Journal of Scientific Innovations in
Anesthesiology

REVO PUBLICATION

Case Report

Balancing sedation and responsiveness with dexmedetomidine–ketamine in pediatric marcus gunn jaw-winking syndrome: A case report

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Abstract

Marcus Gunn Jaw Winking Syndrome (MGJWS) is a rare congenital condition characterized by unilateral ptosis associated with synkinetic elevation of the upper eyelid during jaw movements. Patients with moderate-to-severe MGJWS often require surgical correction for cosmetic improvement and relief of visual axis obstruction. Intraoperative assessment of eyelid symmetry necessitates intermittent patient arousal, which poses a significant anesthetic challenge in pediatric patients. Additional anesthetic considerations include maintaining child cooperation, alleviating anxiety, ensuring airway safety, providing adequate analgesia and optimal sedation and the risk of the oculocardiac reflex. There is a paucity of literature regarding anesthetic management in pediatric patients with MGJWS necessitating intermittent arousal. We describe the successful anesthetic management of a child undergoing ptosis correction with sling suspension using procedural sedation with dexmedetomidine and ketamine combined with a supraorbital nerve block. This technique enabled repeated, smooth awakening, effective analgesia, stable cardiorespiratory parameters, satisfactory patient cooperation, and high surgeon satisfaction.

Keywords: Dexmedetomidine, Ketamine, Marcus Gunn phenomenon, Nerve block, Pediatrics.

Received: 15-06-2026

Accepted: 22-08-2026

Published: 02-11-2026

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https://doi.org/10.65929/JSIA.2026.1.3.007

How to cite this article: Kaasat A, Thakore S, Yadav P, Thakore N. Balancing sedation and responsiveness with dexmedetomidine–ketamine in pediatric marcus gunn jaw-winking syndrome: A case report. *J Sci Innov Anesthesiol.* 2026;1(3):95-99.

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1. Introduction

Marcus Gunn Jaw Winking Syndrome (MGJWS) or trigemino–oculomotor synkinesis, was first reported by the ophthalmologist Marcus Gunn in 1883.[1] The incidence of MGJWS among individuals with congenital ptosis is approximately 2% to 13%.[2]

MGJWS presents with unilateral congenital ptosis and coordinated synkinetic movement of the upper eyelid and the jaw. Etiopathogenesis of MGJWS is not well defined, the likely underlying mechanism is abnormal connection between the motor branches of the trigeminal nerve and superior division of oculomotor nerve.[3]

Most cases of MGJWS are congenital; acquired forms have been reported following ocular surgery, trauma, syphilis, and pontine tumors.

Patients with moderate to severe cases of MGJWS need surgical intervention for cosmetic issues and visual axis obstruction. The standard treatment consists of partial excision of levator muscle and fascia lata sling suspension surgery can be performed under either general anesthesia or local anesthesia. To conduct real-time evaluation of symmetry of eyelids, surgeons require

patients to be easily arousable during the operation. This can be easily achieved in most adult patients with surgery under local anesthesia. However intermittent awakening is challenging in pediatric population.[4] There is a paucity of literature regarding anesthetic management in pediatric patients with MGJWS necessitating intermittent arousal.

Recently a newer sedoanalgesic combination of dexket (dexmedetomidine and ketamine) has been used in pediatric population for procedural sedation.[5] The Dexket offers synergistic benefits for sedation and analgesia, balancing their individual effects.

In the present case, we report a successful anesthetic management for sling suspension and ptosis correction surgery under procedural sedation using combination of dexmedetomidine and ketamine with supraorbital nerve block in a pediatric patient with MGJWS.

2. Case Report

A seven-year-old male, weighing 30kg known case of MGJWS presented with left blepharoptosis and eye blinking associated with jaw movements (**Figure 1**). There was no history of developmental delay, ptosis, or similar complaints among family members, and no history of prior surgery. Pre operative and airway examination was unremarkable. He was posted for staged fascia lata sling suspension surgery.



Figure 1: Preoperative image showing unilateral ptosis with the jaw closed (left) and characteristic jaw-winking phenomenon with elevation of the affected upper eyelid on jaw opening (right).

In the first stage fascia lata was harvested from thigh, constructed as slings and placed in subcutaneous tunnel along with partial excision of the LPS muscle. This critical step abolishes the abnormal "wink" synkinesis by preventing the levator from contracting with jaw movement. This surgery was performed under general anesthesia uneventfully.

In the second stage, the fascia lata slings were attached to the frontalis and levator muscles of the eyelid, and eyelid height was carefully adjusted to achieve symmetry with the contralateral side. Intermittent patient awakening was required intraoperatively to assess eyelid movement, and both the patient and parents were counseled accordingly.

Procedural sedation along with supraorbital nerve block was planned. After taking informed consent from parents, patient was shifted to OT and standard ASA monitors were applied. He was preoxygenated and premedicated with inj. glycopyrrolate 0.1 mg, inj. ondansetron 3 mg, inj midazolam 0.5 mg, inj fentanyl 30 mcg iv. Patient was administered a combination of IV dexmedetomidine (15 mcg) and ketamine (30 mg) diluted with 10 ml normal saline slowly over 2 minutes followed by infusion of dexket (1mg/ml ketamine and 1 mcg/mL dexmedetomidine) at 5-10 mL/hr. The infusion rate was titrated to achieve a sedation score of 2 to 4 on the Ramsay Sedation Scale (RSS) during the surgery.[6]

Five minutes after administration of the dexmedetomidine–ketamine bolus, a supraorbital nerve block was performed using the landmark technique. Following standard skin antisepsis, the supraorbital notch (or foramen) was identified by palpation along the supraorbital margin. A 25-gauge needle was inserted into the subcutaneous plane adjacent to the notch/foramen, and 2 mL of 0.25% levobupivacaine was injected after negative aspiration. (**Figure 2**).



Figure 2: Intraoperative landmark guided supraorbital nerve block

Surgery was started 5 minutes after the block. Patient was awakened twice during procedure on light tactile and verbal stimuli to check symmetry. Patient was arousable, comfortable and cooperative during the procedure (**Figure 3**).



Figure 3: Placement of electrodes for forntalis muscle identification

Patient was maintained on spontaneous respiration with oxygen at 2L/min with nasal cannula, without any episodes of desaturation or airway compromise. End tidal CO_2 varied from 30-35 mmHg with Respiratory rate of 14-16 per minute. Patient remained hemodynamically stable throughout the surgery with HR (80-100bpm), NIBP (90-100/60-70) mmHg.

Intraoperatively, electrodes were placed to identify the frontalis muscle using a nerve mapper. The procedure was completed in approximately 30–35 minutes (**Figure 4**).



Figure 4: Intraoperative image

After the completion of procedure infusion was stopped and patient was awake in 5 minutes, and was shifted to PACU (RSS<2). After 30-minute Aldrete score > 9 was achieved and patient was shifted to ward (**Figure 5**).



Figure 5: Postoperative photograph showing successful correction of ptosis with satisfactory eyelid symmetry

3. Discussion

This case report demonstrated the successful use of dexmedetomidine and ketamine for procedural sedation with supraorbital nerve block in pediatric patient requiring intermittent awakening. This approach facilitated adequate patient cooperation and resulted in high surgeon satisfaction during surgery.

As Marcus Gunn syndrome is an autosomal dominant condition with incomplete penetrance, a detailed family and prior anesthetic history should be obtained to rule out malignant hyperthermia.

Anesthetic implications in MGJWS include ensuring child cooperation, managing anxiety, airway control, optimal sedation, adequate analgesia, risk of oculocardiac reflex[7] and malignant hyperthermia. Balancing sedation for comfort with maintaining patient responsiveness for eye lid symmetry requires precise anesthetic techniques.

Local anesthesia has been safely used in adult patients with MGJWS; however, its use in pediatric patients is challenging.[8] With appropriate conscious sedation and analgesia, surgical procedures can be successfully performed in children.

Tu and Gao successfully used dexmedetomidine along with local anesthesia in a 10-year-old child with MGJWS for levator resection surgery.[9]

Recently a novel sedoanalgesic combination of dexmedetomidine and ketamine has been used in children for PS like UGIE, TTE, minor cardiac procedure, MRI in variable doses etc.[10-13]

Baptista et al used same combination as ours of ketodex with bolus of ketamine (1 mg/kg) and dexmedetomidine (1 mcg/kg), followed by a continuous infusion of ketamine (1 mg/mL) and dexmedetomidine (1 mcg/mL) at 0.2-2 mL/kg/h for a four-year child with a large cervical/facial mass undergoing a diagnostic magnetic resonance imaging.[14]

A limitation of anesthetic management in this case was the unavailability of depth-of-sedation monitoring such as the bispectral index (BIS). Further large scale RCTs are required to evaluate the efficacy in dexket in such surgeries.

4. Conclusion

Procedural sedation with dexket and supraorbital nerve block was seen to be a safe and effective alternative to general anesthesia. It provided adequate analgesia, sedation with intermittent awakening without any cardiorespiratory compromise.

Author Contributions

Ankita Kaasat: Conceptualization, patient management, anesthesia management, literature review, manuscript drafting, critical revision, and final approval of the manuscript.
Sakshi Thakore: Conceptualization, anesthesia management, patient management, clinical interpretation, manuscript drafting, critical revision, and final approval of the manuscript.
Pratibha Yadav: Patient management, data collection, literature review, manuscript review, critical revision, and final approval of the manuscript.
Nirdesh Thakore: Clinical supervision, patient management, clinical interpretation, manuscript review, critical revision, and final approval of the manuscript.

Abbreviations

LPS: Levator Palpebrae Superioris; OT: Operation Theatre; ASA: American College of Anesthesiologists; HR: Heart Rate; MRI: Magnetic Resonance Imaging, NIBP: Non-Invasive Blood Pressure; PACU: Post Anesthesia Care Unit, RSS: Ramsay Sedation Score, UGIE: Upper Gastrointestinal Endoscopy, TTE: Trans Thoracic Echocadiography, RCT: Randomized Control Trial.

Declaration of Patient Consent

Written informed consent was obtained from the patient's parent/legal guardian for publication of this case report, accompanying clinical details, and clinical photographs/images. The parent/legal guardian was informed that appropriate measures would be taken to maintain the patient's confidentiality and anonymity.

Financial Support and Sponsorship

None.

Conflict of Interest

None.

Use of Artificial Intelligence (AI)

The authors declare that no artificial intelligence (AI) tools were used in the generation, analysis, interpretation, or writing of this manuscript.

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