Findings that shed new light on the possible pathogenesis of a disease or an adverse effect

Right main bronchus perforation detected by 3D-image

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Summary

A male metal worker, who has never smoked, contracted debilitating dyspnoea in 2003 which then deteriorated until 2007. Spirometry and chest x-rays provided no diagnosis. A 3D-image of the airways was reconstructed from a high-resolution CT (HRCT) in 2007, showing peribronchial air on the right side, mostly along the presegmental airways. After digital subtraction of the image of the peribronchial air, a hole on the cranial side of the right main bronchus was detected. The perforation could be identified at the re-examination of HRCTs in 2007 and 2009, but not in 2010 when it had possibly healed. The occupational exposure of the patient to evaporating chemicals might have contributed to the perforation and hampered its healing. A 3D HRCT reconstruction should be considered to detect bronchial anomalies, including wall-perforation, when unexplained dyspnoea or other chest symptoms call for extended investigation.

BACKGROUND

The location of lung rupture causing spontaneous pneumothorax (SP) has been supposed to be in the periphery of the lung, with its thinner and possibly more vulnerable anatomy.^{1–2} However, ‘emphysema like changes’ (ELCs) have been documented in central lung regions, in SP patients, who have never smoked.³ We hypothesised that the central location of the ELC may suggest weakness and potential rupture of proximal airways as a possible cause of SP. In this paper, we report on a perforation in the right main bronchus that has caused unusual symptoms for barotrauma.

We call the attention of the reader to such a possibility to motivate further observations and investigations.

This case report shed new light on;

- ▶ a novel diagnostic procedure—the first 3D/radiological documentation of the existence/healing of a bronchial perforation, and its correlation to the clinical symptoms.
- ▶ the existence of a distinct location of a potential rupture in the wall of a presegmental airway generation.
- ▶ a possible pathogenetic mechanism of barotrauma, including spontaneous pneumothorax.

CASE PRESENTATION

We report on a 184 cm tall, never-smoking man born in 1952. He was free of α 1-antitrypsin deficiency, inflammatory, malignant or cardiopulmonary diseases and had worked in the metal industry for 20–25 years before 2003, with exposure to iron, steel, aluminium, copper, different coolants and lubricants. Several members of his family had had spontaneous pneumothorax (SP). He contracted a right-sided SP in 1986, verified by chest x-ray and was treated with underwater seal. He had no relapse of the

SP, but high-resolution CT (HRCT) showed ELCs in the central regions of the lung.³ He sometimes had dyspnoea, which became debilitating from December 2003. The patient suffered from pain in the dorsal part of the thoracic wall, December 2005, with increasing dyspnoea, as if breathing was blocked in his right lung. Cyanotic nails and lips were noted, but no blood gas analysis was made. His symptoms were made worse by occupational exposure to evaporating chemicals. However, several examinations in a local primary care clinic provided no diagnosis.

INVESTIGATIONS

A 3D bronchial image was reconstructed⁴ from both the horizontal and vertical series of 2D sections of a HRCT in 2007. It showed previously undetected peribronchial air located mainly along the presegmental airway generations, on the right side (figure 1). After digital subtraction of the peribronchial air by a special segmentation technique, a hole, approx 1.5×2.0 mm, was detected on the cranial side of the right main bronchus, near the origin of the right, upper third generation of airways (figure 2). Knowing the location of this hole on the 3D-image, we could identify it at a re-examination of the HRCT images in 2007.

DIFFERENTIAL DIAGNOSIS

X-ray and scintigraphy of the skeleton, spirometry and metacholine tests were negative. HRCT of the lungs in 2007 was considered negative, before the 3D-image analysis.

OUTCOME AND FOLLOW-UP

In 2009, we repeated the HRCT with a limiting isotropic spatial resolution of 0.6 mm and the 3D-reconstruction of the airways. It confirmed the results from 2007 and showed some decrease of the lumen of the hole, at the

Figure 1 Previously undetected, peribronchial air located mainly along the presegmental airway generations at the right side. This 3D-image comprises the airways vertically 6 cm above and 14 cm below the carina.

level of the mucous membrane of the main bronchus. Two control HRCT examinations, in August and September 2010, showed that the perforation in the right main bronchus had healed.

DISCUSSION

To our knowledge, no similar cases have been reported. We searched for bronchial anomalies⁵ with HRCT, not visualised by bronchoscopy, in patients who had never smoked but had suffered spontaneous pneumothorax. The case reported here suffered from SP in 1986, verified by a conventional chest x-ray. When severe symptoms reappeared in 2003, SP could not be demonstrated. Since the symptoms persisted, repeated investigations were made and a bronchial perforation could be demonstrated in 2007 using HRCT and 3D-reconstruction of the airways.

The bronchial perforation was documented in 2007 and 2009, its partial healing in 2009 and its final healing in 2010 by 3D and HRCT techniques. These documented facts were in accordance with the debilitating state in 2007, the improvement in 2008–2009, and the total recovery of the patient in 2010. The patient now works full time. The objectively documented healing of the perforation and the clinical improvement and the full recovery of the patient present a correlation between the perforation

and the clinical course of events. We consider these facts to be direct evidence for the existence and role of the perforation between 2007 and 2010. The correlation in time, between the healing of the perforation and the ceasing of the patient's symptoms, supports adequately, indirectly, that the onset of the debilitating dyspnoea in 2003 was caused by the onset of the bronchial perforation with subsequent leakage of air to the peribronchial space. For this reason, we attribute the clinical symptoms and course of events as indirect evidence for the onset, existence and role of the perforation between 2003 and 2007. Further leakage may have caused the dramatic increase in chest pain and dyspnoea which the patient complained of 2 years later.

Furthermore, no indication or any proof was found by an intense search for any other factor which could have been considered to have any relation to the perforation. At the first report of a non-traumatic bronchial perforation on the main bronchus, the detection and documentation of the perforation and its result, the accumulation of the peribronchial air are the relevant, primary issues that the readers should be informed about.

However, the initial SP in 1986 may not have been caused by the bronchial hole; it may not have existed at that time. Rather, the hole may reflect one or more anomalies in the

Figure 2 By digital subtraction of the peribronchial air, a hole was detected on the right main bronchus, near the origin of the third right upper airway generation. The insert to the right in the panel shows the origin of an accessory bronchus³ behind the first segmental airway generation of the right apical posterior segment.

bronchial tree. We assume that the occupational exposure to evaporating chemicals might have contributed to the perforation and hampered its healing.

One may ask why the bronchial hole and peribronchial air passed undetected over a number of years. However, the peribronchial air will not be detected by the highest resolution on 64 slice HRCT (voxel: $0.625 \times 0.7 \times 0.7$ mm, number of rows: 64, thickness of rows: 0.625 mm, interval: 0.625 mm), if its thickness is less than 1.3 mm. We have to assume that the peribronchial air layer was not large enough to be visualised by HRCT in any of the investigations that were performed.

Moreover, the 3D reconstruction applies a numerical correction procedure by adjusting the neighbouring contour lines to each other. This technique enables the visualisation of the airway contour of the 2D images, neglecting all other information of the images. This allows the 3D-reconstruction algorithm to show the surface of the airways in a much more sensitive and precise way than can be achieved by 2D images. By this means, the bronchial hole could be detected.

The risk of barotraumas (SP) is 80%, if patients are ventilated mechanically, with a pressure of 70 cm H₂O (70 millibar).⁶ An atmospheric pressure fall of 10 millibar or more triggers SP.⁷ The hole was approx. 1.5×2.0 mm and the pressure drop across the hole depends on the magnitude of the leak to the peribronchial space. With no leak there will be no pressure drop. With a leak corresponding to the area of the hole in relation to the lumen area of the airway, reflecting potential leak flow, the pressure drop will be no higher than 0.5 millibar,⁸ suggesting that the leak must have been too small to cause any SP.

Figure 1 demonstrates the accumulation of the peribronchial air in the peribronchial space, a proof for the existence of the leakage. Without the leakage, no air could have been accumulated in the peribronchial space. We have further

proved the morphology of the leakage, the perforation on the right main bronchus (figure 2).

The consequence of an existing leakage is three fold;

- a larger proportion of the inhaled air gets into the lungs
- the smaller proportion of the inhaled air, which can not get into the lungs, gets into the peribronchial space (figure 1) and the amount of air which could have been inhaled into the lungs will be reduced by the volume of air, which leaked into the peribronchial space (the peribronchial air is the evidence for the leakage through the perforation)
- the increased/expanded volume of the peribronchial space (figure 1) comprimates the surrounding pulmonary parenchyma, hampering its gas exchange.

These synergetic effects enhance each other and contribute to a deteriorated ventilation, manifested in, besides debilitating dyspnoea, cyanotic nails and lips and the feeling of inability to breathe on the right side of the chest in several years after 2003, while both the open hole and the peribronchial space were documented on the right/affected side.

Regarding the possible need of this method, we present the estimated number of patients who seek ward due to the recognised symptoms of spontaneous pneumothorax, but their complaints cannot be verified by radiologic examinations. Their number is about 20%–25% of all SP patients (personal communication by H Jorulf), that is, approximately 400 patients/year only in Sweden. Such patients, or those who suffer from unexplained dyspnoea, including those with occupational exposure to chemicals, should be considered for HRCT and 3D-image reconstruction, to detect lung abnormalities, for example, bronchial wall perforation.

We should also consider the accidents in the changing ambient pressure especially, pulmonary barotrauma in

divers. In Sweden 30–40 divers are treated for decompression illness and 4–5 lethal diver accidents are registered yearly,^{9 10} while the establishment of the correct morphologic pathogenetic diagnosis regarding their airways is still missing.

The correct estimation of the economic effectiveness of the 3D technique is not possible within the frame of a case report. Considering only the price of investigations we may declare that the price of the more than 100 traditional examinations 2003–2007 was about 150% of the price of the used 3D technique. In the presented case, it is obvious that 3D technique was cost effective and superior compared to the traditional, ineffective methods, which did not establish any diagnosis.

The cost of this specific 3D technique, which can visualise the structure of the bronchial tree, perforation, deformity of the bronchial wall and bronchial lumen, may be reduced by incorporating that software into CT.

In summary, the perforation and healing of a central airway corresponding to the clinical symptoms was detected by 3D image.

Learning points

- ▶ Subjectively recognised but not radiologically verified symptoms of spontaneous pneumothorax/barotrauma or unexplained dyspnoea might be caused by bronchial perforation—accumulation of peribronchial air.
- ▶ Chest x-rays and spirometry do not contribute to diagnosis in such cases.
- ▶ HRCT can visualise peribronchial air better with 3D-reconstruction than without.
- ▶ The right main bronchus may be perforated, at a distinct location.

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