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Original Article

Clinical efficacy of botulinum toxin in salivary duct stenosis: A preliminary study of six cases

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ABSTRACT

Introduction: Salivary duct stenosis is the second most common cause of obstructive pathology after lithiases, and it primarily affects the parotid gland. Salivary duct stenosis is treated with drug therapy and/or sialendoscopy. If unsuccessful, surgical removal of the gland is indicated, but it is associated with a high risk of facial morbidity. The aim of this study is to evaluate the clinical efficacy of an alternate treatment, botulinum toxin, in salivary duct stenosis.
Material and methods: In a preliminary retrospective study from January 2011 to December 2014, six patients with parotid duct stenosis received 50 IU of botulinum toxin in three injections in the parotid gland. The frequency of relapses and the intensity of pain and swelling were recorded before and after treatment. The onset of action and duration of efficacy were also assessed.
Results: Four of six patients showed a decrease in the frequency of swelling episodes and greater pain relief during the first year of treatment, but to a lesser extent after 2 years. The mean duration of efficacy was 3.5 months with an interval between two injections of 5.7 months. Only one parotidectomy had to be performed. No major side effects were observed, with only one case of local infection at the injection site.
Conclusion: Botulinum toxin appears to be a viable alternative in treating salivary duct stenosis before resorting to surgical gland removal.

1. Introduction

Salivary duct stenosis is rare, causing 15% to 25% of obstructive symptoms, after lithiases [1]. If lithiases most often occur in the submandibular gland, stenosis mainly affects the parotid gland and accounts for half of parotid obstructive symptoms [1,2].

Salivary obstructive symptoms are characterised by painful saliva retention with swelling and pain, and infection after a number of swelling episodes. If lithiases cause mealtime obstructive syndrome, stenosis causes obstructive syndrome unrelated to meal times. The absence of lithiasis on imaging (ultrasound, computed tomography scan) confirms the diagnosis of stenosis. This diagnosis could then be confirmed with sialendoscopy (SE), sialo magnetic resonance imaging (MRI), or cone beam three-dimensional (3D) sialography [3]. The various types of stenosis have recently been classified by Marchal et al. [4] (Table 1, Fig. 1).

The aetiology of stenosis is often unknown. In certain cases, stenosis may be caused by ductal injury (e.g., jugal wound, previous ductal surgery), a chronic inflammatory pathology (e.g., lithiases, Sjögren syndrome, connectivitis, juvenile recurrent parotitis) [2], or radioactive iodine in thyroid carcinoma treatment [5].

Treatment is based on drug therapy (i.e., antibiotics, anti-inflammatories, antispasmodics) prescribed in cases of acute swelling and/or inflammation or SE [1,6]. When these protocols are ineffective, the question of parotidectomy with its high risk of postoperative facial palsy can be discussed.

For this reason, botulinum toxin (BTX) has been proposed to reduce salivary flow [7,8].

We report our experience in a preliminary short series of six patients.

2. Material and methods

2.1. Subjects

The inclusion criteria were as follows:

- adults with salivary duct stenosis visible on 3D sialography or sialo-MRI;

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Table 1
Endoscopic classification of stenosis [5].

Score	Endoscopic aspect
S0	No stenosis
S1	Intra-duct stenosis in the shape of a diaphragm (unique or multiple)
S2	Singe duct stenosis (principal duct)
S3	Multiple or diffuse duct stenosis (principal duct)
S4	Generalised stenosis (principal and secondary ducts)

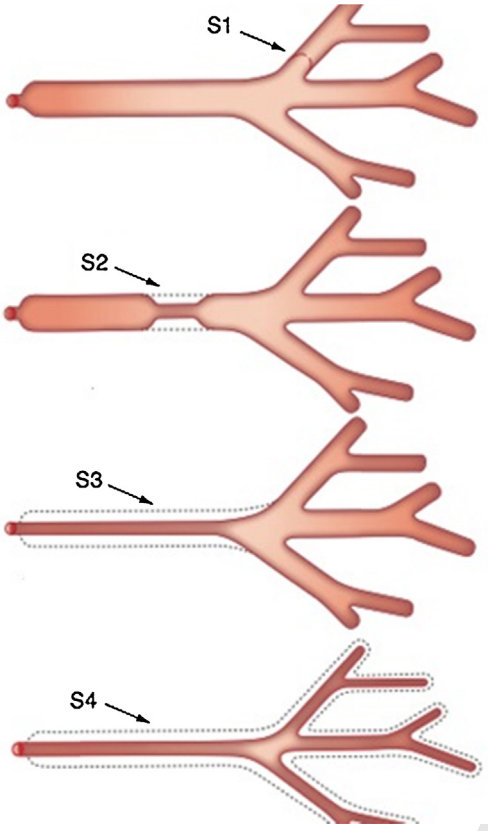

Fig. 1. Different aspects of salivary stenosis (S) [5].

- a previous course of ineffective drug therapy;
- a previous unsuccessful ductal dilatation using a balloon catheter under SE or 3D sialography.

Stenoses in these patients were confirmed on 3D sialography, sialo-MRI, and SE. They were located in the anterior third of the parotid duct in two cases, at the junction of the anterior third to middle third duct in one case, in the middle third in three cases, and in the posterior third in one case (*référence de la classification en tiers*). One patient had a double stenosis associated with Sjögren syndrome. For all six patients, a total of seven stenoses were found. None of them had been treated with radioactive iodine. Patients were followed up in the maxillofacial surgery department of the university hospital centre La Timone in Marseilles between January 2011 and December 2014 (Table 2).

Patients with lithiases, those who were hypersensitive to type A BTX, and those suffering from muscular disorders (e.g., myasthenia gravis, Lambert-Eaton syndrome) were excluded.

BTX protocols have already been validated for the salivary gland, and the ethics committee was briefed on the study. The patients were warned of potential side effects of BTX, which are xerostomia, infection at the injection site, and paresis of the marginal mandibular branch of the facial nerve, and gave informed consent, including for eventual publication.

2.2. Methods

Adapting our protocol from series reported in the literature (doses of 22.5 to 60 UI, depending on the pathology, injected into three or four sites) [7–9], injections were dosed at 50 UI of type A BTX (XEOMIN®), reconstituted in 1 mL of injectable 9 mg/mL (0.9%) sodium chloride in a 1-mL graduated syringe. We used neither ultrasound localisation nor local anaesthetic.

The BTX was injected into three sites of the parotid gland – the upper pole, the lower pole, and the anterior portion of the gland (Fig. 2). The subcutaneous procedure was performed slowly, using a sterile 25–30-gauge needle (0.30–0.50) supplied with the injection kit.

2.3. Assessment

Assessment criteria included the type of symptoms, pain, swelling (or both together), frequency of relapse on an efficacy scale of 0 to 4 (where 0 is the absence of symptoms, 1 is < 1/month, 2 is > 1/month, 3 is > 1/week, and 4 is > 1/day), recorded before and after BTX injections. This assessment was completed with the Visual Analogue Scale (VAS) for intensity of pain, both before and after injections. For greater objectivity in evaluating the effect of BTX, the 0–4 efficacy score was multiplied by the before-and-after VAS scores at 12 and at 24 months' follow-up.

We also collected data on the onset of action (time elapsed between injection and feeling relief), the duration of efficacy (time between injection and point of no longer feeling relief), and eventual side effects. Finally, patient follow-up depended on the evolution of symptoms.

3. Results

During the period under study, we operated on 27 patients for whom drug therapy was ineffective for parotid gland retention pathologies. Of the 27 patients, 13 presented with stenosis. Balloon catheter dilatation was performed on all the patients. The six among them for whom the procedure was ineffective constituted our BTX sample.

Results in Tables 2 and 3 provide data from retrospective follow-up consultations and telephone interviews. All patients were women, with a mean age of 63.8 years (range 48–78). In five of the six cases (83.33%), inflammation was the cause of stenosis. In the remaining case, stenosis was the post-traumatic result of an endobuccal approach in treating the posterior part of the middle third of the parotid duct for lithiasis.

Prior to BTX therapy, three of our patients had both pain and swelling of the parotid gland; one had swelling alone; one, pain alone; and one, discomfort associated with intermittent drooling with a "metallic" taste. Mean VAS was 7.7/10 for four patients during relapse. Frequency of relapse was greater than 1 day in one case, greater than 1 week in two cases, greater than 1 month in one case, and less than 1 month in two cases.

The average time elapsing between the first symptom and the first BTX injection was 12 years (2–40). Patients received a mean of 3.6 injections each. Mean follow-up time was 18.66 months (6–39).

Following BTX therapy, global mean VAS score was 1.92/10. Frequency of relapse was < 1 month in four cases and > 1 day in two cases. Mean onset of action was eight days (2–15), with a mean duration of efficacy of 3.5 months (3–6). Interval between two injections averaged 5.71 months (3–10). Finally, the type of

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