Late-onset inverse Bell’s phenomenon after upper eyelid trauma

ABSTRACT

Bell’s phenomenon evaluation is an important part of preoperative patient assessment for eyelid surgeries. Inverse Bell’s phenomenon is a rare manifestation, usually observed in the early postoperative period following ptosis restoration surgeries, in pathological conditions, and in a small proportion of normal population. In the current case report, a 6-year-old girl presented with late-onset posttraumatic inverse Bell’s phenomenon, 6 months after facial trauma with posttraumatic lagophthalmos. Rehabilitative blepharoplasty for the correction of lagophthalmos led to the restoration of inverse Bell’s phenomenon 3 months postoperatively. Potential causative mechanisms are discussed for this late-onset manifestation of inverse Bell’s response.

Keywords: Bell’s phenomenon, eyelid trauma, inverse Bell’s phenomenon, lagophthalmos

INTRODUCTION

Bell’s phenomenon is a reflex of upward and slightly outward movement of the eyes on attempted eyelid closure. Examination of Bell’s phenomenon is an important part of preoperative patient assessment for eyelid surgeries, since the lack of an adequate Bell’s response increases the risk for corneal exposure in the event of postoperative lagophthalmos. Inverse Bell’s phenomenon constitutes an uncommon variation with hypotropia, instead of hypertropia, found in pathological conditions such as Bell’s palsy, tabes dorsalis, and conjunctival scar, as well as in 2% of normal cases.

An inverse Bell’s phenomenon following ophthalmic surgery has been reported by several previous studies at the early postoperative period, in most cases reverting to normal within 1–4 weeks. The authors herein report an unusual late manifestation of inverse Bell’s phenomenon in a case of posttraumatic lagophthalmos, reverting to normal Bell’s response following the surgical correction of lagophthalmos.

CASE REPORT

A 6-year-old girl was referred 10 days after left upper eyelid and eyebrow laceration, caused by the involvement in a car accident. The facial trauma had been treated with skin and subcutaneous sutures. Previous systematic and ocular histories were unremarkable. On clinical examination, the patient presented with left lagophthalmos, and normal Bell’s phenomenon on voluntary eyelid closure [Figure 1a]. A right IV palsy was also noted, attributed to head trauma, resulting in left-sided ophthalmic torticollis [Figure 2], which necessitated right inferior oblique weakening surgery. Two months later, lagophthalmos was still present, superficial punctuate keratopathy was noted, and Bell’s phenomenon was weakened to a barely noticeable response [Figure 1b]. Artificial tears for corneal protection were prescribed. However, at the 6-month posttraumatic...
interval, the corneal surface displayed localized haze along the interpalpebral fissure [Figure 3]. The degree of lagophthalmos was unchanged and an inversed Bell's phenomenon appeared, along with mild central peaking of the upper eyelid, probably due to fibrotic effects associated with wound healing [Figure 1c]. Surgical correction of lagophthalmos was performed with full-thickness skin grafting to the ipsilateral upper eyelid (the graft was harvested from the ipsilateral preauricular area). During follow-up, eyelid closure was improved with gradual restoration of abnormal Bell’s phenomenon, 3 months after the rehabilitative blepharoplasty [Figure 1d].

### DISCUSSION

Inverse Bell’s phenomenon is a responsive globe hypotropia during voluntary eyelid closure. It has been well documented by several reports to occur in the early postoperative period following levator resection surgeries or occasionally after frontalis sling suspension for surgeries for congenital upper eyelid ptosis. All ten cases described so far reverted spontaneously to a normal Bell’s response between 1 and 4 weeks postoperatively [Table 1]. One possible cause of this transiently altered reflex may be the inflammatory reaction and associated edema of the tissues between the eyelid and the superior rectus.

In another study, the authors evaluated the correlation between the amount of levator resection with Bell’s phenomenon recovery. They reported that 2 out of 32 patients after large levator resection displayed inverse Bell’s phenomenon. In both cases, Bell’s phenomenon was restored to normal along with the recovery of postoperative lid edema and ecchymosis. The authors comment on an additional possible causative mechanism to the one
| References                        | Cases reported | Diagnosis                                                                 | Evaluation                        | Type of surgery                  | Time period to reverse | Complication                                                                 |
|----------------------------------|----------------|---------------------------------------------------------------------------|-----------------------------------|----------------------------------|------------------------|-----------------------------------------------------------------------------|
| Gupta et al\(^{[2]}\)            | 1              | Basal cell carcinoma upper eyelid                                         |                                    | Excision left a wide coloboma on the lateral two-third | Inversed during 2-month follow-up | Inverse Bell's phenomenon, inability to close eyelids, and irritation        |
| Betharia and Kalra\(^{[7]}\)     | 1              | Congenital complicated partial ptosis with epicanthus and antimongoloid slant of palpebral fissures | Levator function: RE=4 mm          | Levator resection                | 12 days               | Inverse Bell's phenomenon                                                  |
| Betharia and Sharma\(^{[4]}\)    | 3              | Residual congenital ptosis (after levator resection 3 years ago)           | Levator function: LE=3.5-4 mm      | Levator resection                | 1 week                 | Inverse Bell's phenomenon and lagophthalmos                                |
| cannons and Sharma\(^{[4]}\)     | 3              | Congenital ptosis                                                         | Levator function: RE=6-7 mm        | Levator resection                | 2 weeks               | Inverse Bell's phenomenon                                                  |
| Cannons and Sterling\(^{[6]}\)   | 1              | Residual congenital ptosis (after frontalis sling with fascia lata 1 year ago) | MRD1: RE=0 mm, LE=1 mm, levator function: 12 mm both eyes | Levator resection                | 4 weeks               | Inverse Bell's phenomenon in both eyes after last surgery and exposure keratitis in both eyes |
| Shitole et al\(^{[8]}\)          | 1              | Congenital ptosis                                                         | MRD1: RE=-1 mm, levator function: RE=8 mm | Levator resection                | 2 weeks               | Inverse Bell's phenomenon                                                  |
| Kumar and Agarwal\(^{[9]}\)      | 1              | Congenital ptosis                                                         | MRD1: RE=-2 mm, LE=-2 mm, levator function: RE=3 mm | Frontalis sling suspension        | 2 weeks               | Inverse Bell's phenomenon, excessive lacrimation, lagophthalmos, and early corneal epithelial defect |
| Goel et al\(^{[10]}\)            | 2              | Congenital ptosis                                                         | MRD1: 1 mm, levator function: 5 mm | Levator resection                | 11 days               | Inverse Bell's phenomenon, ecchymosis, and lagophthalmos                  |
| Pandey et al\(^{[10]}\)          | 1              | Congenital ptosis                                                         | MRD1: 0 mm, levator function: 4 mm  | Levator resection                | 8 days                 | Inverse Bell's phenomenon, eyelid edema, and lagophthalmos                 |
| Morawala et al\(^{[10]}\)        | 1              | Congenital ptosis                                                         | MRD1: LE=2 mm, levator function: LE=8 mm, Marcus-Gunn jaw winking (extrusion 5 mm) | Levator resection                | 1 week                 | Inverse Bell's phenomenon, mild central peaking of the upper lid, mild circumcorneal conjunctival congestion, and diffuse superficial punctuate keratopathy |

MRD1: Margin reflex distance of the upper eyelid from the corneal light reflex, RE: Right eye, LE: Left eye
previously described, associated with abnormal connections between fourth and seventh cranial nerve nuclei, resulting in the infraduction of the eyes from the combined action of the superior oblique and inferior rectus muscles.\textsuperscript{[5,10]} A potential neuro-ophthalmological, instead of mechanistic, origin of a paradoxical inverse Bell’s reaction (with alteration in trigemino-oculomotor nerve and paradoxical eye movements) is also supported by another report of a patient with a wide coloboma on the left upper eyelid following surgical excision of a basal-cell carcinoma.\textsuperscript{[2,11]}

In our case, the inverse Bell’s phenomenon was noted 6 months after the upper eyelid and eyebrow laceration. To our knowledge, late-onset inverse Bell’s phenomenon is a finding not previously reported. In the patient presented in this report, it may reflect an increased infraductive equivalent innervation due to the contralateral IV palsy, possibly associated with juvenile increased brain plasticity. However, the restoration of a normal Bell’s response following the successful surgical treatment of lagophthalmos also supports the concept of an abnormal connection between fourth and seventh cranial nerves.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardian has given his consent for images and other clinical information to be reported in the journal. The guardian understands that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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