



## Original Article

## Multiple calcifications within the parotid gland of patients with Sjögren's syndrome

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## ABSTRACT

**Purpose:** The purpose of this study was to investigate computed tomography (CT) and clinical features relating to calcifications within the parotid gland of patients with Sjögren's syndrome (SS).

**Methods:** Data from 30 patients with SS who had been examined by CT were extracted from our radiological information database accumulated from 2001 to 2011, and their CT images were reread carefully. Of these patients, 14 (all female; age range 20–95 years; mean age 61.4 years) with calcifications within the parotid gland were retrospectively investigated with CT findings. The relationship between calcification occurrence and clinical symptoms including parotid swelling and/or saliva colic was investigated. The degree of destruction of the parotid gland on CT images was also evaluated.

**Results:** All calcifications of 14 patients were located within the parotid gland, not in the parotid duct. CT images of all calcifications showed small and regular round shapes. Multiple occurrences of calcifications were recognized in 10 patients, and a solitary occurrence was seen in 4 patients. Seven patients had bilateral calcifications. There was little relationship between the occurrence of calcifications and clinical symptoms, and the severity of destruction of the parotid gland.

**Conclusion:** The presented CT and clinical features would be peculiar to SS because too many patients lacked the typical features of sialoliths within the parotid gland.

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### 1. Introduction

Sjögren's syndrome (SS) is a multi-system autoimmune disorder mainly targeting the salivary and lacrimal glands [1–4]. Clinical symptoms are characterized by progressive dry mouth and dry eyes [2–4]. In recent years, bilateral and multiple small calcifications in the parotid parenchyma have been reported as a new feature of SS [5–8]. The occurrence of small calcifications was considered to be in the severely destroyed parotid parenchyma of SS [8] and to be extremely rare in the whole SS population [6,7]. To our knowledge, there had been less than 20 cases in the previous numerous studies of SS [5–8]. Therefore, characteristics of small calcifications were not sufficiently investigated.

Several imaging modalities, such as plain X-ray examination [8,9], sialography, ultrasound, and computed tomography (CT) [5,10], are available for detection of calcifications in the parotid gland. CT is the most useful tool in detection and evaluation of small calcifications because of its high spatial resolution [11,12]. CT can

also depict the destroyed parotid parenchyma, which was replaced by fat, due to autoimmune reactions of SS [8,13]. We planned a retrospective CT investigation to clarify some characteristics of small calcifications. The purpose of this study was to investigate CT and clinical features relating to calcifications within the parotid gland of patients with SS.

### 2. Patients and methods

We surveyed patients with SS, who had been examined by CT, from a radiological information database accumulated from 2001 to 2011. Data from 38 patients with SS were extracted. The purposes of CT examination were for diagnosis of inflammation, cysts, tumors, and trigeminal neuralgia. Patients had consented to CT examination and its study application. CT images were reread carefully to confirm the presence of calcifications in the parotid gland by three radiologists. Clinical information was simultaneously investigated from medical records and letters of introduction from previous doctors, in particular past examinations for definite diagnosis of SS, complications of other autoimmune diseases, a history of parotid swelling, and/or saliva colic. If clinical information was not enough, we interviewed the patients again if possible. All patients satisfied the revised Japanese diagnostic criteria for SS of 1999. Of

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**Fig. 1.** The severity of destruction of the parotid gland parenchyma in patients with Sjögren's syndrome. (A) Slight destruction; Slight fat deposits are seen within the parotid gland, and most parenchyma remain. (B) Moderate destruction; Moderate fat deposits are seen within the parotid gland, and parenchyma remain to a degree between slight and severe. (C) Severe destruction; Severe fat deposits are seen within the parotid gland, and most parenchyma disappear.

38 patients, 8 patients were excluded because 5 had sialographic examination before CT examination and 3 patients had insufficient clinical information for this study. Of the remaining 30 patients, 14 (all female; age range 20–95 years; mean age 61.4 years) had calcifications in the parotid gland on CT images.

CT examination was performed with a single slice scanner (Somatom ART: Siemens Medical Systems, Erlangen, Germany), a 2-detector scanner (HiSpeed NX/I Pro: GE Yokogawa Medical Systems, Tokyo, Japan), or a 4-detector scanner (Asteion: Toshiba Medical Systems Corporation, Tokyo, Japan). Scan conditions were 120 kV and 100 mA. Transverse images with 3 mm or 2 mm thickness were continuously acquired with a scan direction parallel to the occlusal or mandibular plane.

CT images of 14 patients with calcifications were investigated for location, distribution, size, and shape of calcifications. The severity of the destroyed parotid parenchyma on CT images was also evaluated based on a consensus of two experienced radiologists. We presumed the destroyed parotid parenchyma, which results from autoimmune reactions of SS, from the extent of fat deposition

in the parotid gland (Fig. 1). Whether these CT findings and clinical features had any characteristics in 14 patients were investigated.

### 3. Results

CT findings and clinical features of the 14 patients are summarized in Table 1. There were many characteristics differing from the typical features of sialoliths.

All calcifications of the 14 patients were located within the parotid gland, not in the parotid duct. CT images of all calcifications showed that the size did not exceed 2 mm and the shape was regular and round. Multiple occurrences of calcifications were observed in 10 patients, and bilateral occurrence was seen in 7 patients.

As to the relationship between the occurrence of calcifications and the severity of the destroyed parotid parenchyma, slight destruction amounted to about 60%. There were a few patients with a history of parotid swelling and/or saliva colic (21%, 3/14) and with complications of other autoimmune diseases (14%, 2/14).

**Table 1**  
Computed tomography findings and clinical features of 14 patients with Sjögren's syndrome.

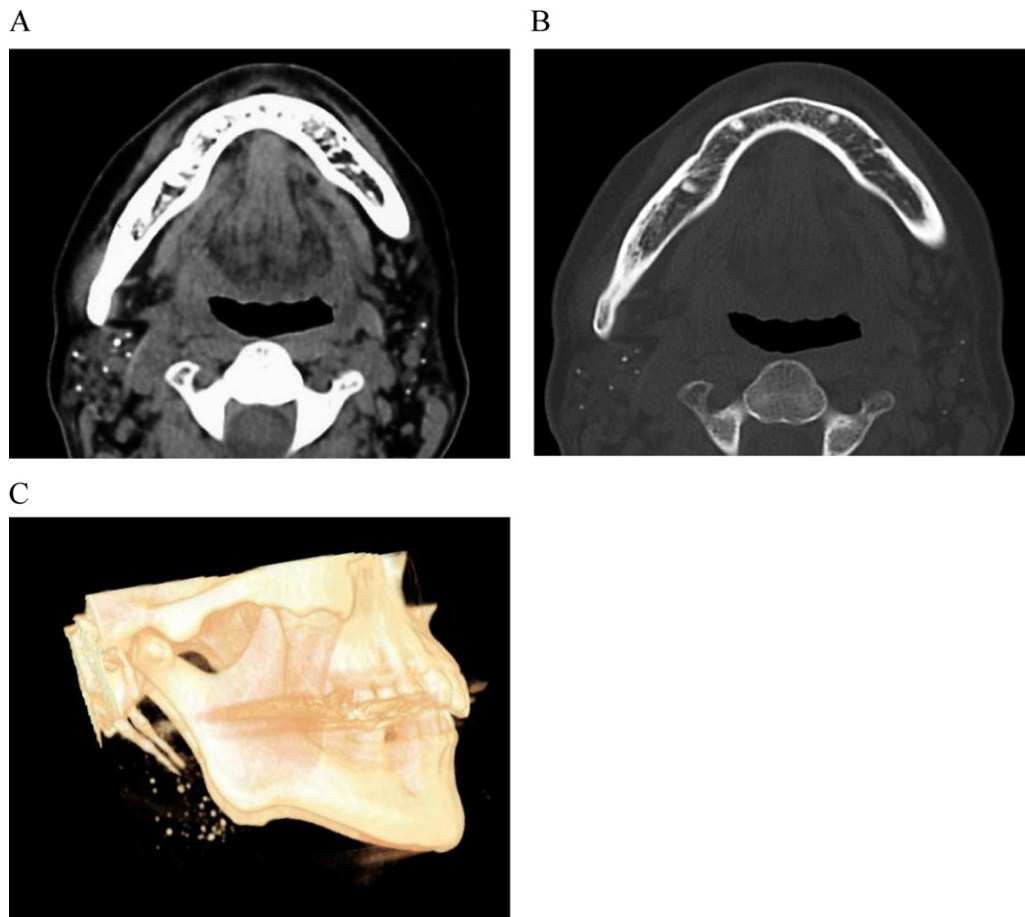
Patient number	Age (years)	Calcifications				Severity of the destroyed PG	History of PG swelling and/or complications pain	Complications
		Location	Distribution	Size	Shape			
1	91	Within Lt. PG	Multiple	<2 mm	Regular round	Moderate	–	–
2	62	Within Lt. PG	Multiple	<2 mm	Regular round	Slight	–	–
3	55	Within Bil. PG	Multiple	<2 mm	Regular round	Moderate	–	–
4	80	Within Bil. PG	Multiple	<2 mm	Regular round	Severe	–	RA
5	58	Within Bil. PG	Multiple	<2 mm	Regular round	Slight	–	–
6	20	Within Lt. PG	Multiple	<2 mm	Regular round	Slight	–	–
7	42	Within Rt. PG	Multiple	<2 mm	Regular round	Slight	–	–
8	68	Within Bil. PG	Multiple	<2 mm	Regular round	Slight	–	–
9	71	Within Bil. PG	Multiple	<2 mm	Regular round	Slight	+	–
10	41	Within Bil. PG	Multiple	<2 mm	Regular round	Slight	+	–
11	95	Within Lt. PG	Solitary	<2 mm	Regular round	Moderate	–	RA
12	40	Within Rt. PG	Solitary	<2 mm	Regular round	Moderate	–	–
13	55	Within Rt. PG	Solitary	<2 mm	Regular round	Slight	–	–
14	64	Within Rt. PG	Solitary	<2 mm	Regular round	Severe	+	–

Rt, right; Lt, left; Bil, bilateral; PG, parotid gland; –, None; RA, rheumatoid arthritis.

Fig. 2 shows typical multiple calcifications within the bilateral parotid gland. Many small calcifications were scattered throughout the whole of the parotid gland. Fig. 3 shows an atypical case of calcifications where isolated calcification was observed within the right parotid gland.

#### 4. Discussion

SS is a multi-system autoimmune disorder that causes abnormalities in many organs and tissues. The occurrence of calcifications in SS has been comparatively known as one of the extra-glandular



**Fig. 2.** Computed tomography (CT) images of a 55-year-old woman with Sjögren's syndrome (Case No. 3). (A) Axial CT image for soft tissue window shows a typical finding of bilateral multiple calcifications in the parotid gland. (B) Axial CT image for bone window of the same slice. (C) Three dimensionally reconstructed CT image clearly shows scattered calcifications in the right parotid gland.



**Fig. 3.** Computed tomography (CT) images of a 40-year-old woman with Sjögren's syndrome (Case No. 12). Axial CT image for soft tissue window shows an atypical finding of solitary calcification in the right parotid gland (arrow).

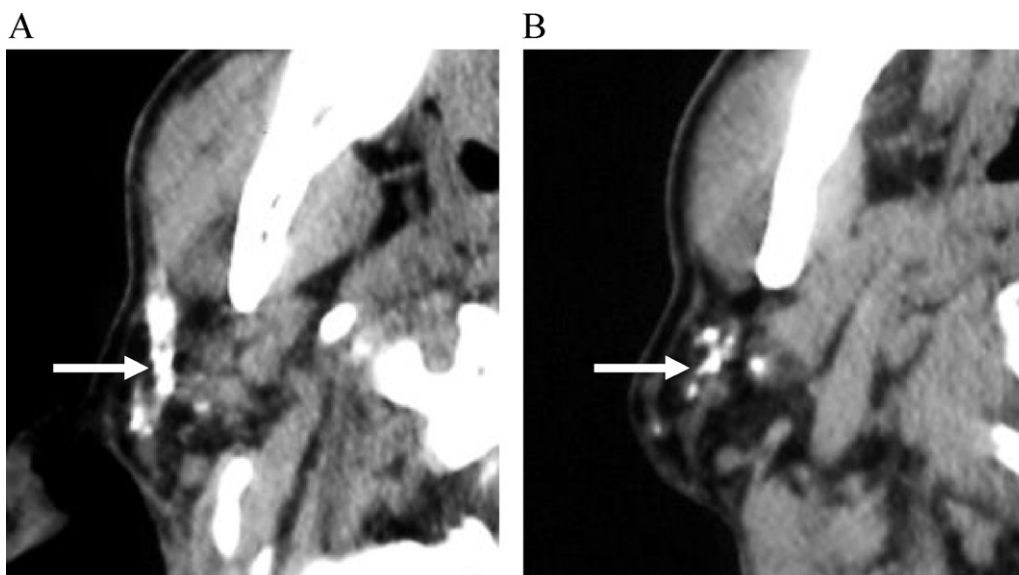
manifestations, such as nephrolithiasis and urolithiasis [14–16]. Distal renal tubular acidosis is considered as a risk factor for development of calcifications [14–16].

In contrast, the calcifications within the bilateral parotid gland of SS were not well known because of their rare occurrence in SS [6,7]. However, considering the incidence of our investigation, calcifications within the bilateral parotid gland appear to be a frequent occurrence (23%, 7/30). Sun et al. also reported that the incidence of calcifications within the bilateral parotid gland of SS was 29.4–35.2% in their CT investigation [8]. A reason for the contradiction between rare and frequent occurrence may be that CT examination is seldom done in patients with SS. Most radiological examinations to SS are scintigraphy and/or sialography, which are inferior to detect small calcifications, for definite diagnosis of SS. If CT examination is performed in higher frequency, the potential existence of calcifications within the parotid gland may be revealed in many patients with SS.

Most previous reports dealt with these calcifications as sialoliths [5–7]. However, in this investigation, a lack of CT and clinical features possibly relating to sialolith occurrence was observed. Ordinary sialoliths occur at an incidence rate of about 1% in the population [17], and 6–20% of the whole sialoliths occur in the parotid gland [10]. In our SS series, the incidence rate of calcifications within the parotid gland was 46% (14/30). Furthermore, although sialoliths are predominantly found in the duct, all calcifications in our cases were found within the gland. The shape of ordinary sialoliths is oblong and is often pointed and sharp [17]. These calcifications were regular and round shapes. Commonly, sialolithiasis presents with painful swelling (59%), painless swelling (29%), and pain only (12%) [10]. Clinical symptoms in our calcification cases were few (21%, 3/14). Although lower salivary flow rate is considered to be associated with sialolith occurrence [10], the severity of the destructed parotid parenchyma was not related to the occurrence of calcifications in our SS series.

Rauch and Gorlin described in the textbook [17] of oral pathology that “The ‘calculus’ of the salivary gland is etiologically and clinically as a relatively heterogeneous term. Apart from sialolith, there are dysplastic calcifications in inflammatory salivary gland tissues and calcified venous thrombi.” As they described, some calcifications in this investigation may be dysplastic calcifications of the salivary gland tissues secondary to chronic inflammation of SS. And CT findings of these calcifications were very similar to the characteristics of angioliths [18] in the parotid gland. Regarding the calculus of the salivary gland, we should keep in mind the existence of calcifications except for sialolith in the duct.

We do not negate a notion of previous reports that all calcifications in SS cases are sialoliths, because of no clear or enough evidence to negate this notion at present. And this investigation has the following limitations: a small number of cases, case bias, and non-analysis of the components of calcifications. Some contradictions between ordinary sialoliths and these calcifications may be a study error. But it is difficult to consider from our overall results that these calcifications occurred from just the same mechanism as ordinal sialoliths in the ducts. Further research by pathophysiological and biochemical approaches is expected to reveal this issue in the future.



**Fig. 4.** Computed tomography (CT) images of a 57-year-old man with Sjögren's syndrome. He had 7 years history of sialographic examination of the right parotid gland before CT examination. (A) High-density structures in the ducts are considered to be the residue of contrast medium (arrow). (B) On the inferior slice, high-density spots cannot be distinguished from calcifications (arrow).

For the evaluation of calcifications on CT images, the history of sialographic examination should be noted. Contrast medium, especially oleaginous, frequently remains in the duct system even in patients without SS [19]. Fig. 4 shows CT images of a patient with SS more than 7 years after sialographic examination. Residual contrast media showing a spot-like appearance occasionally cannot be distinguished from calcifications.

In conclusion, we presented CT and clinical characteristics of high frequency calcifications within the parotid gland in patients with SS. These calcifications would be peculiar to SS, and may differ from ordinary sialoliths in the mechanism of occurrence because too many cases lacked typical features of ordinary sialoliths.

### Conflict of interest statement

None of the authors has any conflict of interest regarding this research.

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