Acute sialadenitis to a gadolinium contrast media

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Iodinated contrast media sialadenitis, or "iodide mumps", is a rare non-allergic adverse reaction that has been described in the literature with several iodinated contrast media. This impressive reaction is most often mistaken for allergic angioedema. However, it is a mild reaction and resolves spontaneously within several days and no severe cases have been reported [1]. We report a case of sialadenitis following gadoteric acid-injected magnetic resonance imaging (MRI).

24 hours after an MRI using gadoteric acid (Dotarem®) to investigate chronic diarrhea, a 37-year-old woman presented with significant bilateral parotid oedema (Figure 1). She did not have any medical history, but a cholecystectomy. Her sole treatment was some diosmectite (Smecta®), for her diarrhea. Given the extent of the parotid and cervical oedema, the patient was hospitalized in a continuing care unit after her visit to the emergency room. The examination showed cervical oedema and enlargement of the main salivary glands. There was not any associated skin reaction, hemodynamic instability, dyspnea or dysphagia or intraoral oedema. She received 130mg of IV methylprednisolone and 100mg of Hydroxyzine, then was discharged home after 24 hours of monitoring. Resolution ad integrum occurred slowly over 7 days despite the fact she continued her treatment at home with corticosteroids and antihistamines. Diosmectite was continued during the hospital care and after, without relapse.

The CT scan showed asymmetric enlargement of the parotid and submandibular glands with infiltration of the adjacent soft tissues (supplementary figures 1, 2). The work-up for parotitis and angioedema, which included a blood count, ionogram, creatinine level, serum protein electrophoresis, autoimmune work-up, tryptasemia, thyroid function, CRP, and a search for active viral infections with EBV, CMV, HIV, hepatitis C and B, and mumps, was negative. The allergology work-up carried out 3 months after the reaction showed negative skin tests. To perform those tests, we used Dotarem® 0,05mmol/ml (279mg/ml) at the concentrations recommended by the EAACI [2]: undiluted for prick-test, and diluted at 1/10 for IDT. Patch-testing was performed using undiluted Dotarem®. Prick and IDT were read at 20 minutes, followed by delayed readings at 48 and 96 hours for IDT and patch-test.

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This patient was referred to the allergy department with a suspected allergy to Dotarem®. The clinical and imaging findings are consistent with what has been described with iodinated contrast media in cases of "iodide mumps"[1]. Since the patient got better without stopping diosmectite, its imputability is very low, thus, Dotarem® was the only suspected drug.

The clinical picture of this sialadenitis is one of unilateral or symmetrical parotid and/or submandibular swelling, usually painless. Allergology tests are negative in these patients, suggesting a non-allergic mechanism. Some studies suggest a higher incidence in patients with renal failure. A. Jiao & al recently conducted a review of 77 cases of iodide mumps. They did not find any statistically significant increase in the incidence of iodide mumps in patients with renal failure.

Paraclinical investigations in the reported cases showed non-inflammatory oedema of the salivary glands in ultrasound, MRI, and fine needle biopsies. Only 2 cases reported inflammatory changes in the surrounding fat. Our patient presented with sialadenitis.

The biological workup was within normal values and the skin allergy workup was negative as in iodide mumps with iodinated contrast media. In addition, she also showed infiltration of the fat adjacent to the salivary glands and asymmetric enlargement of the parotid glands (46*70*79mm on the left and 62*36*66mm on the right) and of the submandibular glands (27*27*39 mm on the left and 28*27*26 mm on the right). There was no detectable abscess, but there were adenopathies in the jugulocarotid territories up to 8mm in diameter, without necrosis. However, there was no fever, nor any local or systemic clinical inflammatory sign.

She was initially treated with antihistamines and corticosteroids, assuming an immuno-allergic mechanism to her reaction, but without real efficacy. The mechanism of the reaction is still unknown, but for sialadenitis to iodinated contrast media, one of the main pathophysiological hypotheses is that of an idiosyncratic phenomenon linked to direct iodine toxicity on the glandular canaliculi, accumulated in the salivary glands. Sialadenitis has been reproduced by the ingestion of Lugol's in patients with a history of iodide mumps [4].

Recurrences of iodide mumps have been reported with the use of different iodinated contrast media, without modification or worsening of the latter [4,5]. Premedication does not prevent recurrence in the reported cases. Iodide mumps is therefore not an absolute contraindication to the use of these drugs. And replacing the molecule, although recommended, does not guarantee the absence of recurrence. The same might be true for gadolinium contrast media.

For now, rechallenge has not been retained in this young woman without comorbidity. The injection of gadolinium contrast media could be considered, if necessary, under prudent medical supervision.

Adverse event associated with gadolinium-based contrast media are infrequent [6]. More reports are needed to better understand this peculiar reaction, potentially associated with

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gadoteric acid (Dotarem®). Therefore, it is important to us that allergists and radiologists are aware of its existence.

All informations and images are being published with the patient's informed and written consent.

This case illustrates an acute sialadenitis, with an iodide mumps-like presentation, possibly caused by an infusion of gadoteric acid (Dotarem®) the day before the symptoms occurrence. It could be a rare, non allergic, adverse event of this gadolinated contrast media.

Conflict of interest

None of the authors have any financial, or material conflict of interest in this work.

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Patients' consent

The authors received the patients' written consent to use her photographs and some of her medical informations to be used for scientific publication.

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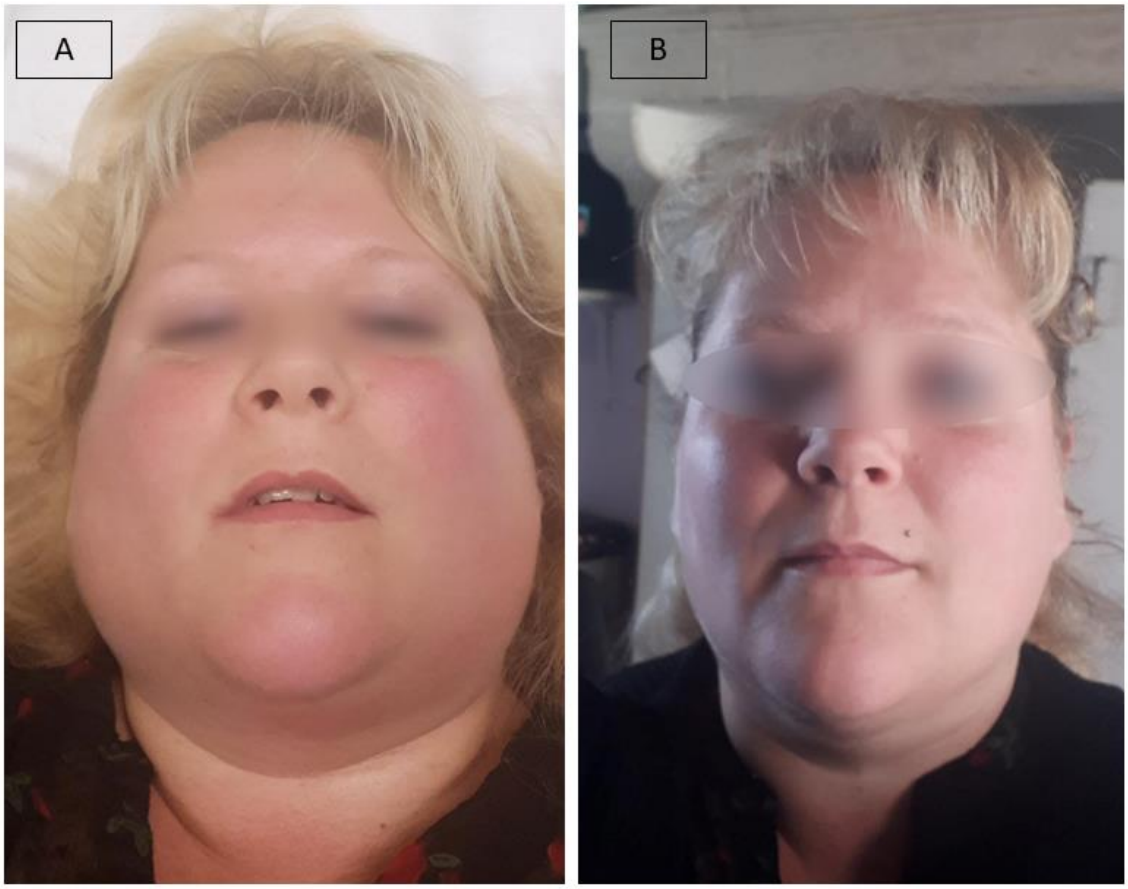

Figure 1 : photographs of the patients face during the reaction (A), and her face 7 days later (B). *These photographs are kindly provided by the patient herself, and they are published with her informed consent.*

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