acute respiratory failure. Our case demonstrates that acute pulmonary oedema after cardiothoracic surgery can be caused or at least be precipitated by OSAS and should be suspected in patients with unexplained episodes of (recurrent) pulmonary oedema. (*Neth Heart J* 2008;16:310-2.)

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A 74-year-old male patient was referred from another hospital for aortic valve and mitral valve replacement. He had aortic valve stenosis and mitral valve regurgitation diagnosed seven years previously. His left ventricular function (LVF) evaluated by echocardiography was normal, and left ventricular hypertrophy was present. His medical history further revealed paroxysmal atrial fibrillation, pacemaker implantation for AV nodal disease, hypertension and renal insufficiency with a creatinine clearance of 30 ml/min. His pulmonary function before surgery showed a forced vital capacity (FVC) of 2.61 litres (73% of predicted), a forced expired volume in one second (FEV₁) of 1.55 litres/second (55% of predicted) and an FEV₁/FVC of 0.55 (75% of predicted), i.e. moderate-obstructive pulmonary function. The diagnosis of obstructive sleep apnoea syndrome (OSAS) had not been made in this patient.

Surgery was performed without complications (Sorin Bicarbon S 21 mm aortic valve and Sorin Bicarbon S 27 mm mitral valve) and after five days the patient was transferred back to the department of cardiology of the referring hospital. Six weeks later the patient was readmitted to the intensive care unit (ICU) because of acute respiratory failure due to pulmonary oedema. Transoesophageal echocardiography revealed intermittent dysfunction of the artificial mitral valve at the original posterior leaflet, with a mitral valve stenosis. During surgical revision it appeared that the caudal leaflet had some resistance causing mitral valve insufficiency. The valve was repositioned in the anatomical position. Two days after surgery, however, the patient again had an episode of pulmonary oedema and respiratory failure (PaCO₂ up to 15 kPa, 122 mmHg measured in arterial blood gas) for which he was readmitted to the ICU. Systemic and pulmonary hypertension (PAP systolic 80 mmHg) was found. A transoesophageal echocardiography at this time showed no valve dysfunction and a normal LVF. Nevertheless, in the absence of other causes, mitral valve dysfunction was suspected and it was decided to replace the artificial

Figure 1. Partial pressure of carbon dioxide ($p\text{CO}_2$). X-axis: days since first operation (dotted vertical lines are days when surgery was performed). Y-axis: $p\text{CO}_2$ in kPa (measured in arterial blood).

mitral valve with a Carbo Medics 27 mm mitral valve. After his third operation, the patient still had recurrent episodes of acute pulmonary oedema, with the same characteristics as before, typically occurring on the second day after surgery. Echocardiography was repeated a number of times and showed that the valves were functioning normally. Following a period of gradually increasing PaCO_2 , each episode of acute pulmonary oedema and respiratory failure occurred at the end of the night/early in the morning, and started with dyspnoea and a high blood pressure followed by respiratory failure within minutes, not relieved by nitroglycerin or diuretics (figure 1).

These episodes were not related to medications nor were there indications of neurological causes or signs of cardiac ischaemia. Closer examination of a chest X-ray during one such episode suggested a narrowing of the trachea (figure 2).

Bronchoscopy revealed coronal narrowing and sagittal widening over the whole length of the trachea, causing severe dyskinesia and stenosis with the tendency to collapse. This tracheal deformity is called a sabre sheath deformity and is strongly associated with coronary obstructive pulmonary disease (COPD).¹ Polysomnography revealed a mixed pattern of obstructive and central apnoeas with oxygen desaturation <70%.

We concluded that the episodes of acute respiratory failure with pulmonary oedema were caused or at least precipitated by repetitive oxygen desaturation due to sleep-disordered breathing. The obstructive apnoeas might have been caused by the tracheal sabre sheath deformity and could have been aggravated by intubation. Sedatives and the exhausted state of our patient after each operation might also have played a role. After starting treatment with continuous positive airway pressure (CPAP) during his sleep, no episodes of acute respiratory failure necessitating artificial ven-

Figure 2A. Chest X-ray during acute respiratory insufficiency.

Figure 2B. Control chest X-ray during a clinically stable episode.

tilation occurred. During the next year of follow-up at our outpatient clinic he was doing acceptably well, with mainly problems of right-sided heart failure, treated with diuretics.

Discussion

OSAS was clearly documented for the first time in 1966² and is characterised by upper airway narrowing or collapse resulting in repetitive episodes of hypoxaemia, swings in intrathoracic pressure and sleep fragmentation from arousals. The prevalence in Western populations is estimated to be 5%.³ Anatomical

causes of upper airway collapse are the most frequent cause of OSAS.⁴ OSAS can result in an increase in sympathetic outflow and arterial pressure by mechanisms not yet fully understood. Negative intrathoracic pressure, asphyxia-induced chemoreflex stimulation and arousal from sleep have all been implicated, although chemoreflex mechanisms seem to be more important than negative intrathoracic pressure.^{5,6}

Although OSAS is usually associated with chronic cardiovascular conditions (hypertension, ischaemic heart disease, congestive heart failure and stroke), acute cardiovascular conditions associated with OSAS have also been described: tachycardia, episodic hypertension, dysrhythmias, heart block and angina.¹ OSAS also creates right ventricle pressure overload by the mechanical effects of pulmonary obstruction and as such can also affect left ventricular diastolic function.^{7,8} We also know from two previous case reports that OSAS may present with recurrent pulmonary oedema.^{9,10} In our patient, the first mitral valve reoperation was necessary, due to a demonstrable dysfunction of one of the mitral valve leaflets. The second reoperation was not effective, and OSAS was now present to explain the events. In retrospect, the typical timing of the events after surgery should have warranted attention towards a post-surgery 'amplification' of a possibly mild OSAS.⁸ Other clues leading to the diagnosis were an inappropriate PaCO₂ rise during pulmonary oedema,¹¹ occurrence of pulmonary oedema in the morning hours and the narrowed trachea on the X-ray of the lungs.

The current treatment of OSAS consists of nasal CPAP, which was introduced by Sullivan et al.¹² It was recently demonstrated that CPAP also improves the condition of some patients with acute cardiogenic pulmonary oedema and can improve left ventricle function.^{7,13,14}

In summary, OSAS should be suspected in patients with (recurrent) respiratory insufficiency due to pulmonary oedema, especially if no other causes are found. ■

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