



# Development of Frog Neck After Coronary Angiography: Contrast-Induced Sialadenitis

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## ABSTRACT

Iodine-related submandibular sialadenitis is a rare complication among the causes of sialadenitis. To date, the pathogenesis of iodide mumps has not yet been ascertained. This situation is a late reaction to iodine-containing contrast agents and can manifest in any patient receiving iodinated contrast agents and repeated exposure. Fortunately, this condition does not require specific treatment and resolution can be expected within one or two weeks. We report an 82-year-old man suffering from iodide-induced sialadenitis following coronary angiography with the use of non-ionic low osmolar contrast media.

**Key Words:** Contrast; coronary angiography; sialadenitis

## Koroner Anjiyografi Sonrası Gelişen Kurbağa Boynu: Kontrastın İndüklediği Sialadenit

### ÖZET

İyot ilişkili submandibular sialadenit, sialadenit nedenleri içinde, nadir görülen bir durumdur. İyota bağlı gelişen tükürük bezi iltihabının patogenezi henüz tam kesin bilinmemektedir. Bu durum, iyot içeren kontrast ajanlara bağlı gelişen geç bir reaksiyondur ve herhangi bir iyotlu kontrast alan ve tekrarlayan maruziyeti olan bir hastada gelişebilir. Neyse ki, özellikli bir tedavi gerektirmez ve bir iki hafta içinde kendiliğinden iyileşir. Burada, iyonik olmayan düşük osmolar kontrast ajan ile yapılan koroner anjiyografiden sonra sialadenit gelişen 82 yaşındaki erkek hastayı sunuyoruz.

**Anahtar Kelimeler:** Kontrast; koroner anjiyografi; sialadenit

## INTRODUCTION

Sialadenitis of the submandibular gland is an important area of clinical interest for otolaryngologists and other specialists. Contrast-induced sialadenitis is a less-known and rare adverse reaction to iodine therapy. In this case, the symptoms are less aggressive than bacterial sialadenitis and a specific treatment is not required. Sialadenitis due to contrast administration has dose-related damage that can follow an acute inflammatory course. The risk for sialadenitis appears to be directly related to serum iodine levels, therefore, renal failure and large iodine load are predisposing factors<sup>(1)</sup>. We report a patient with contrast-related sialadenitis as a result of coronary angiography, who was fully recovered after receiving medical treatment. To the best of our knowledge, this case reports the oldest patient presenting with contrast-induced sialadenitis after coronary angiography.

## CASE REPORT

An 82-year-old man presented with upper abdominal pain and unstable angina pectoris. He was symptomatic for shortness of breath on moderate exertion. He had hypertension, hyperlipidaemia, chronic obstructive pulmonary disease and had undergone a radical prostatectomy to treat prostate cancer.

Coronary angiography was performed using Iohexol (Omnipaque 350, GE Healthcare). A non-ionic, monomeric, low-osmolality contrast medium containing approximately 350 mg/mL iodide was used. A total of 60 mL of the above-mentioned iodinated contrast media was used. A coronary angiogram showed normal coronary arteries, except the proximal left anterior descending artery, which showed 20% narrowing. Thus, the patient was treated medically. About 24 hours after the procedure, the patient complained of bilateral swelling on his upper and front neck. In the physical examination, his blood pressure was 120/80 mmHg, heart rate was 88 bpm and body temperature was 36.8°C. The submandibular gland was enlarged, palpable and

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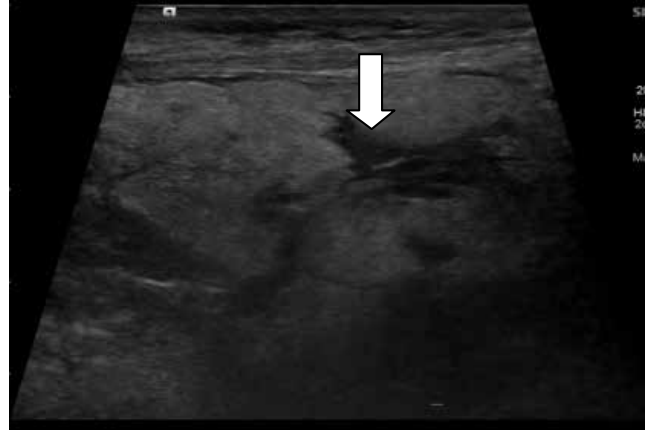


**Figure 1.** Bilateral swelling of submandibular glands at onset.



**Figure 2.** Complete improvement in submandibular gland swelling after six days.

showed no purulent draining in any of the ductal openings. The patient had a non-sensitive, extensive, bilateral and symmetrical, submandibular swelling (Figure 1). The patient was afebrile and there was no enlargement of the parotid gland. The respiratory system examination was normal and there was no observed



**Figure 3.** Ultrasonography of submandibular glands showing oedema (white arrow).

dysfunction in the swallowing function. Clinical presentation of contrast-induced sialadenitis ('iodide mumps') was suspected.

Blood tests revealed a white blood cell count of  $7.02 \times 10^9$ , haemoglobin of 12.5 g/L, serum creatine level of 1.35 mg/dL, GFR according to MDRD as 53 mL/min/1.73 m<sup>2</sup> (mild-moderate chronic kidney disease, Stage 3A), thyroid stimulating hormone at a level of 1.84 mIU/L, amylase at 149 UI/L (normal level), a normal complete blood count and CRP as 16.07 mg/L. Neck ultrasonography showed that the submandibular gland had a significantly longer diameter and thickness. In addition, the gland contour was rough, with increased echogenicity and oedema in the subcutaneous soft tissue (Figure 3). The structure and size of the thyroid and parathyroid gland were normal.

Spiramycin 3 Miu twice a day and prednisolone 80 mg/day were recommended by an otolaryngologist. After six days, the submandibular swelling completely resolved, the serum CRP level decreased to 0.7 mg/dL and the patient's follow-up was uncomplicated (Figure 2).

## DISCUSSION

Aseptic sialadenitis was first described by Sussman and Miller in 1956, and on viewing this on urography, it was named 'iodide mumps'<sup>(2)</sup>. More than 40 cases of iodised mumps have been reported in English scientific literature in the past 60 years.

However, the mechanism of how contrast-induced sialadenitis develops after the use of either non-ionic dimers or non-ionic monomers is still unclear. Iodine-induced sialadenitis is thought to be an ido-synchronous or toxic process, which damages the salivary glands, causes oedema and eventually obstructs the ducts and leads to gland inflammation. The redistribution half-life of the intravascular compartment of contrast agents is fast (2-5 minutes for most agents). In renal insufficiency, the elimination of most contrast agents is delayed<sup>(3)</sup>. Iodinated contrast agents (ICA) are secreted in the first hour, with about 40% of the kidneys remain-

ing mostly unchanged (about 80%-90%). The remainder of ICAs are eliminated within the following hours. If iodine is present at high plasma concentrations (> 10 mg/100 mL), it can be resolved by alternate routes, such as iodine salivary glands<sup>(4,5)</sup>. Consequently, renal insufficiency is a substantial risk factor for sialadenitis. Our patient had mild-moderate renal failure, which exposed his increased iodine load. On the other hand, most cases of ICA management-complicated sialoadenitis have been described in patients with normal kidney function<sup>(6)</sup>.

The specific cause of contrast-induced sialadenitis is unknown, however, it is thought that many factors that contribute to sialadenitis are associated with the use of ICAs. The idiocentric reaction is another cause of sialadenitis that occurs in patients who are repeatedly exposed to ICAs. Iodide mumps can occur after the application of any type of ICA, including both ionic and non-ionic media. In patients receiving an ionic contrast agent, the frequency of development of 'iodide mumps' is greater than in patients receiving a non-ionic contrast agent. When a retrospective analysis of the contrast-induced sialadenitis was performed, 18 of 36 patients received ionic contrast agents, 15 received non-ionic contrast agents and 3 had no media type. The submandibular and parotid glands were involved in 20 of the previous 36 previous cases, 8 patients had pain in the salivary glands and 1 patient developed facial paralysis<sup>(7)</sup>.

While evaluating sialadenitis, ultrasonography can be used to distinguish between solid versus cystic lesions of the gland. Computed tomography (CT) scanning is the ideal modality for recognising and differentiating between intrinsic versus extrinsic glandular disease. Magnetic resonance imaging is effective for appliance for soft tissue imaging and is used in instances of suspected neoplasia. In our case, as ultrasonography is fast and easily accessible, we thought it was appropriate to perform ultrasonography in the first step.

The differential diagnosis of submandibular sialadenitis also includes infectious, inflammatory and autoimmune diseases, along with a number of granulomatous, drug-related, neoplastic, endocrine and metabolic causes.

The management of submandibular sialadenitis involves a wide range of approaches, from conservative medical management to aggressive surgical intervention. However, the treatment of sialadenitis should be directed towards managing the underlying

problem and achieving homeostasis. The goals of pharmacotherapy are to eradicate the infection, reduce morbidity and prevent complications. For patients with acute sialadenitis that does not require hospital admission, a follow-up visit should be scheduled three days from the first visit with another visit scheduled one week later (with improvement). Iodide mumps is usually a self-limiting condition that does not require any intervention.

To determine the progression or improvement of acute sialadenitis, serial USG or CT scanning may be needed. Acute symptoms resolve within one week, however, oedema in the area may last several weeks. Patients with sialadenitis have a good prognosis if their underlying problem is adequately controlled; however, even in this case, bilateral swelling may be persistent.

## CONCLUSION

Sialadenitis is a rare and late adverse reaction to iodine contrast agents. Any iodinated contrast medium, be it non-ionic or hypo-hyperosmolar, may cause sialadenitis. Identifying patients who are at risk of adverse reactions and minimising this risk by following preventive measures can reduce the incidence of adverse effects. Our aim was to present this rare complication and to draw attention to its occurrence in patients with throat and neck swelling that commonly occur after angiography. We also aimed to share the diagnosis, treatment options and prognosis of such patients.

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