ACTA MÉDICA PORTUGUESA
A Revista Científica da Ordem dos Médicos

Symmetrical Neck Swelling After Percutaneous Coronary Intervention

Edema Cervical Bilateral Após Intervenção Coronária Percutânea

Liliana PEREIRA DIAS✉[1], Ana Cristina COELHO[1], Cristina MOTA[2], Maria João SOUSA[1]
Acta Med Port 2025 Apr;38(4):260-263 ▪ https://doi.org/10.20344/amp.22423

ABSTRACT

Cervical edema after iodinated contrast media administration can raise the suspicion of a hypersensitivity reaction, but other diagnoses must be considered. The authors present a rare case of a man who, during the diagnostic investigation of a patient with thoracic pain developed severe submandibular swelling and local pain after a percutaneous coronary intervention with iodinated contrast media. The cervical computerized tomography revealed enlargement of the submandibular glands with adjacent fat stranding, suggesting contrast-induced sialadenitis. In the allergology department skin prick tests, intradermal tests and epicutaneous test with suspected iodinated contrast media and alternative were performed, which were negative. Drug provocation tests with alternative iodinated contrast media were negative. Different approaches can be considered when approaching these patients. Our algorithm includes the evaluation of a potential hypersensitivity reaction with a skin test and drug provocation test with alternative iodinated contrast media.
Keywords: Contrast Media; Edema; Iodine/adverse effects; Sialadenitis/chemically induced

RESUMO

A reação de hipersensibilidade deve ser considerada perante o desenvolvimento de edema cervical após a administração de um meio de contraste iodado. Contudo, devem ser avaliados outros diagnósticos diferenciais. Os autores apresentam um caso raro de um homem, que durante a realização de uma intervenção coronária percutânea com meio de contraste iodado para estudo de dor torácica, desenvolveu um edema e dor submandibular bilateral. A tomografia computadorizada cervical revelou aumento das glândulas submandibulares com densificação da gordura adjacente, sugestivo de sialadenite induzida por contraste. No serviço de imunoalergologia, foram realizados testes cutâneos por picada, intradérmicos e epicutâneos com o meio de contraste iodado suspeito e alternativo, que foram negativos. A prova de provocação com meio de contraste iodado alternativo foi negativa. Assim, podem ser consideradas diferentes abordagens no estudo destes doentes. O nosso algoritmo incluiu avaliação de uma potencial reação de hipersensibilidade com testes cutâneos e prova de provocação com meio de contraste iodado alternativo.
Palavras-chave: Edema; Iodo/efeitos adversos; Meio de Contraste/efeitos adversos; Sialadenite/induzida quimicamente

INTRODUCTION

Iodinated contrast media (ICM) were first implemented in clinical practice in the early 1900s for diagnostic purposes and disease surveillance.[1] Even though adverse reactions to ICM are relatively frequent they are typically mild and can arise from either toxicity or hypersensitivity responses.[2] The frequency of immediate and non-immediate hypersensitivity reactions is described in approximately 0.5% - 3.0% of the exposed patients.[3]

Cervical swelling following the administration of ICM may suggest a potential hypersensitivity reaction, but other diagnoses must be considered: ductal obstruction of salivary glands, trauma, acute infection, neoplasia or contrast-induced sialadenitis (CIS).[4]

Contrast-induced sialadenitis was first described in 1956 by Miller and Sussman, who coined the term 'iodide mumps', likening the facial glandular swelling to viral parotitis.[5] It remains an uncommon reaction to ICM and fewer than 80 cases have been reported to date.[5,6] In the present case, sialadenitis developed from one to 24 hours after administration of ICM and took four to 84 hours to resolve.[4,7] Clinical manifestations were characterized by swelling of the salivary glands and pain.[7] The etiology is not fully known, but may be due to high concentrations of ICM in the salivary glands, causing local inflammatory edema, which leads to the obstruction of the salivary duct.[7] The study by Lee *et al* identified the main risk factors that contribute to the development of CIS after neurovascular procedures, finding higher incidence in the female sex, the volume and type of contrast agent used and the duration of the procedure.[8]

The diagnosis of CIS is primarily based on clinical observation and is corroborated by ultrasound of the salivary glands.[4] Other imaging techniques such as computed tomography may be considered.[4]

A correct diagnosis is essential. Additional exposure to ICM should be avoided, as it is likely that sialadenitis will recur. However, there are no absolute contraindications if urgent life-saving interventions are necessary.

CASE REPORT

We present the case of a 77-year-old male patient referred for evaluation at the Allergology department due to facial edema associated with the administration of ICM.

The patient had a personal history of hypertension, dyslipidemia, and recurrent episodes of thoracic pain. The usual medication was zofenopril 30 mg, atorvastatin 10 mg and

1. Allergy Department. Unidade Local de Saúde Gaia-Espinho. Vila Nova Gaia. Portugal.
2. Radiology Department. Unidade Local de Saúde Gaia-Espinho. Vila Nova Gaia. Portugal.
✉ **Autor correspondente:** Liliana Pereira Dias. lilianadias2493@gmail.com
Recebido/*Received*: 10/10/2024 - **Aceite/*Accepted*:** 13/12/2024 - **Publicado/*Published*:** 01/04/2025
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tamsulosin 0.4 mg. No history of atopy or allergic disorder, such as food, drug or latex allergy.

During the study of thoracic pain, the patient underwent a percutaneous coronary intervention with iopromide 769 mg/mL (Ultravist®), with a total administered dose of 70 mL. Ten hours later, he developed severe submandibular swelling and local pain, without other symptoms. The patient was observed in the emergency department. There were no changes in the blood count, renal function, liver enzymes and C-reactive protein; the cervical CT (without contrast) revealed an enlargement of the submandibular glands with adjacent fat stranding, suggestive of edematous changes (Fig. 1). The patient was treated with a short course of corticosteroids (40 mg prednisolone) with complete resolution on day five. The patient had had a prior exposure to ICM (iopromide) without any adverse reactions.

He was referred to our Allergology department for study of hypersensitivity to ICM. The skin prick tests (SPT), intradermal tests (ID) and epicutaneous test (ET) were conducted in accordance with the European Academy of Allergy & Clinical Immunology (EAACI) and European Network for Drug Allergy (ENDA) guidelines.[9] The SPT with iopromide (769 mg/mL) and iomeprol (714 mg/mL), ID test with iopromide (1/10) and iomeprol (1/1) and ET with iopromide (1/1) and iomeprol (1/1) were negative at the 20 minutes (SPT and ID), and 48 hours reading (ID and ET).

The drug provocation test (DPT) with alternative ICM, iomeprol (Iomeron®), was performed. The DPT with iomeprol was carried out, using incremental doses at one-hour intervals across two sessions separated by one week (5 mL > 10 mL > 15 mL on the first day and 10 mL > 20 mL > 30 mL on the second day, which amounted to a cumulative dose of 90 cc) was negative.

DISCUSSION

Our patient developed bilateral cervical angioedema 10 hours after ICM administration. The differential diagnosis of trauma, infection, ductal obstruction and neoplasms of the salivary glands were excluded based on the clinical history, analytical study and tomographic findings.

Considering the clinical history and time of onset of symptoms after a known trigger, the hypothesis of hypersensitivity to ICM was considered. To investigate a potential hypersensitivity to iopromide, allergological studies (SPT, ID and ET) were carried out, all with negative results, making the diagnosis of ICM hypersensitivity less likely. Considering the cervical CT findings of submandibular gland edema with adjacent fat stranding, but absence of edema in subcutaneous cellular tissues, a characteristic of edema in allergic reactions, the most probable entity was CIS.[10,11]

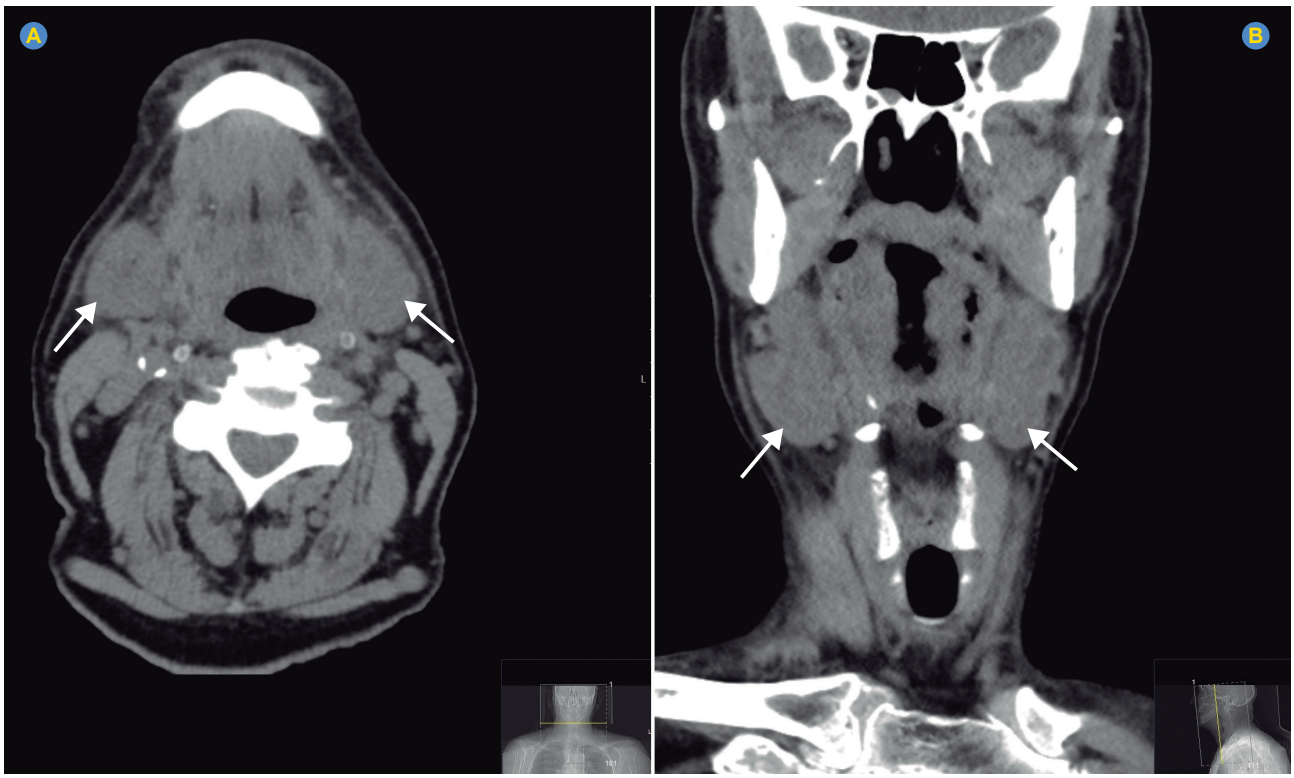

Figure 1 – Axial (A) and coronal (B) submandibular sialadenitis. Non-contrast computerized tomography images reveal enlargement of the submandibular glands with adjacent fat stranding, suggestive of edematous changes. Nonspecific bilateral submandibular lymph nodes are also observed. No obstructive calculi are identified.

The submandibular glands are the most commonly affected salivary glands in this context, as described in our clinical case, followed by the parotid glands, sublingual glands and, in rare cases, involvement of the thyroid, lacrimal gland, and pancreas has been reported.[4,11,12]

The pathophysiology of CIS is debatable; initially it was associated with a mechanism mediated by immunoglobulin E.[4] However, it is now believed that the condition is due to the toxic accumulation of iodine in the ducts of the salivary glands, triggering an inflammatory reaction and consequent obstruction of the salivary duct.[7,13] Premedication with antihistamines and corticosteroids was not proven to prevent episodes of sialadenitis.[7,11]

Current treatments for CIS are supportive therapy and the administration of anti-inflammatory agents, with the expectation that symptoms can fully resolve within a few hours to two weeks.[11,14] In our case, pain and edema of the submandibular glands resolved within five days of treatment with prednisolone.

The management of CIS remains a topic of debate. However, many reported cases indicate that the condition is generally benign and self-limiting, although there is a tendency for recurrence.[11]

The risk of recurrence with the administration of the same contrast is high. A literature review published in 2018 described nine cases of recurrence of sialadenitis after administration of different forms of low-osmolarity non-ionic iodinated contrast media.[12] Other studies suggested there was a lower risk with contrasts with low osmolarity and high solubility.[7]

Due to the risk of recurrence, the patient refused the DPT with the suspected ICM, so the DPT with alternative ICM was performed. The DPT – performed with Iomeprol, a monomeric, non-ionic ICM with low chemotoxicity, osmolality, and viscosity, and high solubility in water compared to other non-ionic ICM – was negative.[1,2]

The benign course of this syndrome should not limit the use of iopramide, but the use of iomeprol should be favored due to the patient's tolerance.

In conclusion, CIS is considered an uncommon reaction to ICM. The condition is typically benign and self-limiting and, although the administration of ICM is not contraindicated in life-saving situations, the risk of recurrence of sialadenitis is high. In the clinical case presented, after evaluating potential hypersensitivity reactions with skin tests, the DPT was performed with alternative ICM, which was negative. Although iopromide administration is not contraindicated, the authors consider that DPT with lower osmolarity ICM can be considered, thus providing the patient with a safer alternative.

AUTHOR CONTRIBUTIONS

LPD: Study design, data analysis and interpretation, writing of the manuscript.

ACC, CM: Data analysis and interpretation, writing of the manuscript.

MJS: Study design, critical review of the manuscript.

All authors approved the final version to be published.

PROTECTION OF HUMANS AND ANIMALS

The authors declare that the procedures were followed according to the regulations established by the Clinical Research and Ethics Committee and to the Helsinki Declaration of the World Medical Association updated in October 2024.

DATA CONFIDENTIALITY

The authors declare having followed the protocols in use at their working center regarding patients' data publication.

PATIENT CONSENT

Obtained.

COMPETING INTERESTS

The authors have declared that no competing interests exist.

FUNDING SOURCES

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

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