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CASE STUDY

Sicca syndrome and silicoproteinosis in a dental technician

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Sicca syndrome and silicoproteinosis in a dental technician. R. Orriols, J. Ferrer, J.M. Tura, C. Xaus, R. Coloma. ©ERS Journals Ltd 1997.

ABSTRACT: Secondary sicca syndrome has been described in silicotic and exposed nonpneumoconiotic individuals. Systemic autoimmune pathogenic mechanisms have been thought to be responsible. Salivary gland mineral deposition has not so far been implicated.

We describe the case of a dental technician who sand-blasted dental prostheses and developed silicoproteinosis and sicca syndrome. Immunological studies were negative. Scanning electron microscopy study and energy-dispersive X-ray analysis were carried out in the patient's lung and salivary gland and in two salivary glands from nonexposed individuals diagnosed with sicca syndrome.

High peaks of silicon and aluminium were detected in the patient's lung and salivary gland. Silicon/sulphur and aluminium/sulphur ratios were higher in the patient's glands (0.612 and 0.402, respectively) than in the two control salivary glands (0.193 and 0 and 0.127 and 0, respectively).

We suggest that, in this case, sicca syndrome could have been caused by inorganic deposition.

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Primary Sjögren's syndrome is now accepted as a generalized autoimmune exocrinopathy [1]. Secondary Sjögren syndrome occurs as a complication of a connective tissue disease [2], or some other entities [3].

The case of a dental technician who sand-blasted dental prostheses and developed lung silicoproteinosis and sicca syndrome is described. Microanalytical study of the salivary gland suggested that inorganic deposition could have played a role in the pathogenesis of the glandular involvement in this patient.

Case report

The patient was a 29 year old, nonsmoking man, who started to work as a dental technician at 16 yrs of age. He had worked in sand-blasting from 16–22 yrs of age, for on average 4–5 h a day, without continuous respiratory protection. He had worked with acrylic resins from 22–26 yrs of age.

When the patient was 24 yrs of age, he began to experience dry cough, dyspnoea, slight fever and weight loss. Chest radiography showed an interstitial pattern, more marked in the upper lobes and parahilar lymph nodes. Respiratory function examination revealed: forced vital capacity (FVC) 3.0 L (65% of predicted); forced expiratory volume in one second (FEV_1) 2.4 L (61% pred); FEV_1/FVC 80%; and transfer factor of the lungs ($T_{L,CO}$) 5.5 mmol·min^{-1}·kPa^{-1} (52%). Gallium scintigraphy disclosed increased uptake in both lungs, lachrymal glands and right parotid glands. Bronchoalveolar lavage (BAL) was performed and showed 1.9×10^9 cells·L^{-1}, with 59% macrophages and 41% lymphocytes (CD4+ 41%, CD8+ 54%). Transbronchial biopsy revealed rounded formations with a lymphoplasmocytic cellular component. The patient stopped working and received treatment with prednisone for 8 months. At 28 yrs of age, he was referred to our centre for evaluation as a possible lung transplant recipient, owing to the progression of his lung disease.

On admission, the patient complained of dyspnoea on slight exertion. He also referred to oral and ocular dryness of 1 year's duration. Bilateral lung rales were heard. Pulmonary functional examination revealed: FVC 1.6 L (35% pred); FEV_1 1.4 L (38% pred); total lung capacity (TLC) 2.6 L (42% pred); $T_{L,CO}$ 3.4 mmol·min^{-1}·kPa^{-1} (32%); $T_{L,CO}$/alveolar volume (V_A) 1.86 mmol·min^{-1}·kPa^{-1}·L^{-1} (89%). Arterial blood gas values breathing room air were pH 7.40; arterial oxygen tension (P_{a,O_2}) 6.3 kPa (47 mmHg); arterial carbon dioxide tension (P_{a,CO_2}) 4.5 kPa (34 mmHg), HCO_3^- base excess (BE) 0.1 mmol·L^{-1} and arterial oxygen saturation (S_{a,O_2}) 80%. Computed tomography (CT) scan revealed: opacities and linear shadows in the upper lobes with destructive changes; ground-glass areas in middle and lower lobes; pleural thickening more prominent within the upper fields; and multiple calcified hilar and mediastinal lymph nodes.

Schirmer's test and rose-bengal staining were positive for keratoconjunctivitis sicca. There was no hypergammaglobulinaemia and immunological studies, including antibodies against extractable nuclear antigens for deoxyribonucleic acid (DNA), Ro (SSA), La (SSB), Sm, nuclear ribonucleic acid (nRNA) and Scl-70, were negative. Lip biopsy yielded a lymphoplasmotic infiltrate, showing cellular clusters and slight glandular sclerosis. Study of the glandular biopsy by polarized light showed no birefringent particles, and pathological features

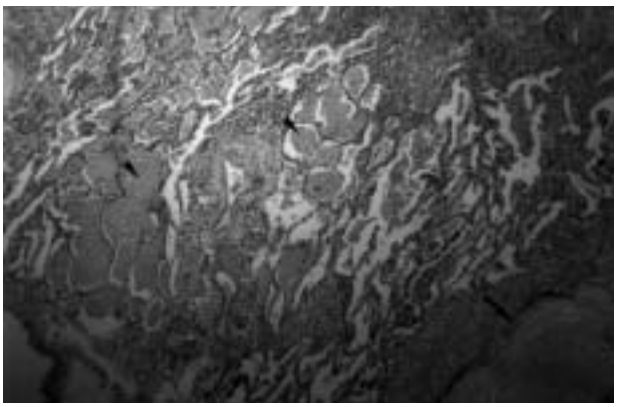

Fig. 1. – Fibrocellular nodule (arrow), interstitial mononuclear cell infiltrate and alveolar proteinosis (arrowheads) in lung specimen. (Periodic-acid-Schiff staining; Internal scale bar=200 µm).

suggestive of acrylic resin accumulation were not observed.

The patient underwent unilateral lung transplantation. Histological study of the excised lung revealed an amorphous granular eosinophilic substance that stained by the acid-Schiff reaction in airspaces, interstitial mononuclear cell infiltrate and some organized reticular nodules, particularly in the area of the respiratory bronchioles, all of which suggested silicoproteinosis (fig. 1). Polarized light study showed birefringent particles, but accumulation of acrylic resin was not found.

Microanalytical studies

Scanning electron microscopy (SEM) and energy-dispersive X-ray analysis (EDXA) were carried out in lung and salivary gland. A specimen of each tissue, embedded in paraffin, was deparaffinated with xylol and transferred to acetone, and later dried by the critical point technique using liquid CO_2. The specimens were attached with albumin to carbon slices and covered with a 20 nm thick conductive carbon layer. The absence of inorganic contamination in paraffin, carbon slices and albumin was verified by EDXA. Element analysis was carried out using an energy-dispersive X-ray analyser with an Si (Li) detector (EDX 9800 PV; IL, USA) fitted to a SEM (Philips 515; The Netherlands). Working conditions for EDXA were as follows: accelerating voltage 20 kV; distance from the specimen to the detector 34 mm; tilt angle towards the X-ray detector 30°; and spot diameter 200 nm. The element content in the particles was determined by EDXA, by peak integrals on the X-ray spectra after subtraction of the background. Moreover, EDXA was carried out at a constant magnification of ×625. Five areas were analysed in each specimen, and the final value determined for each element was the average of the five determinations. Analysis time was always 100 s.

To obtain a value for each element, the height of the peak was divided by the noise (peak/background), since background represents dispersive radiation independent of the element detected [4]. All values are expressed as an element/sulphur ratio since, as previously demostrated in lung [5] and in pleura [6], sulphur (S) is an internal standard of tissue mass. Digital EDXA mappings for Si and Al were carried out in order to determine their distribution in the glands.

Material from the patient's protective mask was analysed by electronspectroscopy for chemical analysis (ESCA) to rule out beryllium exposure [7].

Two samples from individuals with primary sicca syndrome and no known significant exposure to inhaled inorganic dust were also studied as controls. Previous examination by polarized light showed no particles.

Results

A high number of silica and aluminium particles were detected in the patient's lung and mask material.

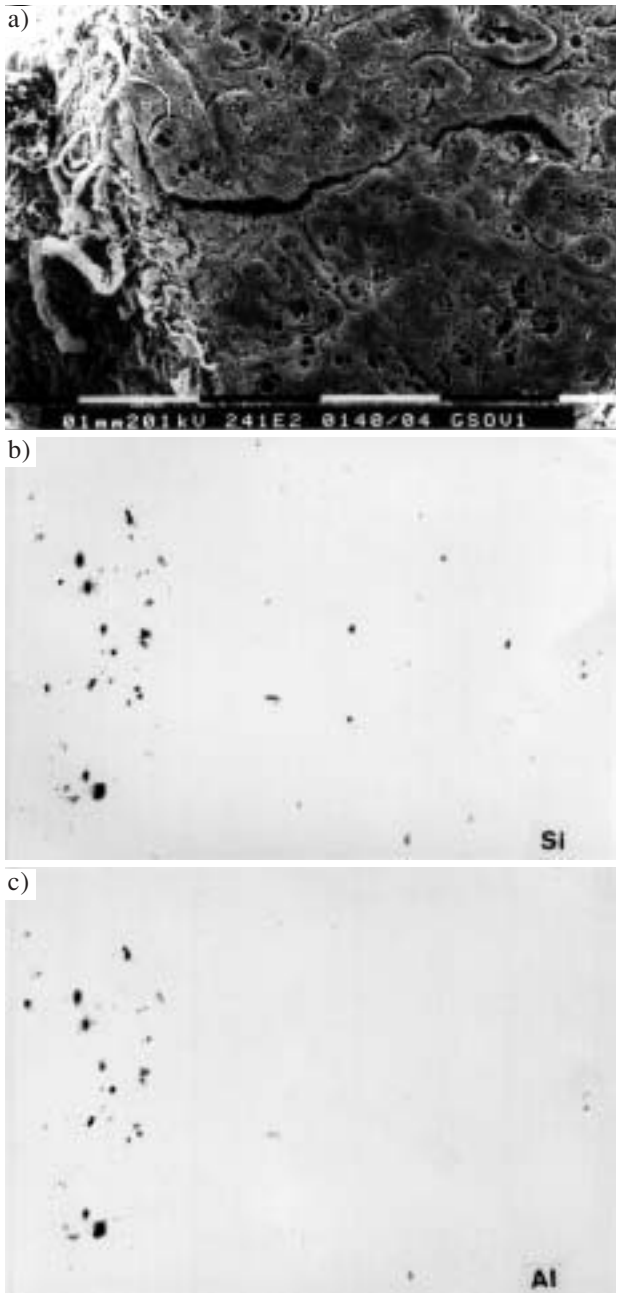

Fig. 2. – Scanning electron microscopy (SEM) and energy dispersive X-ray analysis (EDXA) of a salivary gland cross-section. a) SEM image; b) silicon (Si) digital mapping; c) aluminium mapping. Internal scale bar=100 µm.

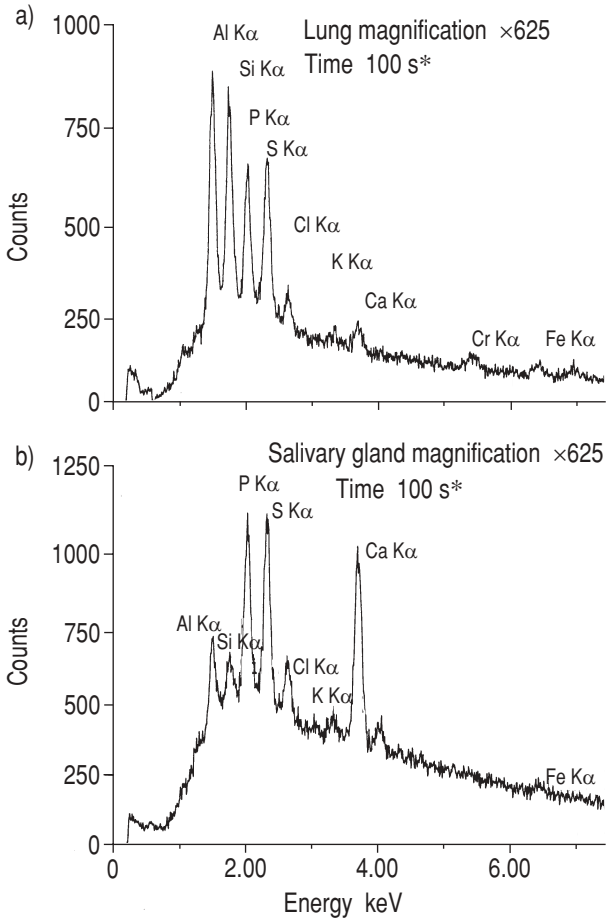

Fig. 3. – Energy dispersive X-ray analysis (EDXA) of the patient's lung and salivary gland. a) EDXA of lung; b) EDXA of salivary gland. Kα: characteristic X-ray line emitted from the K shells of the element during high energy electron bombardment. *: analysis time in live seconds (*i.e.* real time).

Table 1. – Element content of tissue from the salivary gland of the patient and two controls

Element/sulphur ratio	Patient	Controls	
		No. 1	No. 2
Si/S	0.612	0.193	0.127
Al/S	0.402	0	0
Ca/S	0.880	0.814	0.681

Si: silicon; Al: aluminium; Ca: calcium; S: sulphur.

Furthermore, silicon and aluminium, but no beryllium, were detected in the mask material. No particles were detected in the patient's salivary gland or those of the two controls. However, digital EDXA mapping showed silicon and aluminium in the patient's gland, particularly in the outer area (fig. 2). Low magnification EDXA showed silicon and aluminium to be the elements predominantly detected in the patient's lung and salivary gland (fig. 3). The element content of tissue from the salivary glands of the patient and two controls are presented in table 1. Si/S and Al/S values are clearly higher in the gland of the patient than in controls, whilst Ca/S values are similar.

Discussion

In dental technicians, pneumoconiosis is related to silica and other minerals. Noncomplicated silicosis and interstitial fibrosis are the two most common pathological pictures [8]. Although accelerated or acute silicosis with alveolar proteinosis has been described in sand-blasters in several occupations, to our knowledge it has not previously been described in dental technicians. Acrylic resins have been related to alveolitis and interstitial pneumonia in a dental sand-blaster [9], but in the present case the histological and microanalytical data fail to support the aetiological implication of acrylic resins.

Sjögren's syndrome, associated with other collagenopathies, has been described in silicotic and exposed non-pneumoconiotic individuals [10, 11]. It has been suggested that silica might act as a nonspecific activator of autoimmunity, and that, in certain subjects, silica might lead to the appearance of collagenopathies [2]. However, as in the present case, SANCHEZ ROMAN *et al*. [12] described isolated sicca syndrome not associated with other collagenopathies in three workers occupationally exposed to silica. The hypothesis that the glandular disorder is due to a local effect of the mineral should also be considered. In cases of massive exposure, as in the present patient, the mineral deposited in the lung may later be redistributed by the bloodstream and lymphatic routes. Furthermore, extrapulmonary inorganic deposits have been found in patients with inhalatory exposure to mineral dust [13], though not in salivary glands.

In the present patient, the negative immunological tests and the finding of a high glandular content of silicon and aluminium suggest that the glandular involvement might be due to this inorganic deposition. In fact, particles composed of these elements have proved to be pathogens in the lung and other tissues [14, 15]. The presence of calcium, on the other hand, appears to be a secondary fact, since collagen in fibrosed tissue can stimulate deposition of this element [16]. In fact, calcium was also found to be high in the two individuals with sicca syndrome without known exposure to inhaled inorganic dust.

We suggest that the deposition of inorganic material in the salivary glands might have contributed to sicca syndrome in this patient.

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