

# THE CLINICAL OUTCOME OF BOTULINUM TOXIN TYPE A INJECTION IN CONGENITAL ESOTROPIA : A CASE SERIES

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## ABSTRACT

Botulinum toxin type A (BTX-A) is the precise drug to cause temporary paralysis of extraocular muscles and change ocular alignment with few side effects. Congenital esotropia is an ocular misalignment characterized by a convergent non-accommodative ocular deviation which develops at approximately six months of age. Botulinum toxin type A has been used in congenital esotropia as an alternative treatment to surgery. In this study, we report three cases of large angle congenital esotropia more than 45 prism diopter (PD) underwent BTX-A injection simultaneously on both medial rectus muscles. The procedure was done under total intravenous anesthesia (TIVA) with upright position. A unit of BTX-A in 0.1 milliliter was injected through the intact conjunctiva into the medial rectus muscle. Two weeks after injection, two patients developed exotropia and one patient developed ptosis. Four months after injection, all the cases showed improvement in ocular alignment (less than 35 PD) with no other complication. Botulinum toxin type A injection represents a secure alternative procedure in case of congenital esotropia and reduce the angle of deviation before surgical interventions were planned.

**Keywords** : Botulinum toxin, congenital esotropia, case series.

## 1. INTRODUCTION

Different strains of *Clostridium botulinum* produce eight exotoxins (A, B, C1, C2, D, E, F, G).<sup>1,2</sup> Types A, B, and E are commonly associated with human botulism cases and cause prolonged paralysis.<sup>1,3</sup> Botulinum toxin type A (BTX-A), is a high molecular weight protein of about 900 kilo daltons made up of toxic units of 150 kilo daltons.<sup>3</sup> Botulinum toxin type A has been studied for selective weakening of extraocular muscles and can be used to treat small or moderate esotropia and exotropia. However, for large angle esotropia, BTX-A is considered to be less effective and surgery should be considered instead. Botulinum toxin type A was the right drug to cause temporary palsy of extraocular muscles and manufacture permanent changes in ocular alignment with few side effects. The advantage of BTX-A injection is administration under sedation in children with little risk and lower cost. The most common side effect after injection include temporary ptosis and vertical misalignment resulting from spread into surrounding muscles.<sup>1,4</sup>

Congenital esotropia is an ocular alignment disorder characterized by a convergent non-accommodative persistent ocular deviation of relatively large angle which develops at approximately 6 months of age.<sup>5</sup> The etiology of congenital esotropia is still unknown. There are invasive treatments for congenital esotropia include surgery procedure (recession of both medial rectus muscles) and chemodenervation induced by botulinum toxin injection.<sup>4,5</sup> However, there is no case report about the use of BTX-A injection for congenital esotropia in South Sulawesi, Indonesia. Therefore, the aim of this study is to report the result of BTX-A injection in three cases of congenital esotropia.

## 2. CASE PRESENTATION

**Case 1** : A 6-month-old child with inward deviation of both eyes noticed by the parents since 3-month-old. There is no history of trauma and systemic disease. Visual acuity (VA) was central steady maintaining (CSM) fixation. Anterior and posterior segment examination showed normal. Cycloplegic refraction revealed +4.00 D in right eye and +4.50 D in left eye. There is no improvement in ocular deviation using cycloplegic refraction that indicating a non-accommodative esotropia. Ocular deviation using Krinsky test was more than 50 prism diopter (PD) base out. There is no limitation on ocular movement nor palpebral fissure change during ocular movement.

**Case 2** :A 6-month-old child with inward deviation of both eyes noticed by the parents since 4-month-old. VA was CSM fixation. Anterior and posterior segment examination showed normal. Cycloplegic refraction revealed +2.00 D/ -1.00 D X 180° in right eye and +2.00 D/ -0.50 D X 180° in left eye. There is no improvement in ocular deviation using cycloplegic refraction that indicating a non-accomodative esotropia. Ocular deviation using Krimsky test was 50 PD base out. There was no limitation on ocular movement nor palpebral fissure change during ocular movement.

**Case 3** :A 1-year-old child with inward deviation of both eyes noticed by the parents since 5-month-old. Previously, there is a history of varicella when 4-month-old. VA was CSM fixation. Anterior and posterior segment examination showed normal. Cycloplegic refraction revealed +5.00 D/ -1.50 D X 180° in right eye and +4.50 D/ -1.50 D X 180° in left eye. There is no improvement in ocular deviation using cycloplegic refraction that indicating a non-accomodative esotropia. Ocular deviation using Krimsky test was 45 PD base out. There is no limitation on ocular movement nor palpebral fissure change during ocular movement.

All patients were subjected to general anestesiawith total intravena anesthesia (TIVA)during the procedure. A unit of BTX-A in 0.1 mililiter was injected bilateral simultaneously through the intact conjunctiva into the medial rectus muscle, until the eyeball moves into the medial direction that indicating the location of the injection was correct.Botulinum toxin type Awas injected in 90° into the muscle and the needle was removed after 60 seconds have passedto minimized the spread ofBTX-Ato surrounding tissues.The patient's head position was held in an upright position during injection and for one hour after injection.

There is no complication was noted in all cases on first day after injection. Two weeks after injection, two patients developed exotropia (Case 1 and Case 2), and one patient developed ptosis (Case 3). Four months after injection, all the cases showed improvement in ocular alignment (respectively 15 PD base out, 15 PD base out, and 30 PD base out) with no other complication (Figure 1).Evaluation of ocular alignment in six months after injection is the further plan for these patients.



**Figure 1.** The clinical outcome.

### 3. DISCUSSION

Botulinum toxin type A serves on muscle nerve endings by blocking calcium transport, which is not all muscle fibres of the motor unit lose their function concurrently. There is no presynaptic release of acetylcholine, thus muscle fibres do not contract. Once the muscle is paralysed its antagonist becomes stronger, as a result change and provide stable ocular alignment.<sup>4</sup>Clinical trials using botulinum for the treatment of strabismus have shown this agent to be most effective in the following conditions, such as small to moderate angle esotropia and exotropia. In very large angle esotropia, it has been shown that surgery (bilateral medial rectus recession) with bilateral simultaneous medial rectus muscle intra-operative botulinum toxin type A injection into the operated muscle is better than surgery alone.<sup>6</sup>In this case series, all the patients had more than 45 PD of ocular deviation. However, four months after injection, the ocular alignment was improved and no other ocular nor systemic complications were noted. The aim of this procedure is to achieve good ocular alignment to ensure the binocular single vision. The timing of surgical intervention is still a controversial. In congenital esotropia, surgery can lead to permanent

deviation of one eye, which may cause amblyopia. The child often develops exotropia later in life, probably because of the permanent anatomic alterations. Otherwise, BTX-A injection can minimize the angle of deviation for surgical intervention. The use of BTX-A in congenital strabismus carries a success rate of 45% or higher. Therefore, we suggest BTX-A injection in these patients. The use of BTX-A in children is still controversial, as it requires sedation and some complications, as temporary ptosis and vertical strabismus, may be more frequent. Nevertheless, BTX-A is a good alternative, as it is a less invasive procedure than muscle surgery.<sup>4,6,7</sup> In case 3, complication of temporary ptosis was obtained and no systemic effects were observed all the cases, which is also consistent with the literature.

#### 4. CONCLUSION

Botulinum toxin type A injection in case of congenital esotropia represents a secure alternative procedure with excellent result and temporary side effect without systemic complication. Botulinum toxin type A injection can reduce the angle of deviation before surgical interventions were planned.

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