Case Report

Botulinum toxin in the treatment of asymmetric crying face syndrome: a case report

Toxina botulínica para tratamento de síndrome do Choro Assimétrico: relato de caso

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■ **ABSTRACT**

Asymmetric crying face syndrome is a congenital condition secondary to hypoplasia or absence of the depressor muscle at the mouth angle. It is a common condition that presents with facial asymmetry while crying and smiling and may be associated with other congenital malformations. Children with this deformity may experience psychosocial difficulties and introversion. The therapeutic arsenal of this condition has already been studied and discussed in the literature with an emphasis on surgical and invasive approaches. We report here a case of a 9-year-old child with this syndrome, treated less invasively with botulinum toxin, with good result and satisfaction.

Keywords: Botulinum toxin type A; Facial asymmetry; Facial paralysis; Abnormalities of the mouth; Congenital abnormalities.

■ **RESUMO**

Síndrome do choro assimétrico é uma condição congênita secundária à hipoplasia ou ausência do músculo depressor do ângulo da boca. Trata-se de uma condição não tão incomum que pode cursar com assimetria facial ao chorar e sorrir, além de poder estar associadas a outras malformações congênitas. Crianças com essa deformidade podem sofrer dificuldades psicossociais e introversão. O arsenal terapêutico dessa condição já foi estudado e discutido na literatura com ênfase em abordagens cirúrgicas e invasivas. Relatamos aqui um caso de uma criança de 9 anos com essa síndrome, tratada, de forma menos invasiva, com toxina botulínica, com um bom resultado e satisfação.

Descritores: Toxinas botulínicas tipo A; Assimetria facial; Paralisia facial; Anormalidades da boca; Anormalidades congênitas.

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Article received: May 1, 2018.
Article accepted: June 4, 2018.

Conflicts of interest: none.

DOI: 10.5935/2177-1235.2018RBCP0104

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INTRODUCTION

Asymmetric crying face (ACF) syndrome was first described in 1931 by Parmelee while observing the crying of a newborn[1]. It is a congenital alteration secondary to hypoplasia or absence of the depressor muscle at the mouth angle. It can develop in up to 0.6% of births[2] and present with facial asymmetry when crying and loss of saliva in the oral commissure of the affected side. Other facial movement muscles are unaffected[3].

The diagnosis may be clinical or aided by electromyography. The association with other congenital malformations is possible and may be from the gastrointestinal, cardiovascular, skeletal, genitourinary, and even central nervous system. Congenital hypoplasia of the depressor muscle of the mouth angle develops in isolation in most cases[4], and the investigation of other congenital anomalies is controversial[2]. Its etiology is not yet well defined but is believed to be multifactorial.

The clinical diagnosis is more difficult with the child's growth, since, with the passage of time, the risorius and other muscles begin to dominate facial expression[4]. The main differential diagnosis of this condition is with obstetric traumas and congenital facial paralysis, in which there are other facial expression alterations such as deficient ocular closure, absence of frontal wrinkles, and deletion of the nasolabial sulcus.

The diagnosis and follow-up of this syndrome are well discussed and established in the literature. Nevertheless, the treatment of facial asymmetry is rarely studied, and there are differences in the initial conduct of these cases. Thus, we present here a case of a child with ACF syndrome successfully treated with botulinum toxin on the unaffected side.

CASE REPORT

A 9-year-old female patient, with complaint of mouth asymmetry since birth, currently suffers from *bullying* at school. She had stable asymmetry throughout growth, with greater prominence of the deformity when crying and smiling.

According to the mother, gestation was uneventful. Personal history is only bronchial asthma with eventual use of inhaled corticosteroids.

The patient was evaluated by a pediatrician, who ruled out associated congenital malformations. Upon examination, there were no significant changes at rest (Figure 1), but the smile was asymmetric due to the absence of action of the depressor muscle at the left mouth angle (Figure 2). The remaining facial muscles had no clinically detectable changes (Figure 3).

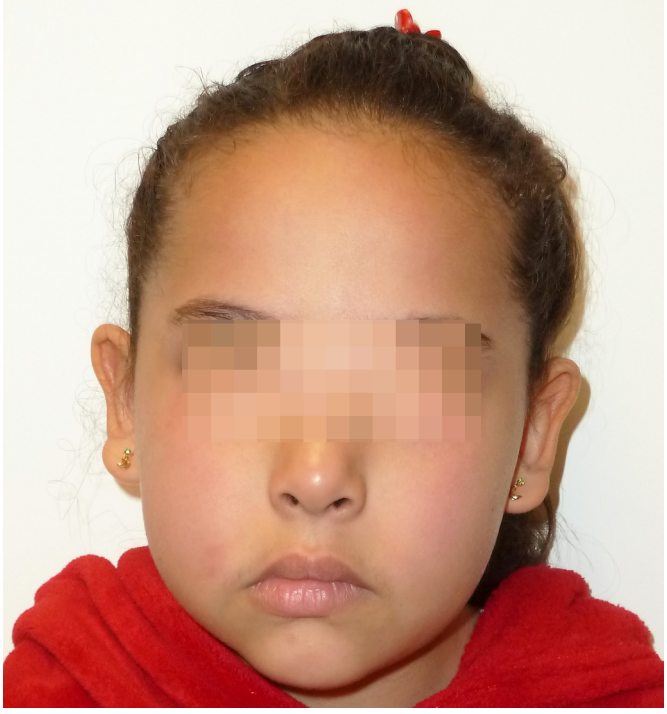

Figure 1. Resting - without significant asymmetry.

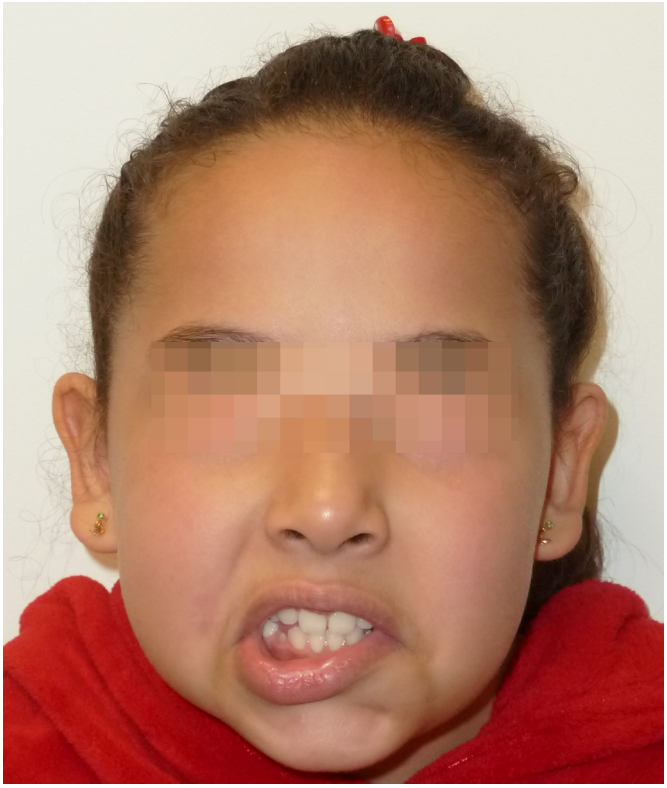

Figure 2. Mouth asymmetry when smiling - absence of depression action at the mouth angle on the left side.

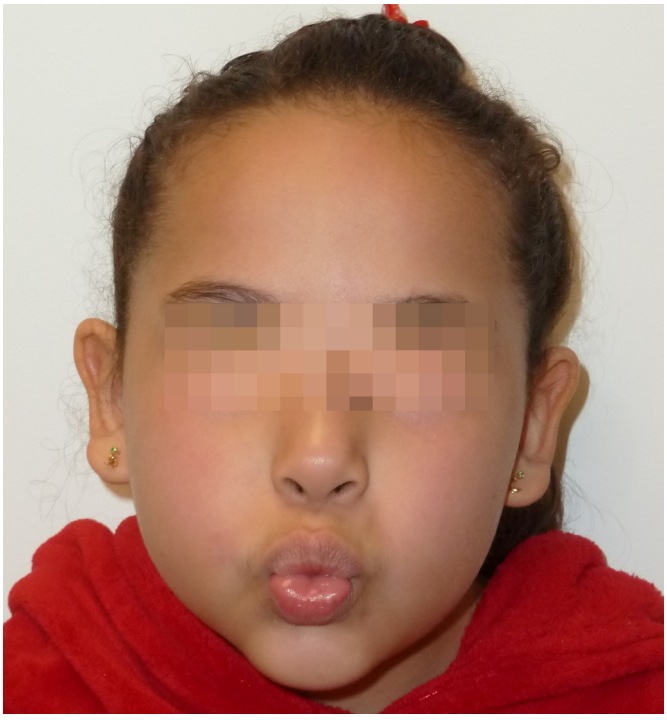

Figure 3. Dynamic physical exam normal resting - good function of the orbicular mouth muscle.

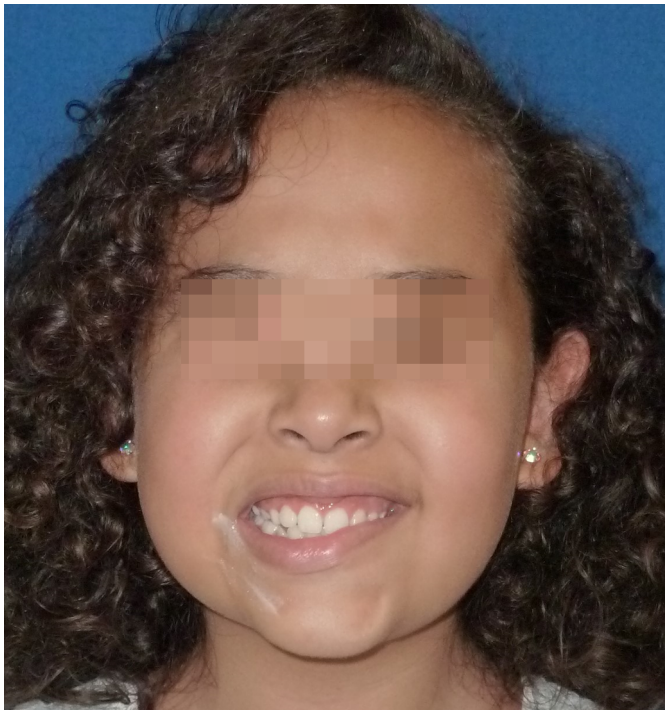

Figure 4. Result after the first application - application of 5 IU of botulinum toxin type A (Botox®; Allergan).

The therapeutic possibilities were proposed for the patient and mother. After the consent of the patient and her caregiver, 5 IU of botulinum toxin type A (Botox®; Allergan) was directly applied to the depressor muscle at the right (unaffected) mouth angle. After 14 days, an insufficient result was verified (Figure 4), and another 5 IU (total of 10 IU) was applied.

The patient returned satisfied with the result, showing symmetry when smiling, as seen in Figures 5 and 6.

DISCUSSION

ACF syndrome has been extensively studied in pediatric publications with a focus on diagnosis and follow-up necessary for these children and differential diagnoses and associations with other congenital malformations, but there are few discussions on the treatment of the central characteristic of this condition: facial asymmetry secondary to hypoplasia of the depressor muscle at the mouth angle.

The most studied and described treatment in the world literature is the weakening of the sound side through selective neurectomy of a marginal nerve branch of the mandible or myectomy of the depressor muscle at the mouth angle[5]. Udagawa et al.[6] described a surgical intervention on the affected side, in which a fascia lata graft was performed in 7 children with facial asymmetry when crying, showing good results. Other

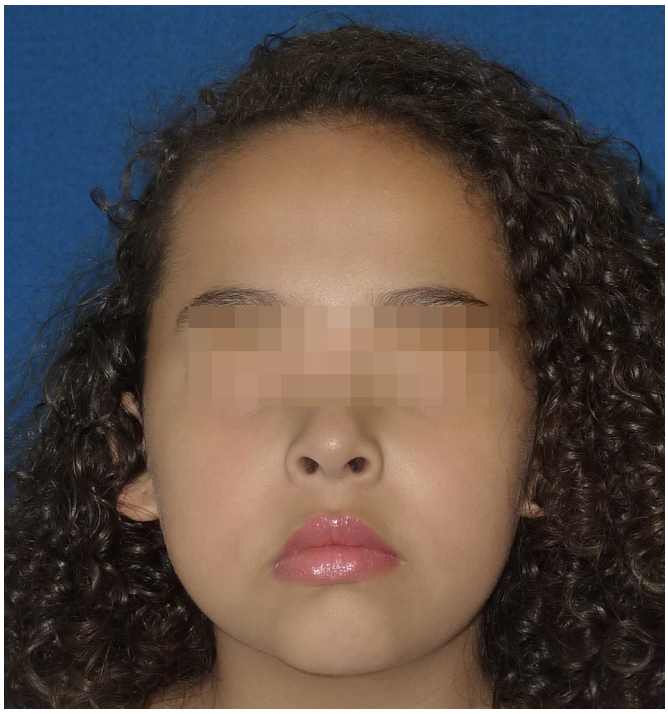

Figure 5. Final result after the second application. Resting - total dose of 10 IU botulinum toxin type A (Botox®; Allergan).

innumerable invasive forms have also been published such as functional microsurgical transfer, fascia graft, and even transposition of the digastric muscle.

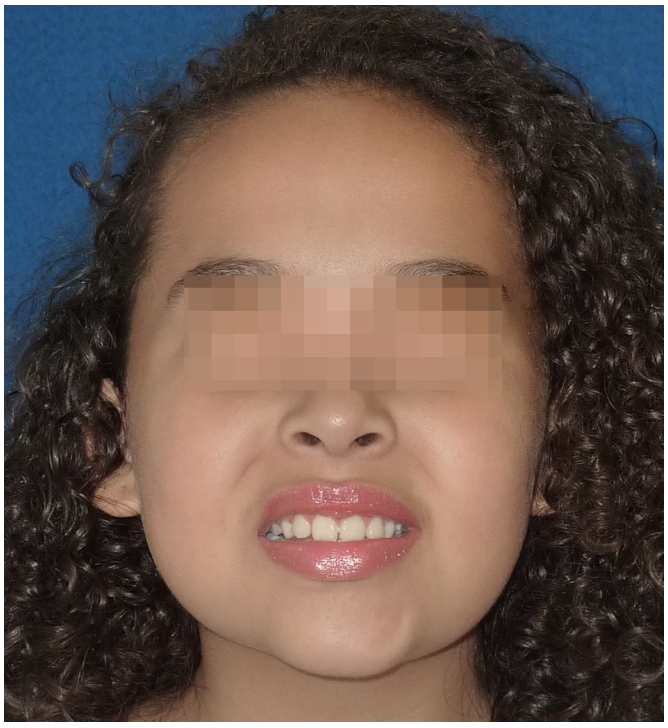

Figure 6. Final result after the second application. Smile - total dose of 10 IU of botulinum toxin type A (Botox®; Allergan).

The treatment modalities proposed in other studies have the need for surgical intervention as a disadvantage, thus exposing children to anesthetic risk. Therefore, less invasive alternatives such as the selective blockage of the marginal nerve branch of the mandible and application of botulinum toxin were also studied.

Tulley et al.[7] demonstrated in 2000 the application of botulinum toxin in 5 adult patients who presented with isolated facial paralysis at the marginal branch of the mandible, with excellent results. Isken et al.[8] published in 2009 two cases of children, one 4 years of age and the other 16 months, with facial asymmetry when crying successfully treated with botulinum toxin on the unaffected side, discussing in this work the benefits of a simple therapy for children with facial deformity that may eventually present psychosocial difficulties and introversion.

Botulinum toxin is already used safely in several childhood conditions such as spasticity in cerebral palsy, strabismus, dystonia, and hyperhidrosis, so there is sufficient evidence to demonstrate safety in its use in children[8].

Here, we present the case of a 9-year-old child with facial asymmetry of the mouth when smiling and crying, extremely uncomfortable with her physical condition. A safe, quick, and easy treatment was performed with the use of botulinum toxin, demonstrating a good result and with total patient satisfaction.

COLLABORATIONS

LGMPM Analysis and/or interpretation of data; study design and design; methodology; writing - preparation of the original; writing - revision and editing.

HAN Analysis and/or interpretation of data; conceptualization; investigation; conducting operations and/or experiments; writing - revision and editing; supervision.

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