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CASE REPORT

Diagnosis and management of multiple salivary gland anomalies: A Case report

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Abstract:

Anatomical anomalies in the salivary glands, such as ductal ectasia and stenosis, are rare and can be idiopathic, congenital, or acquired. The diagnosis of these anomalies is based on imaging examinations and is sometimes challenging. A 65-year-old female patient complained of a recurrent volume increase in the right preauricular region for 43 years. The swelling was exacerbated during the periprandial period. Physical examination with palpation revealed a flaccid swelling. Aspiration showed clear liquid compatible with saliva. Ultrasonography and magnetic resonance (MR) sialography showed changes in ductal anatomy and sialocele. MR sialography allowed the diagnosis of sialocele secondary to idiopathic duct anomalies, ectasia, and strictures in the parotid ducts. The treatment was sialoendoscopy. The patient presented a good clinical evolution with a significant reduction in swelling in the preauricular region due to less saliva accumulation. Sialoendoscopy is an effective method for the diagnosis and treatment of major salivary gland anomalies.

Keywords: Ectasia, Parotid diseases, Sialoendoscopy, Sialocele, Stenosis.

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INTRODUCTION

Salivary gland obstruction is most commonly caused by salivary calculi or strictures of the duct[1]. Approximately 60–70% of cases of obstruction are caused by sialolithiasis and are sometimes not identified on imaging examinations, both with ultrasound and computed tomography, which have sensitivity and specificity rates of 95% and 98%, respectively[2,3].

Salivary gland swelling of unclear origin is frequently associated with duct strictures, which cause 25% of all glandular obstructions. Among the strictures of the salivary glands, 75% occur in the parotid duct. These strictures or stenoses develop secondary to inflammation in the duct wall and may be single or multiple[1].

Strictures are classified through sialoendoscopic evaluation into three types: type I, inflammatory; type II, fibrous with partial obstruction; and type III, fibrous with complete stricture. The degree of stricture represents the severity based on narrowing, mild to moderate (up to 50%), and severe (>50%)[4]. Constriction of the parotid duct usually occurs in the middle third in 39.6% and 37.8% of cases in the proximal third[1]. Strictures are responsible for a large proportion of obstructive sialadenitis[5].

Ductal ectasia or sialectasis is a rare type of heteroplasia that develops in the ducts of the parotid and submandibular glands[6]. This heteroplasia may have a congenital[6,7] or acquired origin, characterised by dilation of the salivary duct, which frequently occurs secondary to obstruction caused mainly by sialolithiasis and strictures of the duct[8]. They can cause an increase in recurrent swelling in the affected duct region[9], sometimes followed by painful symptoms not associated with eating[6], making the differential diagnosis with other diseases is essential.[8] Hereditary factors were considered a central element in the development of ectasia[7].

Only 5–10% of salivary duct obstructions occur due to chronic inflammatory processes, 60–70% due to sialolithiasis, and 15–25% due to strictures[2]. The other causes correspond to a rate of 1–3% and include allergic processes (granulomatous and autoimmune conditions, radiotherapy)[9], presence of anatomical abnormalities of the parotid duct[5], trauma[10], juvenile chronic parotitis[7], autoimmune disease[4], among others.

Sialoendoscopy is a minimally invasive technique that enables better visualisation and diagnosis of the ductal system and conservative treatment of benign salivary gland diseases[11]. This procedure is safe[11], and the treatment of obstructive duct conditions by sialoendoscopy has shown a success rate of >81%[12], which is considered the treatment of choice for salivary duct strictures[13].

Herein, we report multiple and uncommon anatomical anomalies of the parotid gland that triggered sialocele and management by sialoendoscopy.

CASE REPORT

A 65-year-old Caucasian female patient reported recurrent swelling in the cheek region, anterior to the right ear. In anamnesis, the patient reported hypothyroidism and was being treated. The patient had no history of facial trauma and was referred for mild xerostomia. The patient had consulted several dentists and physicians, and no effective diagnosis or therapy was found. The patient signed an informed consent form.

The swelling was asymptomatic, had been present for 43 years, started primarily around the eating period, and regressed with self-massage. Physical examination revealed a unilateral swelling of approximately 1.5 cm, with a soft, floating consistency anteriorly to the right ear, below the zygomatic arch. The adjacent skin did not show any colour change and was intact (Fig. 1). Intraoral findings were unremarkable.

Fig. 1. The frontal view of the patient shows swelling in the preauricular region.

Extraoral fine needle aspiration was performed, which presented a translucent, fluid, foamy-looking liquid compatible with saliva. Biochemical analysis confirmed the clinical findings (amylase levels, 169,660 U/L; total proteins, 0.83 g/dL). Panoramic radiography revealed no calcification or bone lesions. Ultrasonography demonstrated diffuse dilation of the right parotid duct and findings compatible with ductal ectasia.

A provisional diagnosis of sialocele associated with ductal ectasia was made. Since biopsy did not appear adequate and a more accurate evaluation of the ductal system (including the gland itself) was necessary, magnetic resonance (MR) sialography was requested.

MR sialography showed important anatomical changes in the right parotid gland and revealed changes in the left parotid and submandibular glands. The section showed a hypodense image on the medial border of the masseter muscle and dilation of the right parotid duct, showing a well-defined image of approximately 12 mm along the longest axis. In addition, there was a marked dilation of the right parotid duct, which was more important in the posterior region, associated with the prominence of the interglandular ducts. Important ductal constriction was observed before ectasia, absence of sialolithiasis, and lymph node enlargement. A slight ectasia of the left parotid duct was more evident in the anterior portion. The images showed a slight prominence of the posterior portion of the submandibular ducts, with no evidence of intraglandular dilation (Fig. 2). To confirm these anomalies and provide a noninvasive approach, sialoendoscopy was proposed.

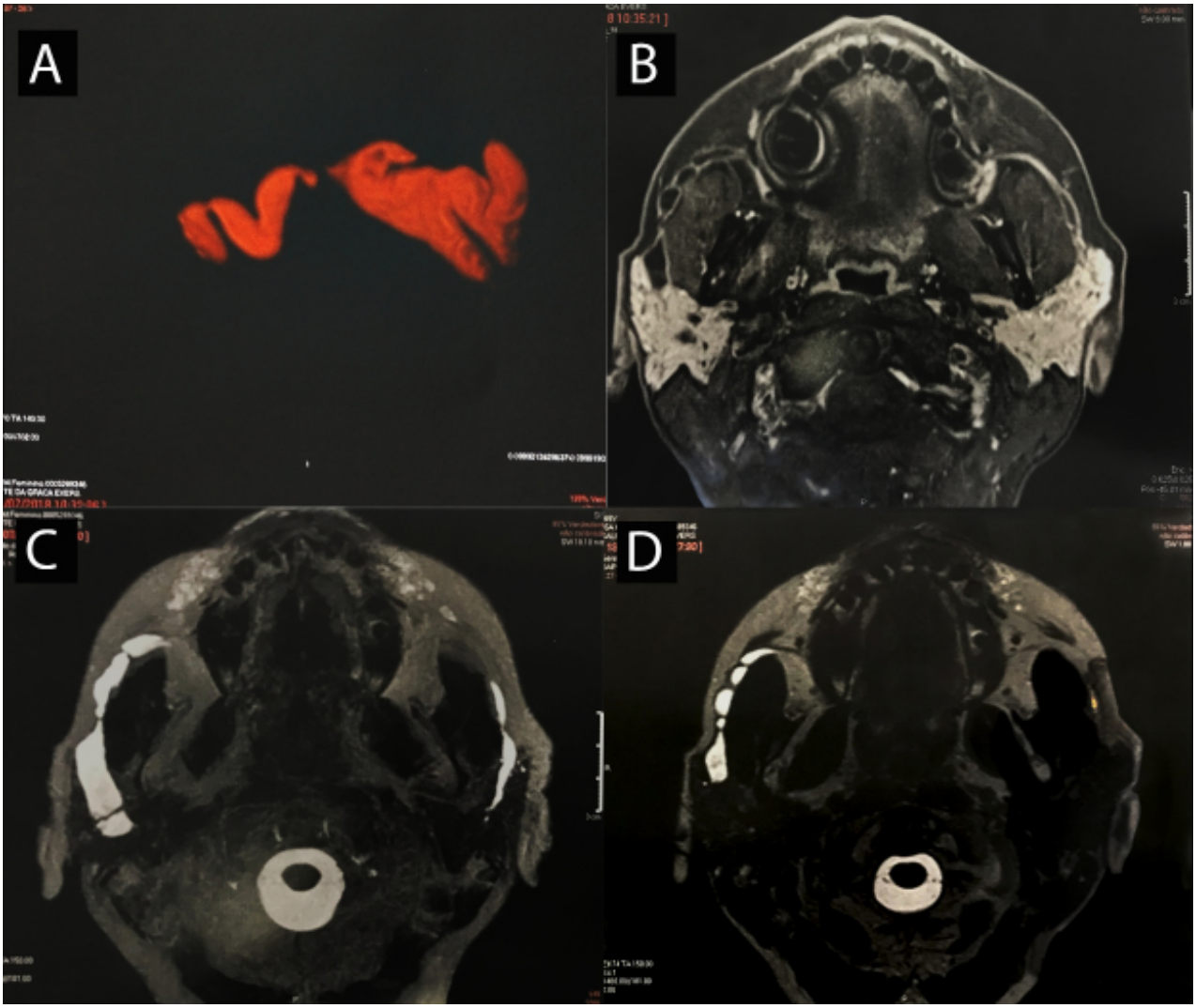

Fig. 2. MR sialography shows in (A) (T2-weighted image) stricture of the right main duct followed by stenosis. (B) Axial section (T1-weighted image) of the right and left parotid glands and its main ducts. (C) Axial section (T2-weighted image) ductal stricture, ectasia, and sialocele in the right parotid gland, and stenosis in the left parotid gland. (D) Axial section (T2-weighted image) shows stenosis of the right parotid gland.

Sialoendoscopy was performed in the right and left parotid ducts. The procedure was performed under general anaesthesia. The sialoendoscope was inserted through the natural orifice of the parotid duct. Probes of 1.1-mm and 1.5-mm diameter were conducted through the ducts. The irrigating solutions used were saline and one dexamethasone (4 mg, 2.5 ml) in each parotid gland. In the right parotid duct, a stricture was observed that embraced the 1.1-mm probe in its middle third, passing through it without much resistance and without the need for too much force to pass through it (Fig. 3A-D). The ductal system was explored, and sialolithiasis or mucous plugs were not found during sialoendoscopy. Anatomical changes in the ductal system of the right and left parotid glands on MR sialography were confirmed. The postoperative instructions consisted of cold compresses for 48 h and diet restriction of citrus and dry foods for 30 days. The patient recovered without complications.

Given the MR sialography images and the diagnostic exploration by sialoendoscopy, the definitive diagnosis was sialocele secondary to idiopathic duct anomalies (ectasia and strictures in the parotid ducts).

The patient was followed-up for 9 months after sialoendoscopy and showed excellent clinical evolution. The patient reported that there were still some episodes of swelling in the cheek, only when eating citrus foods or in a few periprandial moments. These swellings were significantly smaller than before. There were no abnormalities on physical examination during visual inspection and palpation (Fig. 4). Follow-up MR sialography at 9 months after sialoendoscopy showed dilation and tortuosity of the right parotid duct up to 1.3 cm in diameter. The ducts of the submandibular glands and left parotid gland had mild ectasia (Fig. 5).

Fig. 4. The frontal view of the patient 9 months after sialoendoscopy.

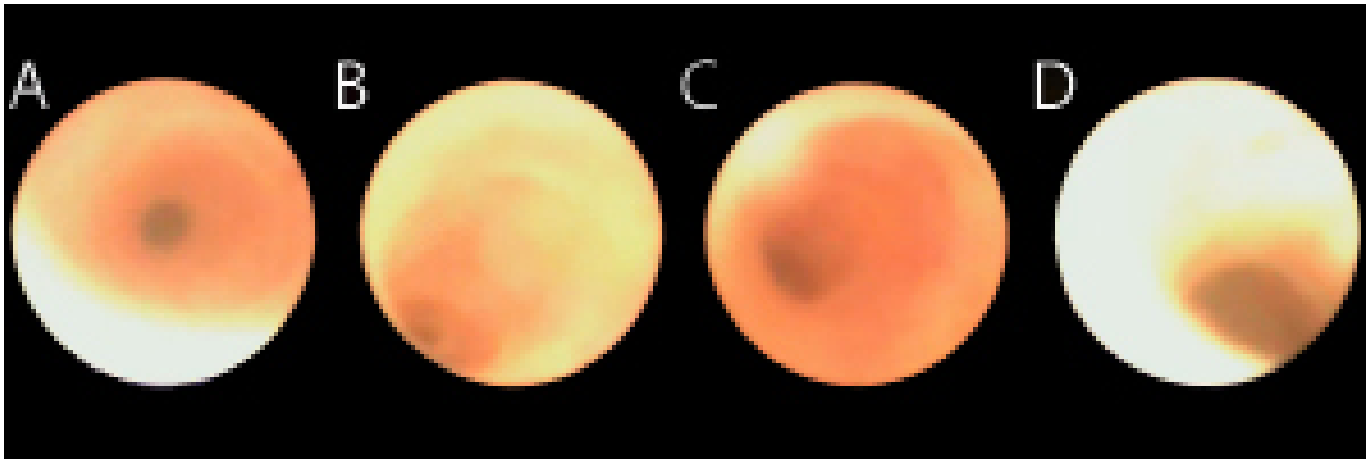

Fig. 3. Sialoendoscopy of the right parotid gland. (A) Normal duct lumen and ectasia are demonstrated in the background. (B) Stenosis of the duct. (C) Duct ectasia in the middle third, after passing through the stenosis. (D) Duct ectasia in the proximal portion.

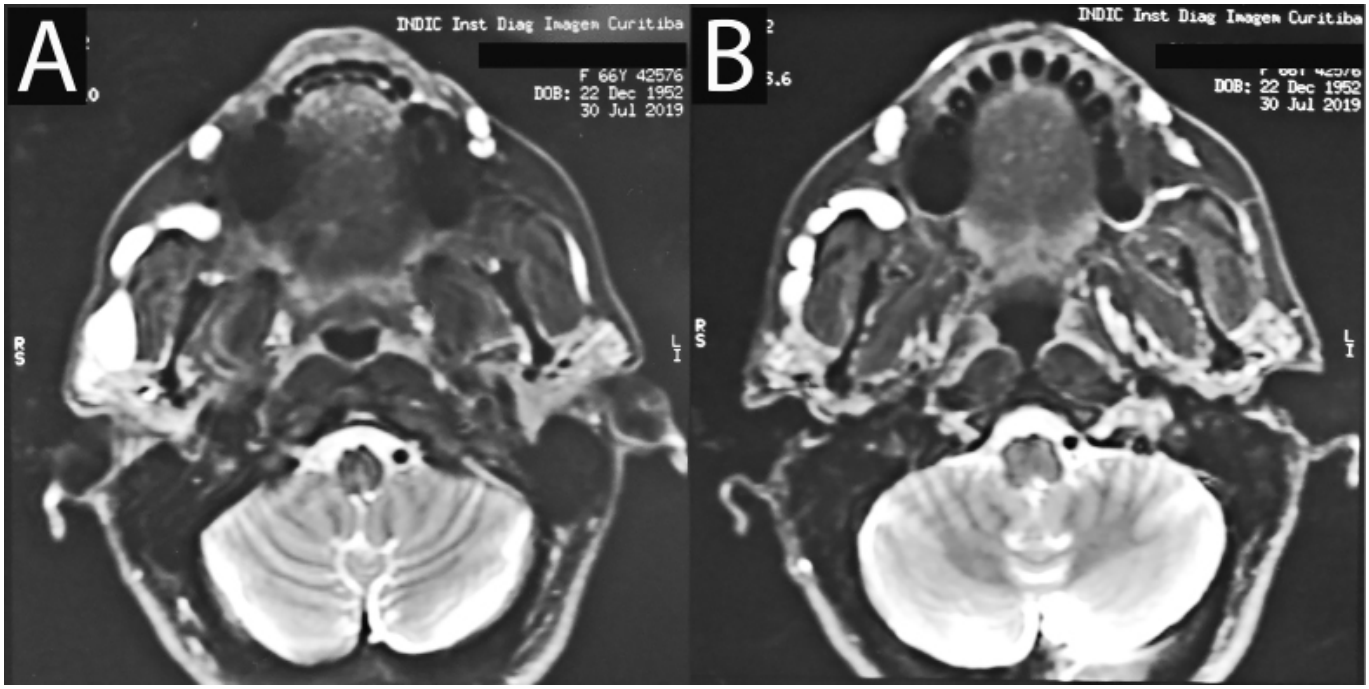

Fig. 5. Magnetic resonance sialography 9 months after sialoendoscopy. (A) Axial section (T2-weighted image) stenosis, ectasia, and sialocele of the right parotid duct. (B) Axial section (T2-weighted image) shows stenosis of the right parotid duct.

DISCUSSION

The diagnosis of a few salivary gland anomalies is a real challenge. Considering the patient's history of swelling, clinical presentation, and region of the lesion, differential diagnoses were focused on salivary gland disorders or reactive lesions. Swelling in the preauricular region may be the result of different diseases and anatomical anomalies. Among the anomalies that affect the salivary glands, tumours and sialadenopathies are the most frequent[3].

Infectious diseases, such as sialadenitis, chronic obstructive parotitis[5,7], and juvenile chronic parotitis[7], can affect the parotid region. The absence of purulent secretion, fever, erythema, and pain, which are characteristics and symptoms of an infectious condition, ruled out these entities.

Unilateral swelling concomitant with salivary stimulus and filled with saliva indicated a local association with the parotid gland. The extraoral massage manoeuvre and the consequent partial reduction in oedema reject the hypothesis of total duct stricture, as well as disorders of the salivary glands characterised by permanent oedema, such as sialoadenosis described in polycystic ovary syndrome, alcoholism, and malnutrition[14].

Swelling, characterised by a collection of extraglandular saliva, was not associated with a history of trauma or previous surgery in the face[15] or exposure to radioactive iodine[13], which would trigger cystic cavities. Sialocele or salivary pseudocyst is a rare clinical condition in which saliva leaks into the periglandular tissue due to inadequate salivary excretion due to rupture of the glandular parenchyma leading to swelling of the affected glandular region[15]. Although ultrasonography has reported diffuse dilation of the parotid duct and ductal ectasia, the cystic cavity has not been reported.

Sialolithiasis can lead to partial obstruction of the salivary duct, with a consequent reduction in salivary flow, which is usually asymptomatic. The imaging examinations performed, transcutaneous ultrasound and panoramic radiography, did not reveal sialolithiasis. Although ultrasonography is a diagnostic test that identifies sialolithiasis and strictures of the duct with high sensitivity and specificity[10], it has some limitations, especially when obstructions are located in the distal ductal portion, where visualisation is limited[3].

Therefore, diagnostic hypotheses led to salivary retention in the parotid duct. Salivary fluid retention can occur due to ectasia, sialocele in the glandular parenchyma or parotid duct, and partial or total strictures. Strictures are responsible for the obstruction of salivary ducts in 15–25%[1,2], and 75% of them are located in the parotid duct[1]. This change is due to several local factors that can cause a delay in saliva excretion, resulting in retrograde infection and swelling of the gland[5].

There are some imaging modalities available to diagnose salivary gland anomalies, among which ultrasound is widely used because it is a cost-effective, fast, and noninvasive method with high sensitivity to identify strictures in the middle third of the anterior duct[3], as well as dilation and signs of infection[10]. However, in this case, transcutaneous ultrasound has not been completely elucidated and conclusive. MR sialography is essential for diagnosing anatomical glandular anomalies and provides sufficient information for diagnosis and appropriate treatment plans. Vogl et al.[10] highlighted that MR sialography is a safe, painless, and reliable method of identifying obstructions in the salivary ducts and changes in the adjacent soft tissues.

Sialoendoscopy is a therapeutic procedure for the diagnosis and management of obstructive salivary disorders. Diagnostic sialoendoscopy allows the identification of different gland pathologies, such as localised duct stenoses, mucous plugs, and duct anomalies. This approach as a treatment method allows conservative management and leaves a functional gland in most patients[3,4,11]

MR sialography and sialoendoscopy showed that the patient had short stenosis in the middle portion of the duct, type II, and moderate degree. In addition, the patient did not have an infection associated with swelling. This is probably because the patient used to do self-massage to reduce swelling and maintain saliva excretion, making it difficult for bacteria to establish upward colonisation via the duct. Usually, a reduction in salivary flow allows bacteria to ascend through the duct, resulting in infection, in addition to the fact that flow stasis can result in the development of a mucous or fibrous plaque and constriction or stricture of the duct[5].

Additionally, there are complementary imaging modalities to diagnose anatomical anomalies in the salivary glands. The factors that cause these anomalies are often unknown and considered idiopathic causes[1]. Given the imaging findings and sialoendoscopy, bilateral ductal ectasia with prominence in the posterior region of the right side of the parotid gland and discreetly also in the ducts of the submandibular glands, it is suggested that these anomalies are congenital. Congenital dilation of the parotid duct can affect the parotid glands bilaterally; however, the increase in volume can only be seen unilaterally[7].

The surgical treatment, parotidectomy, had previously been proposed for the patient; however, she refused this approach due to possible sequelae to the facial nerve and the invasiveness of the procedure. Sialoendoscopy was performed despite the limitations found as only a few centres in this country perform it, associated with the high costs of the procedure. During sialoendoscopy, saline and anti-inflammatory corticosteroids were used as irrigating solutions. The administration of irrigating solutions, such as corticosteroids, antibiotics, and saline[11], is intended to remove debris and dilate the duct by hydrostatic pressure to facilitate visualisation[9], in addition to having therapeutic properties, such as anti-inflammatory and antibiotic actions[9]. Corticosteroids have anti-inflammatory and antiproliferative effects, significantly reducing the formation of primary strictures or their recurrence[1,13]. In this case, during sialoendoscopy, according to clinical criteria, it was considered unnecessary to use artifices to maintain the ductal lumen. The use of mini-forceps, mini-baskets, balloons, tweezers, and microdrills described in the literature is very useful in treating strictures caused by sialolithiasis[11].

Mini-baskets are used for inflammatory or fibrous structures. Baskets associated with microdrills can also be indicated in cases of complete duct obstruction or filiform healing. Stents are indicated for cases in which the stricture is in the region of the parotid papilla or in the distal duct, the condition of which prevents the insertion of the endoscope[13]. Intraluminal dilation balloons guided by sialography or sialoendoscopy can also be used in cases of punctual or diffuse strictures[13]. The use of a stent to enlarge strictures substantially reduces the rate of recurrence[13].

Technological advances, such as diagnostic imaging using MR sialography and a conservative approach using sialoendoscopy, were essential in this case. Both allowed functional maintenance of the parotid gland[12] and mainly eliminated the need for invasive surgery, which, although normally effective[11], implies known undesirable sequelae, such as facial nerve injury and aesthetic problems.

Some authors recommend the use of imaging tests after sialoendoscopy to confirm the elimination of constriction[10]; however, we emphasise that clinical information is sovereign. The patient remains in follow-up due to discrete and transient swellin, and she is very satisfied with the results obtained. For the subsequent treatment of persistent swelling, although scarce, the literature reports that the condition may revert spontaneously, or conservative treatments may be performed. Among these, repeated fine-needle aspiration, compression, the use of anticholinergics, or botulinum toxin are cited.

Alternatively, when the major duct of the deep parotid lobe is affected, the proposed treatments are intraoral drainage by catheter, surgical excision of the cyst, and parotidectomy associated with conservative treatment[15].

CONCLUSION

The patient had uncommon and rarely described clinical conditions due to different anatomical salivary gland anomalies that triggered a sialocele. MR sialography showed accurate diagnostic information on ductal changes in the parotid and submandibular glands. A successful conservative approach was performed using sialoendoscopy to avoid parotidectomy. The key role of sialoendoscopy is gland-preserving management.

Ethical approval: Not required. The patient has authorised the publication of this case report, respecting complete anonymity and ethical aspects.

Conflict of interest: The authors declare that they have no conflict of interest.

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