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Case Report

A case of IgG4-related disease founded from sclerosing sialadenitis



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ABSTRACT

IgG4-related disease, a new disease entity proposed in Japan, is characterized by the chronic inflammation of various organs, enlargement of affected organs, and reversible hypoadenia. Here, a case of a condition suspected to be IgG4-related sclerosing sialadenitis is reported.

A 58-year-old woman consulted us with a primary symptom of bilateral, painless, and submandibular swelling. While salivary gland MRI revealed the symmetric enlargement of the lacrimal and salivary glands, no mass formation was noted. Also, while autoantibody was negative, a diagnosis of IgG4-related disease was made because of a high serum IgG4 level and fibrosis accompanied by IgG4-positive plasma cell infiltration demonstrated by sublingual gland biopsy. Subsequent examinations of the whole body also disclosed autoimmune pancreatitis. Enlargement of the salivary glands rapidly reversed, and pancreatomegaly also gradually regressed, after the beginning of steroid therapy.

Since IgG4-related disease is systemic, it is necessary to examine various organs and prevent the progression of their impairment. This disease remains unclear in many respects, and multidisciplinary cooperation is considered important.

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1. Introduction

IgG4-related disease, which was reported in Japan as pancreatitis with a high serum IgG4 level and tissue infiltration of IgG4-positive cells [1], is presently considered a systemic disorder as cases with generalized extrapancreatic lesions have been accumulated. Among salivary diseases, chronic sclerosing sialadenitis such as Mikulicz's disease and Kuttner's tumor has been reported to show a high serum IgG4 level and to be histologically characterized by IgG4-positive cell infiltration. Mikulicz's disease, which is generally included in Sjögren's syndrome, has now been proposed as an independent and new IgG4-related disease entity [2–4].

While IgG4-related disorders of various organs are essentially benign, they may have been mistaken for malignancies of organs or blood and subjected to extended surgery or anticancer chemotherapy. However, they respond markedly to steroid therapy. Here, we present a case of IgG4-related disease that showed symmetric swelling of the salivary glands and pancreatomegaly and was diagnosed by sublingual gland biopsy.

2. Case report

The patient was a 58-year-old woman. She first noted swelling of the bilateral lower jaw regions around February 2013. While there was no fever, pain, or progressive enlargement, she consulted a local dentist, was recommended to undergo close examination and treatment, and visited Fukui Prefectural Hospital. Her clinical history included hypertension and osteoarthritis of the knee, but there was no particular familial history.

The face was symmetric, and the palpebral conjunctivae were normal. Despite swelling of the bilateral parotid and submandibular regions, no tenderness was noted (Fig. 1A and B). Also, there was no enlargement or tenderness of the cervical lymph nodes.

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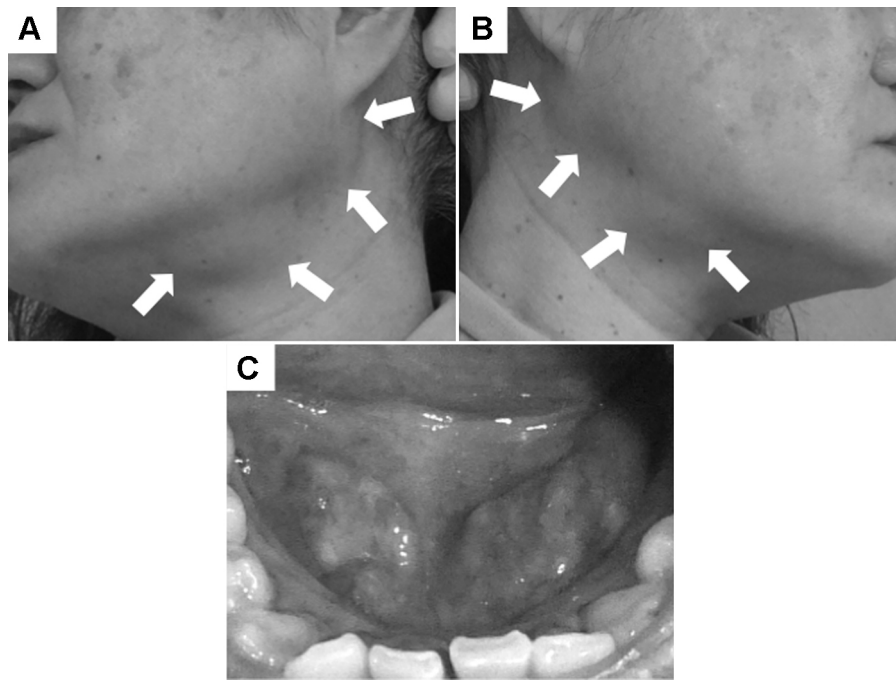


Fig. 1. (A and B) Elastic, hard, movable, and painless swelling was noted in the bilateral parotid and submandibular regions. (C) Painless swelling was noted in the bilateral sublingual regions.

While both the submandibular and sublingual glands were enlarged, no adhesion was observed, and in addition, all these glands were elastic hard (Fig. 1C). No paralysis of the lingual, hypoglossal, or glossopharyngeal nerve was observed.

Salivary gland MRI revealed symmetric enlargement of the bilateral lacrimal, parotid, submandibular, and sublingual glands but no clear mass formation, and the possibility of a neoplastic disease was considered low (Fig. 2).

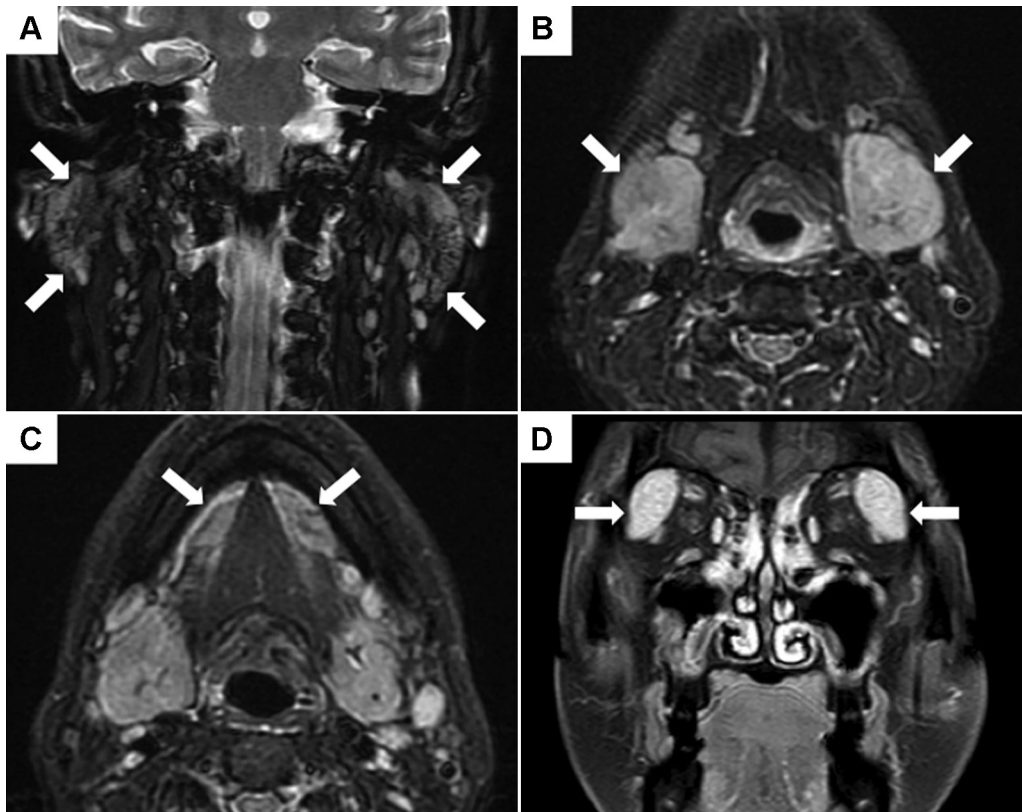


Fig. 2. Contrast-enhanced MRI. Bilateral enlargement of the parotid, submandibular, sublingual, and lacrimal glands was noted. (A) Parotid gland; (B) submandibular gland; (C) sublingual gland; (D) lacrimal gland.

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