Sicca syndrome and silicoproteinosis in a dental technician


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.

*Servei de Pneumologia, Hospital General Universitari Vall d’Hebron, and **Consell Superior d’Investigacions Científiques, Barcelona, Spain.

Keywords: Sicca syndrome silicosis X-ray microanalysis

Received: March 4 1996
Accepted after revision October 25 1996

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 (FEV1) 2.4 L (61% pred); FEV1/FVC 80%; and transfer factor of the lungs (TL,CO) 3.4 mmol·min⁻¹·kPa⁻¹ (32%); TL,CO/alveolar volume (VA) 1.86 mmol·min⁻¹·kPa⁻¹·L⁻¹ (89%). Arterial blood gas values breathing room air were pH 7.40; arterial oxygen tension (Pa,O₂) 6.3 kPa (47 mmHg); arterial carbon dioxide tension (Pa,CO₂) 4.5 kPa (34 mmHg); HCO₃⁻ base excess (BE) 0.1 mmol·L⁻¹ and arterial oxygen saturation (Sao₂) 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 lymphoplasmocytic infiltrate, showing cellular clusters and slight glandular sclerosis.

Study of the glandular biopsy by polarized light showed no birefringent particles, and pathological features...
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 CO₂. 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 demonstrated 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 electronmicroscopy 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. 1. – Fibrocellular nodule (arrow), interstitial mononuclear cell infiltrate and alveolar proteinosis (arrowheads) in lung specimen. (Periodic-acid-Schiff staining; Internal scale bar=200 µm).

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.
Ca/S values are similar.

In the gland of the patient than in controls, whilst presented in table 1. Si/S and Al/S values are clearly high-
dominantly detected in the patient’s lung and salivary
gland (fig. 3). The element content of tissue from the
lung magnification ×625

Silicon and aluminium in the patient’s gland, particu-
larly in the outer area (fig. 2). Low magnification EDXA
detected in the patient’s salivary gland or those of the
mask material. No particles were

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, particu-
larly in the outer area (fig. 2). Low magnification EDXA
showed silicon and aluminium to be the elements pre-
dominantly 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 pre-
ferred in table 1. Si/S and Al/S values are clearly high-
er in the gland of the patient than in controls, whilst

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

<table>
<thead>
<tr>
<th>Element/sulphur ratio</th>
<th>Patient</th>
<th>Controls</th>
</tr>
</thead>
<tbody>
<tr>
<td>Si/S</td>
<td>0.612</td>
<td>0.193</td>
</tr>
<tr>
<td>Al/S</td>
<td>0.402</td>
<td>0</td>
</tr>
<tr>
<td>Ca/S</td>
<td>0.880</td>
<td>0.814</td>
</tr>
</tbody>
</table>

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

Discussion

In dental technicians, pneumoconiosis is related to sil-
lica and other minerals. Noncomplicated silicosis and in-
terstitial 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 pre-
viously been described in dental technicians. Acrylic
resins have been related to alveolitis and interstitial pneu-
monia in a dental sand-blower [9], but in the present
case the histological and microanalytical data fail to sup-
port the aetiological implication of acrylic resins.

Sjögren’s syndrome, associated with other collageno-
pathies, 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 auto-
immunity, 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 col-
gennoopathies 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 con-
sidered. 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 min-
eral dust [13], though not in salivary glands.

In the present patient, the negative immunological
tests and the finding of a high glandular content of sil-
icon and aluminium suggest that the glandular involve-
ment 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 stim-
ulate 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 inor-
ganic dust.

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

References

1. Strand V, Talal N. Advances in the diagnosis and con-
cept of Sjögren’s syndrome (autoimmune exocrinopa-
WN, ed. Textbook of Rheumatology. Third edition. Phila-
4. Boekestein A, Stols ALH, Stadhouders AM. Quantitation
in X-ray microanalysis of biological bulk specimens. SEM
5. Funahashi A, Siegemund KA, Dragen RF, Pintar K. Energy
dispersive X-ray analysis in the study of pneu-
X. Energy-dispersive X-ray analysis and scanning


