Short report

The Marin-Amat syndrome: an unusual facial synkinesia

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SUMMARY Three cases of facial nerve palsy with abnormal synkinetic movement manifesting with eye closure on jaw opening (Marin-Amat Syndrome) are described. The eye closure occurred only with wide jaw opening and it is felt that the disorder represents aberrant regeneration within the facial nerve with proprioceptive impulses associated with muscle stretch acting as the trigger. Though the movement is the exact opposite of what happens in the Marcus-Gunn phenomenon it is suggested that the term inverse Marcus-Gunn phenomenon be reserved for a congenital lesion with a different pathogenesis.

Bell's palsy is the commonest lesion affecting the facial nerve. A full recovery is seen in the majority1 while in others a variable degree of weakness persists. Other residual deficits described are autonomic phenomena (for example, crocodile tearing and auriculo-temporal syndrome), mass actions and associated movements.2 Observation of an unusual associated movement in three patients following Bell's palsy led to this report and a review of the literature. The condition referred to here as the Marin-Amat Syndrome3 with eye closure on jaw opening is similar to the inverse Marcus-Gunn phenomenona but we believe that the pathogenesis is different.

Case reports

Case 1 A 28-year-old male had suffered from right facial paralysis for two years. Routine laboratory investigations, ENT examinations, skull radiographs and CSF analysis were normal. He was diagnosed as Bell's palsy and was treated with steroids. He showed delayed but good functional recovery. When seen 3 months later he was asymptomatic. Examination revealed mild facial asymmetry (fig. a). Full opening of the mouth led to closure of the eye on the affected side (fig. c) though partial opening of the mouth had no such effect (fig. b). No reciprocal movement was seen on voluntary closure of the eye.

Electrophysiological studies revealed normal conduction in the facial nerve. Muscle action potentials were normal. There were no denervation potentials. EMG studies of facial and masticatory muscles revealed that clenching of the teeth, moving of the jaw from side to side and slight opening of the mouth did not produce any contraction in orbicularis oculi. Active contraction of facial-innervated muscles including platysma had no effect on the eye. It was only when the jaw was opened to its maximum limit, either actively or passively, did the orbicularis oculi muscle contract. Simultaneous EMG studies in the levator palpebrae superioris revealed no change with wide opening of the mouth.

Case 2 A male aged 30 years, who came with respiratory tract infection was noted to have a peripheral facial palsy due to injury sustained near the angle of the jaw three years previously. The facial muscles showed residual weakness. He showed synkinetic movements similar to Case 1: full forcible opening of the mouth caused eye closure on the affected side, while partial opening of the mouth had no such effect. Eye closure did not occur with other movements. EMG showed that firing of units was consistently seen only with wide opening of the jaw and not during slight opening.

Case 3 A 35-year-old lady suffered from Bell's palsy for two years followed by right sided palsy one year later. When seen she had residual facial paresis on the right side. She had anomalous movements and EMG findings similar to Cases 1 and 2.

Discussion

Marin-Amat's syndrome is a rare associated movement, wherein contraction of orbicularis oculi is
brought about by opening of the jaw. This is the opposite of the Marcus-Gunn phenomenon; hence it has also been called Inverted Marcus-Gunn Phenomenon.\textsuperscript{4} Isolated case reports of this syndrome are noted in the literature\textsuperscript{1–3} where it followed peripheral facial paresis. Surprisingly most of the text books of neurology have not mentioned it when discussing facial nerve lesions. Zulch\textsuperscript{2} described closure of eye-lids on the affected side in Bell’s palsy while smiling or whistling as a result of mass movement.

Many views have been put forward to explain the phenomenon.\textsuperscript{2} Orbicularis oculi is richly supplied by branches of the facial nerve which have a connection with branches of the maxillary nerve and buccal branches of the mandibular division of the trigeminal nerve. This peripheral anastomosis led to it being regarded as a trigemino-facial associated movement. This would be in keeping with the view about the more well known Marcus-Gunn phenomenon, where abnormal connection between the nerves to external pterygoid and the levator palpebrae is postulated. Wartenberg,\textsuperscript{7} did not agree with this view and we also do not accept it. The Marin-Amat syndrome is mostly an acquired phenomenon occurring after peripheral facial palsy and in our cases there is no doubt about the site of lesion. To involve faulty regeneration via trigeminal nerve in such cases seems unsatisfactory; we believe that the lesion is due to faulty regeneration within the facial nerve itself. The proprioceptive impulses of muscle stretch of the facial muscles are carried in the facial nerve.\textsuperscript{9} Ascending impulses during full mouth opening and muscle stretching could then trigger contraction of orbicularis oculi owing to faulty regeneration at the site of nerve lesion.

Lubkin,\textsuperscript{10} reported the first EMG study in the ‘Inverse Marcus Gunn Phenomenon’. This patient had a congenital problem. Her father had a similar disorder. When she opened her mouth or moved the jaw to the left, her right eye would close. During EMG studies Lubkin noted that closure of the eye was due to inhibition of the levator palpebrae superioris and not due to orbicularis oculi contraction. Thus, his case was truly an inverse Marcus-Gunn Phenomenon. Whereas the Marcus-Gunn phenomenon is due to contraction of the levator, this case showed inhibition of the levator. Lubkin’s case\textsuperscript{10} was also congenital, like cases of Marcus-Gunn Phenomenon. The cases best called Marin-Amat syndrome are acquired and follow facial paralysis. In our cases the eye closure is clearly due to orbicularis contraction, a feature different from the inverse Marcus-Gunn phenomenon as described by Lubkin. We believe that this differentiation which has not so far appeared in the literature is worthy of mentioning. It is to be noted that neither Marin-Amat\textsuperscript{3} nor other earlier authors were able to show that eye closure was due to orbicularis contraction. We can only note that their cases were clinically similar and use the eponym the Marin-Amat syndrome for this combination. We believe this syndrome will be found more frequently if looked for carefully.

Fig. (Case 1): Showing facial asymmetry on clenching teeth (a) and effect of partial (b) and full (c) opening of mouth on ptosis.
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References


