



Extended frontalis orbicularis oculi muscle flap shortening for treating refractory apraxia of eyelid opening associated with blepharospasm

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Abstract

Background: Refractory apraxia of eyelid opening (AEO) is mostly unresponsive to botulinum toxin (BTx) and inevitably leads to functional blindness. To treat this challenging condition, an innovative surgical technique was proposed.

Methods: The extended frontalis orbicularis oculi muscle (FOOM) flap shortening consisting of frontalis suspension, partial myectomy, and myotomy in situ of eyelid protractors was applied to treat refractory AEO associated with blepharospasm. The postoperative outcomes and patient satisfaction were evaluated.

Results: Seven patients (mean ages 64.1 ± 3.9 years) of 14 eyelids in total had an average flap shortening distance of 24.4 ± 1.3 mm. During a mean follow-up of 31.6 ± 11.4 months, the average BTx dosage reduced from 58.6 ± 12.1 units to 30.0 ± 8.2 units, with a mean injection interval decreasing from 2.3 ± 0.5 months to 4.1 ± 0.9 months ($p < 0.05$). Palpebral fissure height increased from 1.4 ± 0.5 mm to 7.9 ± 0.7 mm, and the disability scale decreased from $78.8\% \pm 7.2\%$ to $12.6\% \pm 7.0\%$ ($p < 0.05$). The postoperative BTx dosage and frequency were significantly reduced. All patients restored voluntary eyelid opening and reported high postoperative satisfaction (average Likert scale 4.6 ± 0.5).

Conclusion: Extended FOOM flap shortening is an effective treatment to solve refractory AEO associated with blepharospasm.

Keywords: Apraxia of eyelid opening; Blepharospasm; Botulinum toxin; Frontalis orbicularis oculi muscle flap; Frontalis suspension

1. INTRODUCTION

Apraxia of eyelid opening (AEO) is defined as the inability to open the eyelids voluntarily with sustained brow elevation in the absence of obvious blepharospasm.^{1,2} Although the etiology and pathophysiology of AEO are poorly understood, idiopathic focal dystonia is the most common cause.¹ Other causes, infection, toxic exposure, autoimmune, neurodegenerative, neuromuscular diseases, and ocular/central nervous system lesions have all been reported.¹⁻⁶ AEO is most commonly associated with blepharospasm and is occasionally seen in patients with Parkinson's disease or other movement disorders,^{7,8} which is caused by prolonged involuntary pretarsal orbicularis oculi muscle (OOM) contractions, involuntary levator palpebrae inhibition, or a combination of the two.⁹ Tawfik and Dutton¹⁰ proposed that a spasmodic

contraction of the muscle of Riolan may be the etiological basis for levator inhibition. Sleep-induced AEO that occurs only on awakening from sleep is a rare entity.^{11,12} Several studies have found that the imbalance of dopamine neurotransmission is linked to abnormal eyelid muscle contraction.^{1,13}

With AEO being identified as the most common cause for botulinum toxin (BTx) therapy failure in blepharospasm, refractory AEO can even cast severe socioprofessional impact and inevitably lead to functional blindness. The overall prevalence of AEO was approximately 7% to 10.8% in the general population of blepharospasm,^{14,15} but the incidence was as high as 88% in the patients suffering from blepharospasm refractory to BTx.¹⁵⁻¹⁸ In the previous literature, radical myectomy¹⁶⁻³⁰ or myotomy in situ combined with partial myectomy of eyelid protractors¹⁶ were performed separately from frontalis suspension in patients who sustained concomitant refractory blepharospasm and AEO.^{14,31-34}

We herein proposed a novel technique "the extended frontalis orbicularis oculi muscle (FOOM) flap shortening" for treating refractory AEO associated with blepharospasm. This is a previously unreported integrative procedure that combines frontalis suspension, partial myectomy, and myotomy in situ of eyelid protractors. The long-term postoperative outcomes as well as patients' quality of life (QOL) were assessed.

2. METHODS

2.1. Study design

We retrospectively reviewed the patients who had received extended FOOM flap shortening between April 2013 and June

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Video content is available on the journal website at <https://journals.lww.com/jcma>.

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2018. These patients were diagnosed with refractory AEO associated with blepharospasm. Cases with incomplete data or follow-up courses lasting less than 18 months were excluded. The study was approved by the Institutional Review Board [KMUHIRB-E(II)-20180282] and adhered to the tenets of the Declaration of Helsinki. All patients completed the written informed consent prior to surgery, and the presented photographs and videos were provided in accordance with patient written consent. The surgeries and perioperative evaluations were performed by a same experienced surgeon (C.-S.L.).

2.2. Surgical technique

The operations were performed under either general or local anesthesia. The FOOM flap with 20 to 24 mm in width was designed lateral to the supraorbital notch as described in our previous reports.³³⁻³⁶ Hydrodissection of the subcutis from the upper eyelid margin to the upper border of the eyebrow was performed with the injection of 2% lidocaine in 1:100 000 epinephrine using a 26-gauge needle, the redundant eyelid skin with its underlying OOM measured by pinch test was resected first (“partial myectomy”) (Fig. 1A and Video 1). Biplane dissections of the subcutaneous and submuscular layers were proceeded superiorly beyond the upper border of the eyebrow with meticulous hemostasis by bipolar electrocauterization. Anatomically, the upper margin of the orbital OOM was approximately located in the upper border of the eyebrow. The orbital septum was maintained intact except for the need of fat bag removal. Bilateral perpendicular cuts of the flap through the upper border of the orbital OOM (“myotomy in situ”) was done to form a superiorly-based extended FOOM flap. With gentle downward traction of the extended FOOM flap by using a muscle clamp, any tethered fibers were dissected until the flap was free for downward traction. The flap was inferiorly advanced and anchored to the mid-portion of the upper one-third of the tarsus with 6-0 nylon sutures. The flap serves as a suspension force that lifts the upper eyelid (“frontalis suspension”). The desired height of the upper eyelid margin was placed at the upper border of the limbus in supine position, or 1 mm below the upper border of the limbus in sitting position.²⁵⁻²⁷ The excess FOOM flap distal to the anchored line was trimmed off (“partial myectomy”). The upper eyelid crease

was subsequently performed with 6-0 nylon sutures, and the wound was closed with continuous suture (Fig. 1B and Video 1). The details of the operation are demonstrated in the Video 1.

Covering the eyes with moisture wrap dressings immediately after operation and during sleep is essential for postoperative care of lagophthalmos. Eye lubricants and artificial tear solution are routinely applied to alleviate eye irritation. The patients were encouraged to wear the sunglasses to prevent photophobia postoperatively.

2.3. Clinical evaluations and follow-up

All patients were followed up in outpatient clinic in “neurology and plastic surgery departments.” BTx (type A, Botox; Allergan, Inc, Irvine, CA) injection was done by neurologists who referred cases for surgery based on patients’ AEO severity and response to the treatment. After patients underwent extended FOOM flap surgery, BTx injections was resumed 4 weeks later when the eyelid swelling was mostly relieved. The dose started at half of the BTx dose which prescribed preoperatively. The interval was prolonged to 4 months per injection. Of note, to minimize BTx effect on frontalis suspension and levator complex function, BTx injection to the extended FOOM flap of the upper eyelid is avoided. Instead, postoperative BTx is injected into lower lid OOM, upper orbital OOM (except the flap), corrugator, and procerus.

Perioperative assessments were done and documented by an experienced plastic surgeon, including palpebral fissure height (PFH), the therapeutic dosage and injection interval of BTx, as well as the length and the width of flap shortening were recorded. Functional disability measured by a scoring scale that contained six items—reading, watching television, household activities, mobility, driving, and working for each patient—was documented, with the score of each item ranging from 0 to 4, representing the degree or percentage of disability from “no disability” to “activities impossible”.³⁷ The total disability scores ranged from 0 to 24, but the item “working” was not applicable in some patients due to their retirement or freelance status. Therefore, the denominator score was 20 in some patients without a professional status.¹⁶ The preoperative and postoperative

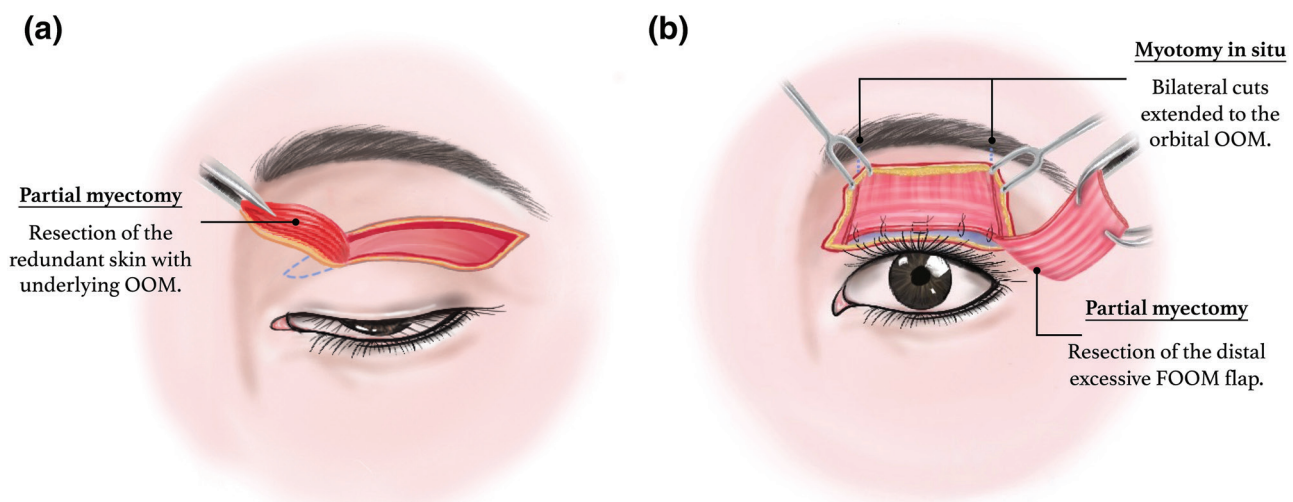


Fig. 1 Illustration of the surgical procedure of Extended frontalis orbicularis oculi muscle flap shortening. A, The redundant skin with underlying OOM is resected along the eyelid crease. B, Extending the bilateral cuts of the FOOM flap into the upper end of the orbital OOM, the flap is advanced and anchored to the mid-portion of the tarsus so as to lift the upper eyelid margin at the upper border of the limbus in supine position, and any distal excessive FOOM flap is resected. FOOM = frontalis orbicularis oculi muscle; OOM = orbicularis oculi muscle.

data were analyzed using paired Student *t* tests. Patients' satisfaction scores of the postoperative outcomes were subjectively assessed by the 5-point Likert scale ranging from highly unsatisfied to highly satisfied.

3. RESULTS

Seven patients with 14 eyelids including five females and two males received the extended FOOM flap surgery. The patients aged between 60 and 72 years (average 64.1 ± 3.9 years). During the procedures, the shortened length of the FOOM flap (partial myectomy) was 24.4 ± 1.3 mm on average (range, 22-26 mm), and the width of the flap base was 22.0 ± 1.6 mm (range, 20-24 mm). The postoperative follow-up period ranged from 20 to 49 months with a mean interval of 31.6 ± 11.4 months. The average PFH improved from 1.4 ± 0.5 mm preoperatively to 7.9 ± 0.5 mm postoperatively ($p < 0.05$) (Table 1). No hematoma, seroma, infection, or scar contracture was recorded. All postoperative lagophthalmos following frontalis suspension with the extended FOOM flap shortening subsided within 6 months after surgery. The preoperative and postoperative average treatment dosage of BTx were 58.6 ± 12.1 units and 30.0 ± 8.2 units, with a mean injection interval of 2.3 ± 0.5 months, and 4.2 ± 0.9 months, respectively. Both the dosage and frequency of BTx significantly reduced after surgery ($p < 0.05$) (Table 1). The functional disability scale markedly decreased from $78.8\% \pm 7.2\%$ preoperatively to $12.6\% \pm 7.0\%$ postoperatively ($p < 0.05$). QOL improved from severe to minimal or no disability after surgery. All these patients have restored voluntary eyelid opening and highly satisfied with the surgical outcomes (Likert scale 4.6 ± 0.5) (Fig. 2 and Video 2) (Table 1).

4. DISCUSSION

AEO is the most common cause of unsuccessful BTx treatment in blepharospasm patients. Frontalis suspension was first introduced to treat blepharospasm by Roggenkämper and Nüssgens³⁸ in 1993 and has been adopted to alleviate refractory AEO with a success rate around 70%.^{9,14,31-34,38-42} According to Georgescu et al,¹⁷ radical myectomy of eyelid protractors was performed in patients with refractory blepharospasm in the past decades, with 33% and 44% complete and partial response in refractory AEO, respectively. In recent studies by Lai et al,¹⁸ myotomy in situ with limited myectomy of the eyelid protractors also yielded favorable outcomes. According to previous studies, combining protractor myectomy with BTx treatment is more effective than any single intervention for blepharospasm with AEO.^{17,40,43} Tozlovanu et al⁴⁴ described that an abnormal persistence of OOM activity, detectable electromyographically but not clinically, could be the primary cause in AEO. Combining the fundamental concepts of these methods and findings, the integrative technique of extended FOOM flap shortening including frontalis suspension partial myectomy, and myotomy in situ of eyelid protractors was used to treat refractory AEO associated with blepharospasm.

FOOM flap advancement is derived from the traditional frontalis muscle flap³⁶ and the orbicularis muscle flap,⁴⁵ and is anatomically characterized by the interdigitation of the longitudinally oriented frontalis muscle and horizontally oriented orbital OOM.^{36,45} It is effective in treating severe blepharoptosis with poor levator function.^{37,46} The FOOM flap functions as a dynamic and autologous suspension tissue, reducing the complications associated with exogenous materials, such as infection, rejection, chronic granuloma, or material exposure.³² The bilateral edges of the extended FOOM flap are completely cut through the upper border of the orbital OOM, similar to the effect of myotomy in situ, whereas only partial cuttings of the orbital OOM are performed in traditional FOOM flap

Table 1
Demographics of patients with refractory AEO associated with blepharospasm receiving extended FOOM flap shortening

Patient No.	Sex	Age	Follow-up (mo)	Flap shortening		Flap based width (mm)	PFH (mm) preop/postop (F/L)	BTx dose (unit) (preop/postop)	BTx duration (mo) (preop/postop)	Disability scale preop DP/postop DP (DS/TS)	Likert scale
				distance R/L lid (mm)	distance R/L lid (mm)						
No. 1	M	62	28	25/25	22	(2/2)/(8/8)	40/20	2/3	70.8% (17/24)/8.3% (2/24)	4	
No. 2	F	63	30	24/24	20	(1/1)/(7/7)	60/30	2/4	75% (18/24)/12.5% (3/24)	5	
No. 3	F	65	24	22/22	20	(2/2)/(8/8)	50/20	2/4	80% (16/20)/20% (4/20)	5	
No. 4	F	62	46	24/24	22	(1/1)/(9/9)	60/40	3/6	90% (18/20)/0% (0/20)	5	
No. 5	M	60	24	26/26	24	(1/1)/(8/7)	80/40	2/4	70.8% (17/24)/12.5% (3/24)	4	
No. 6	F	65	20	24/24	22	(2/2)/(7/8)	60/30	3/4	80.0% (16/20)/15.0% (3/20)	4	
No. 7	F	72	49	26/26	24	(1/1)/(8/8)	60/30	2/4	85.0% (17/20)/20.0% (4/20)	5	
Mean ± SD		64.1 ± 3.9	31.6 ± 11.4	24.4 ± 1.3	22.0 ± 1.6	1.4 ± 0.5/7.9 ± 0.7	58.6 ± 12.1/30.0 ± 8.2	2.3 ± 0.5/4.1 ± 0.9	78.8% ± 7.2%/12.6% ± 7.0%	4.6 ± 0.5	
<i>p</i>						<i>p</i> < 0.05	<i>p</i> < 0.05	<i>p</i> < 0.05	<i>p</i> < 0.05		

AEO = apraxia of eyelid opening; BTx = botulinum toxin; DP = disability percentage; DS = disability score; F = female; FOOM = frontalis orbicularis oculi muscle; L = left; M = male; PFH = palpebral fissure height; postop = postoperative; preop = preoperative; R = right; TS = total score.



Fig. 2 A 62-y-old woman (patient 4 in Table 1) suffered from blepharospasm for the past 9 y. A, Refractory AEO unresponsive to BTx injection for the past 3 y. B, Application of an elastic band over the forehead would temporarily enable her to maintain a narrow vision, but undesirable ectropion of upper eyelids could easily occur from this maneuver. C, In addition to the procedure of extended FOOM flap shortening, lateral tarsoplasty with a pentagonal resection of 4-mm of tarsus was performed to prevent postoperative ectropion due to the coexisting floppy eyelid syndrome. D and E, Excellent functional and aesthetic results were observed 3 y and 10 mo after surgery, and voluntary eye opening was completely recovered (D). Eye closing without lagophthalmos was observed (E). AEO = apraxia of eyelid opening; BTx = botulinum toxin; FOOM = frontalis orbicularis oculi muscle.

shortening. The function-preserving FOOM flap was designed to eliminate postoperative lagophthalmos by preserving most OOM.⁴⁷ However, most OOM is deconstructed to weaken the blepharospasm in the presented technique for refractory AEO. The average amount of partial myectomy performed during the flap shortening procedure was $22.0 \pm 1.6 \text{ mm} \times 24.4 \pm 1.3 \text{ mm}$ (flap base width \times flap shortening distance). Frontalis suspension with partial myectomy and myotomy in situ of eyelid protractors demonstrated the effectiveness in alleviating refractory AEO in this report. It is important to note that frontalis suspension supplied by the extended FOOM flap shortening technique acted in synergism with BTx injections, thereby significantly reducing the dosage and frequency of BTx required postoperatively.

In light of restoring functional blindness to voluntary eyelid opening, the extended FOOM flap has shown significant clinical improvements in the PFH and QOL of these patients after a long-term follow-up period, as evidenced by the effectively reduced dosage and frequency of BTx needed for postoperative maintenance.

The limitation of our study is the noncomparative study design as a case series with a relatively small patient number. In the treatment trajectory of refractory AEO associated with blepharospasm, prospective studies with larger case numbers are expected.

In conclusion, the extended FOOM flap shortening, which consists of frontalis suspension, partial myectomy, and myotomy in situ of eyelid protractors, is effective for treating refractory AEO associated with blepharospasm. All patients regained voluntary eyelid opening, the dosage and frequency of BTx required for postoperative maintenance were significantly reduced in a long-term follow-up.

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