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## Clinical Characteristics of Parotid Gland Sarcoidosis: A Population Based Study

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### Introduction

Parotid gland involvement is a well-recognized extra-thoracic feature of sarcoidosis. However, the data on epidemiology and clinical characteristics of parotid gland disease in sarcoidosis are limited. The objective of this study was to describe the epidemiology of parotid gland involvement of sarcoidosis, with an emphasis on clinical characteristics, in a geographically well-defined population.

### Methods

Approval for this study was obtained from the Mayo Clinic and Olmsted Medical Center institutional review boards and the need for informed consent was waived. A cohort of Olmsted County, Minnesota residents diagnosed with sarcoidosis between January 1, 1976 and December 31, 2013 was identified using the resources of the Rochester Epidemiology Project.<sup>1</sup> Potential cases were screened from diagnostic codes related to sarcoidosis. Diagnosis of sarcoidosis and parotid gland involvement were then confirmed by individual medical record review. Cases included were cases of pulmonary sarcoidosis with parotid gland involvement and cases of isolated parotid gland sarcoidosis without intra-thoracic

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#### Author contribution:

**Patompong Ungprasert:** 1. Conception and design 2. Acquisition and interpretation of data 3. Drafting of the manuscript 4. Statistical analysis

**Cynthia S. Crowson:** 1. Conception and design 2. Analysis and interpretation of data 3. Critical revision of the manuscript for important intellectual content 4. Statistical analysis

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Patompong Ungprasert and Cynthia S. Crowson had full access to all of the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis.

disease. Diagnosis of pulmonary sarcoidosis required physician diagnosis supported by histopathologic evidence of non-caseating granuloma and/or radiographic findings of intra-thoracic sarcoidosis and compatible clinical presentations, without evidence of other granulomatous diseases. The only exception was stage I pulmonary sarcoidosis that required only radiographic evidence of symmetric bilateral hilar adenopathy. Diagnosis of parotid gland involvement required signs and symptoms of parotid gland infiltration such as parotid gland enlargement. Biopsy confirmation was not required if the patient had pulmonary sarcoidosis. If the patient did not have pulmonary involvement, parotid gland biopsy with demonstration of non-caseating granuloma was required. In these cases without intra-thoracic involvement, biopsy-proven isolated granulomatous disease of other specific organs except for the skin were also included if there was no better alternative diagnosis. Cases with a diagnosis of sarcoidosis prior residency in Olmsted County were excluded.

## Results

345 incident cases of sarcoidosis were identified. Of those, only 7 patients had parotid gland involvement (mean age 44 years; 43% female, 86% Caucasian and 14% African-American). The majority of patients had parotid gland disease in association with intra-thoracic sarcoidosis as isolated parotid sarcoidosis was observed in only one case. Parotid gland disease was usually painless and unilateral and was the initial presentation in 4 patients. Angiotensin-converting enzyme (ACE) level was elevated in only 25% of patients while none had hypercalcemia. Gland swelling regressed after steroid treatment in all patients though one relapse was seen. Table 1 describes the clinical characteristics of these patients.

## Discussion

Only 2% of patients with sarcoidosis in this population-based cohort developed parotid gland involvement, a frequency considerably lower than previous reports of 5–30%.<sup>2–4</sup> The difference could be due to the difference in ethnic background of the cohorts, as this cohort was predominantly Caucasian.<sup>5</sup> Another possible explanation was related to the study design as this study was a population-based study the might capture a more complete spectrum of the disease, in contrast to referral-based cohort design utilized in previous studies.<sup>2–4</sup>

The most common presentation of parotid gland disease was unilateral painless gland swelling which was also different from the previous studies that found bilateral involvement in more than 70% of their cohorts.<sup>3, 4</sup> More importantly, parotid gland disease was often the initial manifestation, highlighting the importance of the otolaryngologic assessment in the diagnosis of systemic sarcoidosis.

The demographics of patients with parotid gland involvement were similar to the complete cohort of 345 patients (mean age 45.6 years, 50% female and 95% Caucasian). ACE level was elevated in 3 out of 6 tested patients which was also not significantly different from the complete cohort (41%).

In conclusion, the prevalence of parotid gland involvement was lower than previously reported. Prognosis was favorable.

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**Table 1**

Patients with sarcoidosis with parotid gland involvement

Age and race	Presentations of parotid gland disease	First presentation of sarcoidosis	Biopsy	Chest imaging	Other extra-thoracic involvement	ACE level	Serum Calcium	Treatment	Response to treatment
Female/White	Painless bilateral parotid gland swelling <i>JAMA Otolaryngol Head Neck Surg. Author manuscript; available in PMC 2017 May 01.</i>	Yes	Performed; non-caseating granuloma	Bilateral hilar adenopathy and pulmonary infiltration	None	Normal	Normal	Oral prednisone	Parotid gland regressed to normal size
Female/White	Painless unilateral parotid gland swelling and xerostomia	No	Performed; non-specific inflammation	Bilateral hilar adenopathy	Eyes and liver	Normal	Normal	Oral prednisone	Parotid gland regressed to normal size
Male/White	Painless unilateral parotid gland swelling	Yes	Not performed	Bilateral hilar adenopathy	Erythema nodosum and liver	Elevated	Normal	Oral prednisone	Parotid gland regressed to normal size
Female/Black	Painless unilateral parotid gland swelling	No	Not performed	Bilateral hilar adenopathy and pulmonary infiltration	Skin rash and lacrimal gland	Elevated	Normal	Oral prednisone	Parotid gland regressed to normal size
Male/White	Painless unilateral parotid gland swelling and numbness over the swollen area	Yes	Performed; non-caseating granuloma	Bilateral hilar adenopathy	None	Normal	Normal	Oral prednisone	Parotid gland regressed to normal size. Relapse once, also responded to prednisone
Female/White	Painless unilateral parotid gland swelling	No	Performed; non-caseating granuloma	Normal	None	Elevated	Normal	NSAIDs and oral prednisone	Did not respond to NSAIDs but parotid gland regressed to normal size after switching to prednisone
Male/White	Painless unilateral parotid gland swelling	Yes	Performed; non-caseating granuloma	Fibrosis in lung parenchyma with minimal Bilateral hilar adenopathy	Skin rash	Not performed	Not performed	Oral prednisone	Parotid gland regressed to normal size

NSAIDs = non-steroidal anti-inflammatory drug