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Successfully Modified Dillman-Anderson Myectomy for Marcus Gunn Jaw Winking Ptosis Repair: A Case Report

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Abstract

Introduction: Marcus Gunn jaw winking ptosis (MGJWP) is a type of ptosis present from birth which is characterized by winking of the ptotic eyelid during jaw movement. Case illustration: A 6-years-old child came with a complaint of a right eyelid drooping since birth. The eyelid droop was not progressive. Patient had difficulty to read at a distance, since two years ago. The eyelids moved when the patient chews, sucks, or moving jaw from side to side. Examination did not demonstrate any apparent ptosis of either eyelid nor the movement of eyelids when chewing. On examination of ptosis status on right/left eye, MRD1:0/3, MLD:1/5,LA:4/12 and FIP:6/10.Modified Dillman and Anderson's myectomi with removal of levator muscle above whitnall's ligament followed by frontalis suspension using autogenous fascia lata was performed successfully on this patient. Discussion: MGJWP is ptosis in one eye since birth which causes the ptosis eyelid to wink due to provocation of the same side pterygoid muscle. The jaw winking movement is caused by mouth movements such as opening, moving the mouth to the right and left, chewing and sucking. Modified Dillman and Anderson's myectomi with removal of levator muscle followed by frontalis suspension was chosen because it has a better outcome. In the postoperative period, there were no more complaints of ptosis and the jaw winking was resolved. Serious postoperative complications of this surgical technique such as entropion, worsening jaw winking and exposure keratopathy were not found. Conclusion: For severe MJGWP , Modified Dillman and Anderson's myectomy technique gives satisfactory results in both ptosis correction and jaw winking without serious complications.

Keywords: *Ptosis, Marcus Gunn Jaw Winking, Levator Resection, Autogenous Fascia Lata, Frontalis Suspension*

INTRODUCTION

Synkinesis is a coordinated contraction of several muscles innervated by different nerves or peripheral branches of the same nerve, which occurs simultaneously or in a succession (Prakash MV, et al, 2002) Dr. Robert Marcus Gunn reported a special ptosis case since birth in a 15-year-old girl in 1885. The child's condition featured an associated blinking motion of the afflicted eyelid on the movement of the jaw. Ptosis that is neurogenic and congenital is known as the Marcus Gunn phenomenon (MGP). Also referred to as pterygoid-levator synkinesis or Marcus Gunn jaw winking trigemino-oculomotor synkinesis, it has been seen in 2-13% of patients with congenital ptosis and is equally common in males and females (David, D., et al, 2021).

This condition is believed to be caused by a congenital defect in the branch of the fifth cranial nerve to the branch of the third cranial nerve responsible for the contraction of the levator muscle. Jaw movements such as smiling, sucking on a drink, chewing food, lateral mandibular movements, sternocleidomastoid contractions, protrusion of the tongue, Valsalva technique, or even breathing can cause the affected eyelid to lift or even retract in Marcus Gunn Jaw Winking Ptosis (MGJWP) (Ziga, N., et al,2019). When the jaw moves, the ptotic, previously lower upper eyelid rises to a higher position than the normal eye. In complete MGJW, the eyelid covers half of the cornea at rest (partial ptosis). With each jaw movement, there is a rapid winking movement of the ptotic eyelid (Cavuoto, K. M.,et al, 2020). When the patient wants to look up, they usually open their mouth, causing the eyelid to rise. When the mouth is opened, the eyelid shifts upward and then falls back into the ptotic position even though the mouth remains open. Winking ptosis of the jaw is almost always sporadic, but can be bilateral in rare cases. Familial inheritance of MGJWS has been reported (Kannaditharayil, D.,et al, 2015).

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Strabismus is linked to MGJWS in 50–60% of instances. 5 to 25% of cases are anisometropia. Thirty to sixty percent of patients with MGJWS experience amblyopia, which is nearly invariably owing to anisometropia or strabismus and very seldom results from ptotic eyelid occlusion. As people age, the disease tends to become less visible. Firstly, strabismus and amblyopia need to be treated. Surgical procedure to excise the levator in the eye affected by ptosis and frontalis brow suspension surgery may be required in extreme cases. Because suspension in one eye has a high risk of asymmetry, some eye surgeons recommend frontalis suspension in both eyes to prevent it (David, D., et al, 2021).

When ptosis in MGJWP results a severe impairment in functional and also cosmetic, surgical treatment may be explored. The levator resection, Fasanella-Servat operation, levator excision with frontalis suspension, and bilateral levator muscle excision with bilateral frontalis suspension are among the surgical approaches that have been proposed. Patients with mild to severe ptosis who still have good levator muscle function can undergo levator excision, but jaw winking is not immediately resolved (Ning, Q., et al, 2019). We report the successful treatment of Marcus–Gunn jaw-winking ptosis by frontalis suspension surgery using autogenous fascia lata after removal of the aponeurosis and terminal levator muscle.

CASE ILLUSTRATION

The patient came with the complaint that right eyelid down. Right eyelid drooping felt since birth. Complaints of drooping right eyelid did not get worse. The patient's parents complained that the patient had difficulty reading at a distance, when he started school 2 years ago. The patient's parents also felt that the patient's eyelids moved when chewing. The patient was examined by an ophthalmologist at Mojokerto Hospital 3 months ago, then referred to Doctor Soetomo Hospital. There is no previous medical history, no history of allergies, no history of glasses, similar complaints in the family are denied. Patient was born at term with a normal vaginal birth, birth weight 2900 grams. The patient immediately started crying. History of trauma at birth was denied, history of using forceps or vacuum was denied. History of illness during pregnancy denied.

On examination, visual acuity of the right eye was 1/16-1 with pinhole correction 2/32 with logmar scale. Left eye visual acuity 5/6.5 with pinhole correction 5/5. Right eye pressure and left eye pressure was normal palpation. Hirschberg's test in both orthotropic eyes. On examination of the right and left eyelids, the reflex margin was 0 mm and 3 mm. The limbus margin was 1 mm and 5 mm. Levator action right eye 4 mm and left eye 12 mm. The intra-palpebral fissure in the right was 6 mm and the left was 11 mm. There is a lid crease in both eyes. Bell's phenomenon both eyes are good. Normal head position. There was no lower eyelid retraction in both eyes. The patient also found Marcus Gunn Jaws Winking in the right eye.

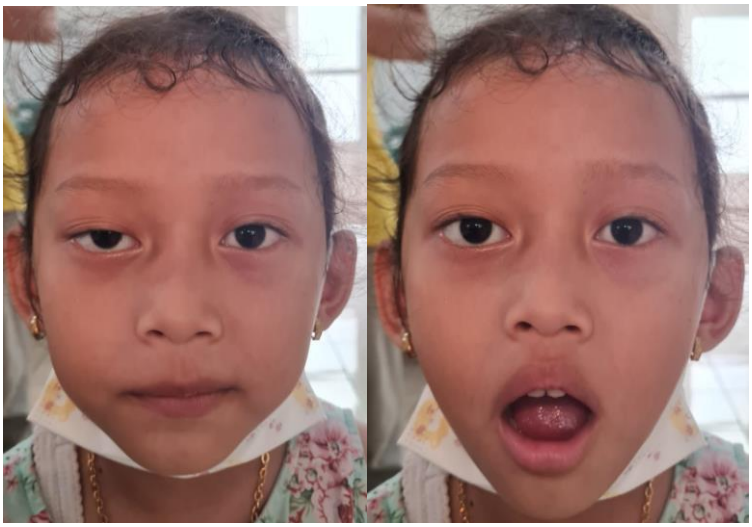

Figure 1.

In the conjunctiva of both eyes there were no signs of infection, secretion and hyperemia. Clear corneas were found in both eyes. In both eyes, the pupils were round, 3 mm in diameter, positive light reflex, and no relative

afferent pupillary defect was found. The lenses of both eyes are clear. on fundoscopic examination of the right and left eyes found in both eyes the fundus reflex was positive for the two nerve heads with normal color limits. There was no bleeding or exudate in the retina of both eyes. Positive macular reflexes were also found in both eyes. The patient was planned to undergo with Dillman-Anderson levator resection technique and frontal suspension surgery under general anesthesia.

The marker was made in the area of the lower third of the right lateral thigh (drawing a straight line from the lateral condyle of the right patella to the anterior superior iliac spine, the marker was made 5 cm above the lateral condyle for 7 mm in the direction of the anterior superior iliac spine).

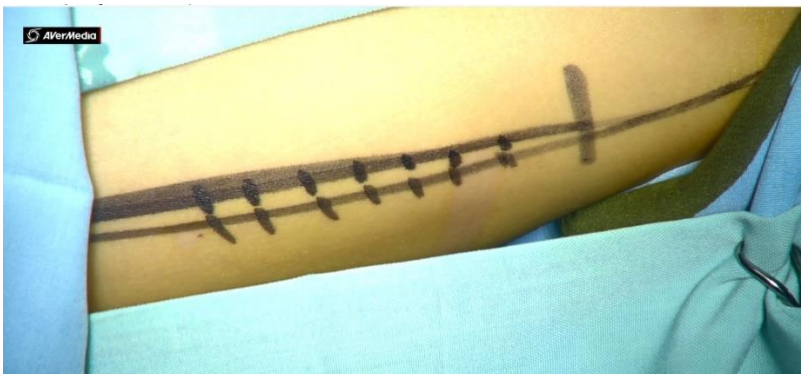

Figure 2.

After disinfection, pehacain was injected subcutaneously and a 5 cm long incision was made with blade number 15.

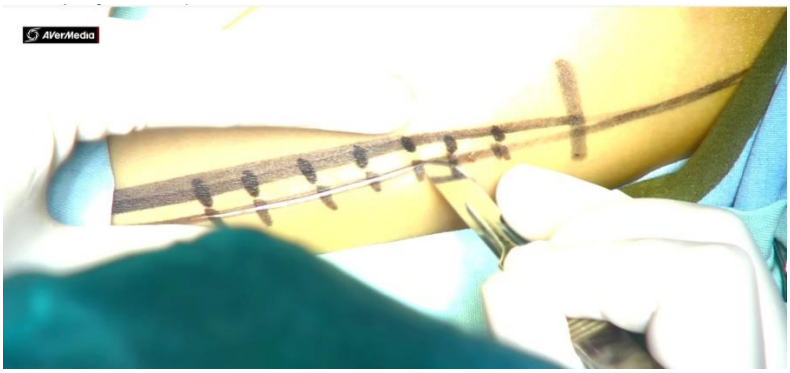

Figure 3.

It was undermined until the fascia lata was found.

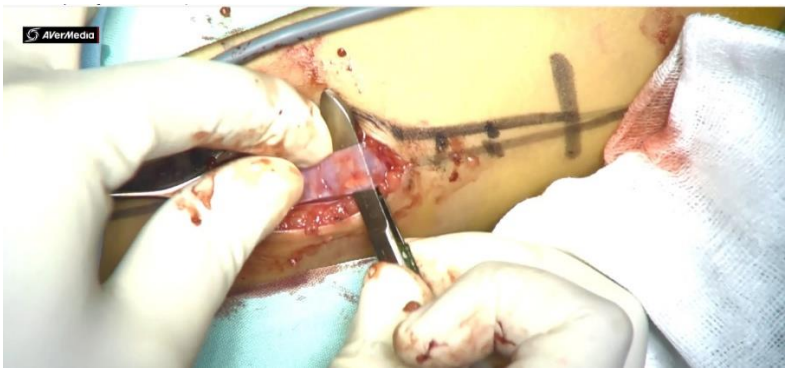

Figure 4.

A 3.5 x 1.0 cm fascia lata incision was made. Suture fascia lata defect with vycril 3-0, subcutaneously with vicryl 3-0. sewing leather with nylon 3-0.

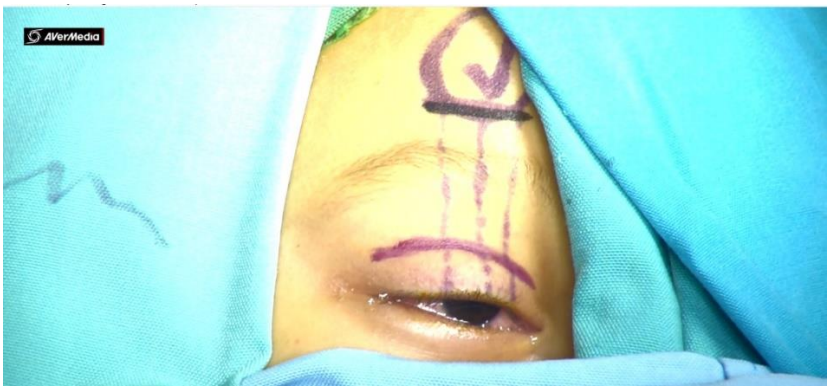

Figure 5.

Make a frontal sling marking. make an incision on the superior lid with a mesh (at the previously marked lid crease area). undermine and identify tarsus.

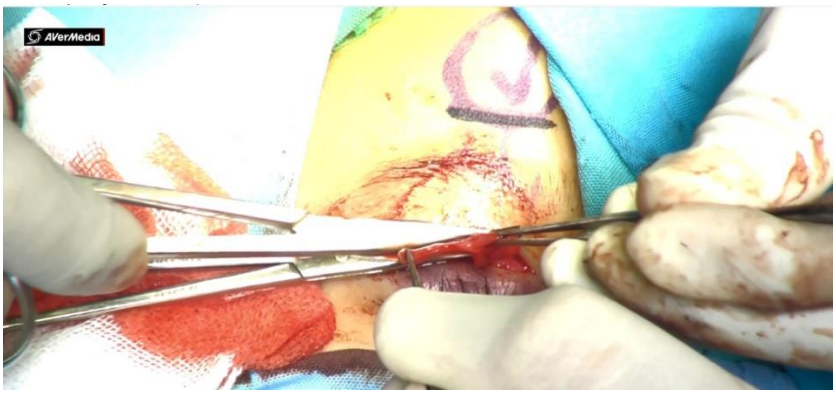

Figure 6.

Separate the levator muscle aponeurosis from the tarsus and conjunctiva by blunt dissection. The levator muscle was cut above the Whitnall ligament. suturing the palpebral conjunctiva with Vicryl 6-0

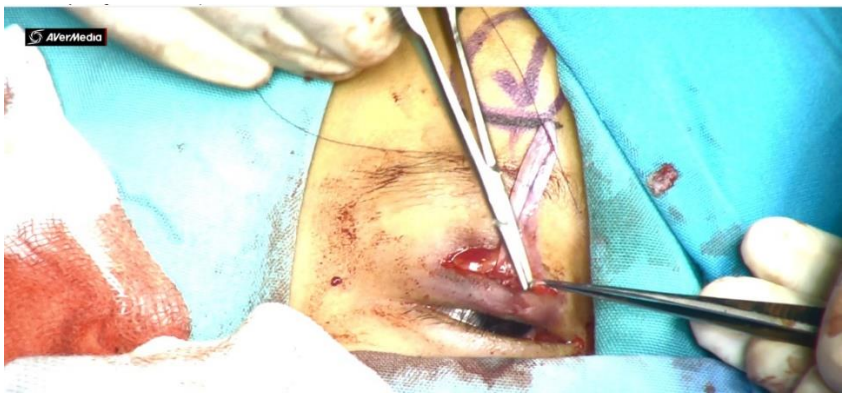

Figure 7.

The fascia lata is sutured to the tarsus. Subcutaneous pehacaine injection is performed at the marker 1 cm above the eyebrow. An incision is made at the marker using a scalpel. Bleeding is controlled using cautery.

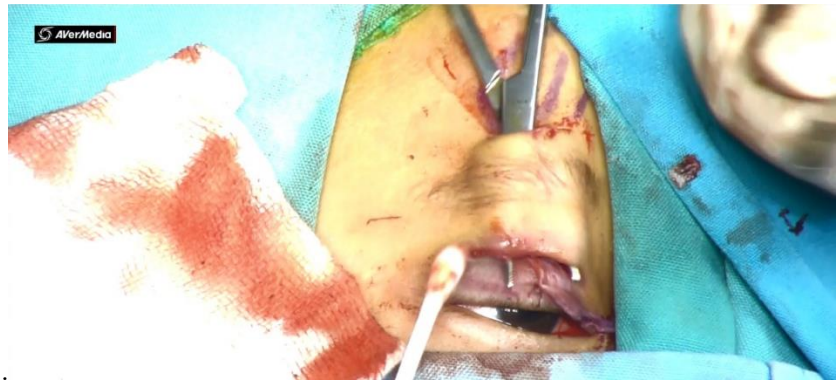

Figure 8.

Undermines from the superior brow towards the palpebral superior (creating a tunnel).

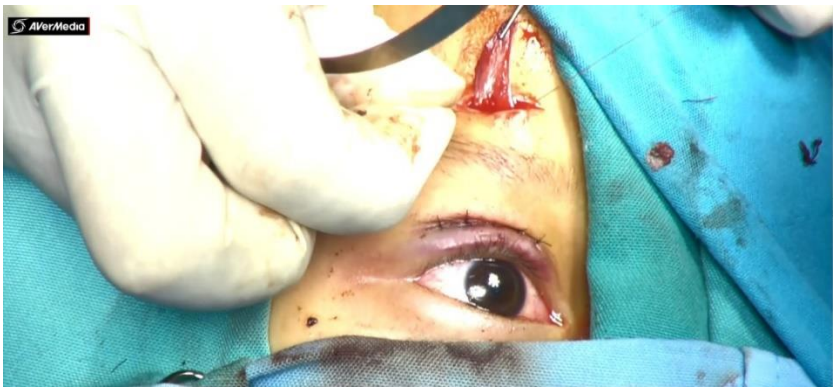

Figure 9.

The fascia lata is pulled through the tunnel to the upper eyebrow until the superior eyelid margin is high enough to evaluate the superior limbus.

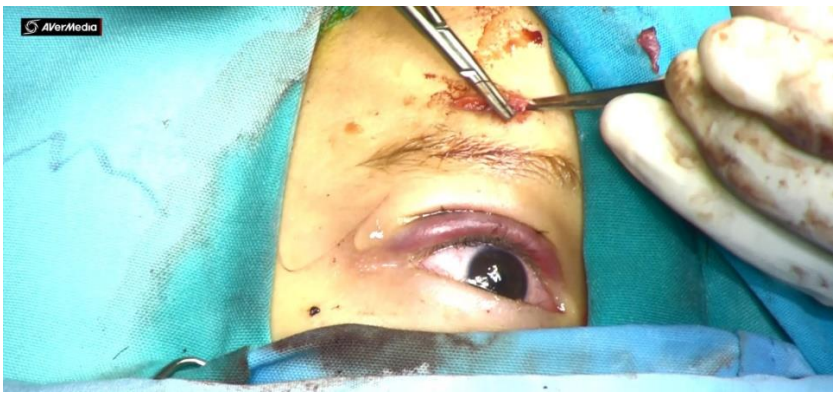

Figure 10.

Stitching was performed on the eyelid crease with 6-0 nylon. Then the fascia lata was stitched to the frontal muscle with 5-0 vicryl. Then the skin of the incision area on the upper eyebrow was stitched with 6-0 nylon.

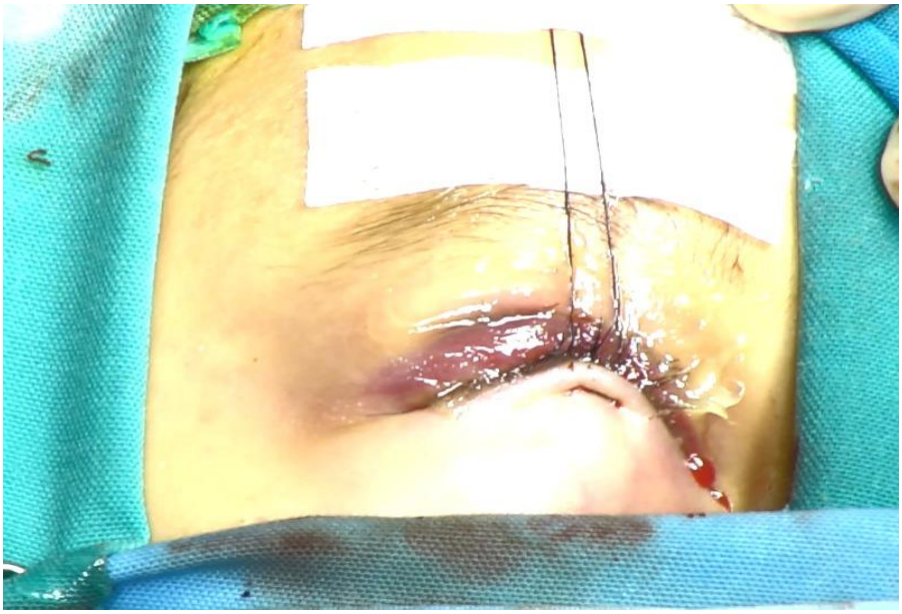

Figure 11.

Frost sutures were performed on the lower eyelid. The patient was discharged from the hospital the next day after surgery.

DISCUSSION

The seven structural layers of eyelids are as follows: tarsus, conjunctiva, muscles of retraction, orbital septum, skin and subcutaneous connective tissue, muscles of protraction, and orbital fat (Cochran, M. L., et al, 2019; Korn, B.S.,et al, 2019). Ptosis, or inferior displacement of the upper eyelid, is also known as blepharoptosis. It is frequently the cause of loss of reversible peripheral vision. While the superior visual field is typically impacted, central vision may also be compromised (Shahzad, B., & Siccardi, M. A., 2023). Reading becomes more difficult for many ptosis patients since their ptosis gets worse when they downgaze. Ptosis can also impair visual acuity because it reduces the total amount of light that reaches the macula. Ptosis can be categorised as either involutional or acquired and congenital or since birth (Shukla, U. V., & Patel, B. C., 2021). Ptosis can also be classified based on its cause into myogenic, aponeurotic, neurogenic, mechanical, or traumatic (Sabatini, S.L., & Erna,R., 2016). The most prevalent kind of acquired ptosis is aponeurotic, which is brought on by stretching or disinsertion of the levator aponeurosis; the most common type of congenital ptosis is myogenic, which is brought on by a poorly developed levator palpebrae superioris (LPS) muscle (Bacharach, J., et al, 2021).

Synkinesis is coordinated contraction of several muscles innervated by various nerves or different peripheral branches of the same nerve, occurring simultaneously or in a succession. One commonly known synkinesis in congenital ptosis is the Marcus-Gunn jaw winking (Joshi, M., et al., 2014) Congenital ptosis results from LPS muscle dysgenesis, a condition in which fat and fibrous tissue replaces healthy muscle fibres. Other names for MGJWS are congenital cranial dysinnervation disorder (CCDD), pterygoid-levator synkinesis, Marcus-Gunn phenomena, and maxillopalpebral synkinesis (Sathish, S., et al, 2023).

Chewing, suction, lateral mandibular movement, smiling, sternocleidomastoid contraction, projecting tongue, Valsalva technique, and even breathing might cause the afflicted eyelid to elevate or even retract in MGJWS (Ziga, N., et al,2019). Although in the literature the incidence of MGJW on the left side of the face has been reported to have a prevalence, several studies have found that the incidence of MGJW on both sides of the face is almost the same, or sometimes MGJW on the right side of the face has a slightly higher frequency (Dżaman, K., et al, 2019).

The etiopathogenesis of MGJWS is still very poorly understood by experts. It is believed that the occulomotor nerve that innervated the levator palpebra is the origin of an aberrant branch of the trigeminal nerve that was

misdirected since birth (Ghodasra, D. H., et al, 2009). Premature babies do not have a higher risk of developing MGJWS. Cases of MGJWS in babies born at 37-42 weeks of gestation were reported, which is also the case in our report (Koelsch, E., & Harrington, J. W., 2007). However, cases of MGJWS have also been reported in premature patients (Kassem, I. S., & Kodsi, S. R., 2009).

The most widely used theory to explain MGJWP is the concept of aberrant connections of the nerves innervating the levator palpebrae. The location of these aberrant connections can vary as follows (Freedman, H. L., & Kushner, B. J., 1997):

Cortical or sub cortical connections

Internuclear connections or faulty distribution in the posterior longitudinal bundle

Infranuclear connection exists between motor branches of the trigeminal nerve (CN V3) innervating the external pterygoid and the fibers of superior division of the oculomotor nerve (CN III) that innervates the levator muscle of the upper lid

Peripherally - some CN V fibers may reach the levator via the auriculo-temporal nerve.

Some authors speculate that it may be caused by facial nerve trauma, which results in aberrant growth branches to the mandibular branch. On the other hand, some authors think that rhythmic jerking of the eyelid is stimulated by excitation of the cranial third nerve, which innervates the levator palpebrae superioris on the ipsilateral side, when the trigeminal nerve innervates the pterygoid muscles in response to stimulation (Demirci, H, et al, 2010; Alam, M. S., et al, 2020).

There has only ever been one case of unilateral jaw-winking ptosis linked to syphilis. The 1937 case report details the patient's recuperation from a unilateral oculomotor palsy following antisyphilitic therapy. This infection may have caused abnormal connections in the midbrain or peripheral nerves, or it may have been the cause of an atavistic release event (Johnson, A, et al,2023).

There are gaps in the mechanism's illustrations. Two primary theories exist. The first one declared the abnormal relationship between the third cranial nerve and the mandibular ramification of the fifth cranial nerve. Studies using electromyography have validated this idea. Proprioceptors located within the pterygoid muscle of affected patients give abnormal signals to the oculomotor nucleus in the midbrain. This nucleus and the trigeminal nerve nucleus may have an aberrant link that is triggered in utero (Shah, A.D, et al,2012; Kannaditharayil, D.,et al, 2015).

According to the second one, MGP is revealed to be a primitive pathway. As many as twenty percent of healthy individuals contract their LPS muscle in response to stimulation of the motor root of the fifth cranial nerve in the brainstem (Lehman A.M., et al., 2014) This also explains why ptosis occurs due to decreased innervation of the third cranial nerve, and this is also supported by histological studies showing neurogenic atrophy of the levator muscle in both eyes in affected individuals (Dżaman, K., et al, 2019).

In addition to olfactory issues, MGJWP may be a component of congenital abnormalities such as cleft palate and lip. CHARGE syndrome has been documented by certain writers, particularly in relation to bilateral MGP (Alshamrani, A. A., et al, 2019). As a result, every patient with an MGS diagnosis needs to be evaluated to rule out deafness, ear abnormalities, or choanae atresia. Dzaman et al. validated the six odours test to check children's olfactory function(Dżaman, K, et al, 2013).

Jaw-winking ptosis is a rare occurrence that mostly affects one side, however it can occur on both, usually on the left. Early diagnosis is typical, mostly based on carer descriptions of the synkinetic movement that occurs during feeding. This syndrome may remain undetected until adolescence in certain situations. A range of degrees of lid elevation has also been noted, as evidenced by the movement of the upper eyelid, which is triggered by the levator palpebrae and ipsilateral external pterygoid muscles. As people age, the condition known as jaw-winking ptosis usually gets better. However, the patient's persistent jaw tightness makes gains invisible(Wong, J. F., et al, 2001). In fact, patients learnt to limit or conceal the illness over time by being aware of the trigger movements and learning how to avoid them (David, D., et al, 2021). The patient came with the

complaint that right eyelid down. Right eyelid drooping felt since birth. The patient's parents complained that the patient had difficulty reading at a distance, when he started school 2 years ago. The patient's parents also felt that the patient's eyelids moved when chewing.

A thorough clinical examination includes measures such as visual acuity to rule out amblyopia, examination of pupillary condition and reflexes, cycloplegic refraction examination should be performed to determine whether there is anisometropia, fundus evaluation, examination of extraocular eye muscle motility, examination of cover test to determine whether there is paralysis of the double elevator or superior rectus, examination of Bell's phenomenon whether it is adequate or decreased due to MGJW because this condition can reduce it, and paying attention to the position of the head because children are often accustomed to lifting the chin to be able to see better without being obstructed by the drooping eyelid due to ptosis (Park, D. H., et al, 2008). After briefly occluding the eyes to interrupt the fusion, the jaw should be immobilised in a central position in order to measure the degree of ptosis. There are three categories for ptosis: mild (≤2 mm), moderate (≤3 mm), and severe (≥4 mm) (Wong, J. F.,et al, 2001; Senthilkumar, V. A., & Tripathy, K., 2020).

On examination of the eyes, right visual acuity was 1/16-1 with pinhole correction 2/32 and left visual acuity was 5/6.5 with pinhole correction 5/5. Both eyes had normal intraocular pressure. On examination of the anterior segment, the right eye reflex margin was 0 milimeter and the left eye was 3 mm. Limbus margin for the right and left eye is 1 mm and 5 mm, respectively. Levator action was 4 mm and 12 mm. Intra-palpebral fissure was 6 mm and 11 mm. Lid crease was found in both eyes. There is no Bell's phenomenon on both eyes. The head position in our patient was normal. There was no lower eyelid retraction in both eyes. Finally, Marcus Gunn Jaws Winking was found in the right eye.

The distance between the upper and lower eyelids is very important to measure because it is the largest gap between the eyelids called the vertical palpebral fissure. Marginal reflex distances 1, 2, and 3 (MRD1, MRD2, MRD3), are examined with the eyes in the primary gaze position and are determined by the examiner and patient aligned at the same level. The corneal light reflex to the central upper eyelid margin is measured using MRD1. The corneal light reflex to the central lower eyelid margin is measured using MRD2. MRD3, which measures the amount of LPS resection in ptosis surgery, is when the patient looks upwards maximally and very high, the distance from the light reflex formed in the ocular to the middle edge of the upper eyelid is measured. Levator function is a measure of the distance between the lids in a relaxed position compared to when the eyes gaze maximally upward and the brow is raised. When the patient does this, the frontalis muscle will contract maximally along the eyebrows. The levator function grading system includes normal if greater than or equal to 15 millimeters, good if between 12 and 14 millimeters, fair if between five and eleven millimeters, and poor if equal to four millimeters or less. The presence of eyelid lag should be measured along with examination of the eyelid position during downward gaze (Senthilkumar, V. A., & Tripathy, K., 2020).

Developmental abnormalities of the extraocular muscles and anomalies of innervation were considered in the differential diagnosis of congenital ptosis. Anomalies related to the development of extraocular muscles include a number of conditions, including Duane's syndrome, MGJWS, and the inverse Marcus Gunn phenomenon (Senthilkumar, V. A., & Tripathy, K., 2020). Congenital ptosis caused by innervation abnormalities may be brought about by neurologic disorders, levator muscle neuromuscular junction failure, or sympathetic nervous system malfunction (Marenco, M., et al, 2017). The range of cranial dysinnervation syndromes, which are defined by deficiencies in the development of different cranial nerves or nuclei, may be associated with the MGS (Graeber, C. P., et al, 2013; Gutowski, N. J., & Chilton, J. K.,2015). Additionally, anomalies of the eyes and other diseases such spina bifida, ectrodactylies, and undescended testicles may coexist with MGP (Dżaman, K., et al, 2019). Other synkinetic disorders that have manifestations and may be associated with eyelid ptosis are Martin-Amat syndrome (Myers, K. A., et al, 2016). Inverse Marcus Gunn phenomenon causes illnesses and Ptosis was observed in Martin-Amat syndrome, which is more noticeable when the patient moves their mandible. Further congenital condition where the levator muscle of the eyelid is affected called Marcus Gunn Phenomenon. After facial paralysis, if the function of the levator muscle and orbicularis oculi is not compounded, acquired Martin-Amat syndrome occurs. Furthermore, the presence of ptosis may indicate Horner's disease or myasthenia gravis (Odehnal, M., & Malec, J., 2002).

Table 1. Differential diagnosis for Marcus Gunn jaw-winking syndrome (Johnson, A, et al,2023).

	Marcus Gunn jaw-winking ptosis	**Inverse Marcus Gunn jaw-winking syndrome**	**Marin–Amat syndrome**	**Duane syndrome**
Physical examination	Synkinetic contraction, specifically eyelid raising, with jaw movement	Synkinetic blinks with mouth closing	the eyes will close due to blepharospasm when smiling or opening the mouth	When the eye adducts, there is narrowing of the palpebral fissure (and sometimes other associated features)
Proposed etiology	Aberrant innervation in midbrain or peripheral cranial nerves	Aberrant innervation in midbrain or peripheral cranial nerves	Aberrant innervation between trigeminal and facial nerves	Functional disorders abducens motor nucleus and nerve with aberrant innervation of lateral rectus by oculomotor nerve

When a child has modest congenital ptosis without significant refractive defects, surgery should only be done for cosmetic purposes and after considering the psychological effects on the child. On the other hand, the most severe cases necessitate a surgical procedure, which involves frontalis brow suspension and unilateral levator excision. It has been suggested that bilateral frontalis suspension prevent imbalance brought on by unilateral suspension. Gene therapy could be a revolutionary way to replace defective genes with a healthy copy of the gene (Marenco, M.,et al,2017).

Surgery for MGJWP remains controversial at present. Because general anesthesia is not safe in infancy, some advocate delaying surgery until age 1–2 years (Feng, Y. P., et al,2020). If surgical treatment of ptosis is performed without addressing the synkinetic winking associated with jaw movement, the patient's abnormal eyelid movements may worsen, which may be cosmetically disfiguring. Surgery is indicated for correction of severe ptosis with normal levator function, in the presence of amblyopia, and in the presence of vertical strabismus. Relative contraindications to surgery for MGJWS include reduced corneal sensitivity and poor Bell's phenomenon, as well as dry eye. Exposure keratitis, a visual-threatening disorder, may arise in several clinical situations (Koka, K., & Patel, B. C.,2019)

The severity of ptosis and jaw winking movement must be taken into account when planning jaw winking ptosis surgery. According to Doucet and Crawford, a jaw-wink of two millimetres or more was deemed to be aesthetically important. It is best not to proceed with surgery for minimal degrees of ptosis or the winking movement of jaw is not noticeable from a cosmetic standpoint. Procedures of choice for correcting minor ptosis include a Fasanella-Servat operation, a Muller muscle and conjunctival resection, or a typical external levator resection (Karabulut, G. O., et al,2019). Regardless of the degree of ptosis, frontalis suspension followed by obliteration of the levator's activity is the recommended course of treatment if the winking movement of jaw is considered to be bothersome or in moderate to advanced severity conditions (Bair, H., et al, 2019).

Levator resection surgery for the treatment of ptosis with jaw winking movement in moderate to advanced severity conditions generally has less than satisfactory results because it does not resolve the problem of jaw winking movement. Even after surgery, the jaw winking movement is even more obvious. Levator excision is a classic technique that has been chosen for use by many eye surgeons. There are several surgical techniques on the eyelid that have been suggested by experts to eliminate the function of the levator palpebra. Dryden et al. performed a transposition of the proximal levator muscle toward the marginal arch after transection above Whitnall's ligament. Bullock suggested a total surgical removal of the entire aponeurosis and muscle of the levator toward the orbital apex. Dillman and Anderson performed an operation to isolate the levator muscle above Whitnall's ligament and also performed a partial muscle removal. The total levator muscle removal procedure is a major surgical procedure that carries several hazards to the superior orbital tissues, including the potential for disruption of the nerve supply to the two rectus muscles (superior and superior oblique). Blind dissection of the underlying superior rectus muscle is required to perform a levator muscle transection above Whitnall's ligament or, if necessary, a myectomy. Furthermore, it has been documented that even after a levator transection above Whitnall's ligament, jaw winking is still complained of by patients (Mandal, S. K., et al, 2021). The patient was planned to undergo with Dillman-Anderson levator resection technique and frontal suspension surgery under general anesthesia. Finally, the patient was discharged from the hospital after receiving one day after surgery.

Marcus Gunn jaw winking syndrome is best treated surgically, functionally, and cosmetically using the unilateral posterior technique (Whitnall's ligament approach), especially in younger patients. There is no chance of needing significant surgery on both sides. The primary gaze facial photo identity was widely acknowledged in official government documents following the procedure. Unlike the conventional approach of downgaze similarities, this study places emphasis on bilateral lid height similarity in primary gaze. The patients reported great emotional and physical satisfaction (Mandal, S. K., et al, 2021).

Surgical follow-up is necessary every two to four weeks to detect complications as described below (Sweeney. A.R., et al, 2024). The Marcus Gunn jaw-winking (MGJW) has a tendency to be associated with strabismus (50–60%) and anisometropia (5-25%) as associated sequelae. Amblyopia affects 30 to 60% of people and is typically a subsequent condition to anisometropia or strabismus. It is necessary to look at the coexistence of these problems, particularly in those whose syndrome is discovered throughout adolescence. Before considering surgical care of the jaw wink, any related conditions—especially strabismus and amblyopia—should be treated (David, D., et al, 2021).

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