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OPSOCLONUS-MYOCLONUS-ATAXIA SYNDROME FOLLOWING A FEBRILE ILLNESS IN A TODDLER: A CASE REPORT

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ABSTRACT

Background: Opsoclonus-myoclonus-ataxia syndrome (OMAS) is a rare, immune-mediated pediatric disorder that may follow neuroblastoma or infection, and has occasionally been reported in temporal proximity to vaccination, though causality with vaccination remains unestablished. Diagnostic delay and relapse on corticosteroid tapering can mask the true course; earlier escalation to combination immunotherapy is associated with better outcome. **Clinical description:** A 1-year-11-month-old boy developed gait difficulty, speech regression, opsoclonus, generalized tremor, irritability, and sleep disturbance within 10 days of receiving the DPT booster and second doses of measles-rubella and Japanese encephalitis vaccines, having tolerated the primary doses previously. Examination confirmed opsoclonus, truncal ataxia with titubation, and four-limb action tremor. **Management:** Serial brain MRI, an autoimmune encephalitis panel, abdominal ultrasonography, and whole-body PET-CT were normal, excluding demyelination, encephalitis, and occult neuroblastoma. After relapsing on intermittent oral corticosteroids over six months, he was escalated to intravenous dexamethasone, IVIg, and rituximab, with significant improvement in gait, opsoclonus, and tremor. **Conclusion:** OMAS should be considered in any toddler with sub-acute opsoclonus, ataxia, and developmental regression, even with a negative autoimmune encephalitis panel. Relapse on tapering should prompt escalation to combination immunotherapy rather than repeated steroid courses.

KEYWORDS: Kinsbourne syndrome, dancing eye syndrome, corticosteroid relapse, rituximab, pediatric neuroregression.

INTRODUCTION

Opsoclonus-myoclonus-ataxia syndrome (OMAS), also called Kinsbourne syndrome or “dancing eye-dancing feet” syndrome, is a rare immune-mediated disorder of early childhood characterised by the triad of opsoclonus, myoclonus/ataxia, and behavioural or sleep disturbance, with an underlying neuroblastic tumour identified in up to half of affected children.[1] An international consensus approach recommends a diagnosis of OMAS when three of these four features are present, since no single confirmatory biomarker exists.[1] Misdiagnosis as acute cerebellar ataxia or other encephalopathies is common, and a recent international patient-reported registry of 194 children found that nearly half were initially misdiagnosed, despite an overall median time to correct diagnosis of 25 days, underscoring how delayed recognition can postpone immunotherapy and worsen developmental outcome.[2] Roughly half of non-paraneoplastic cases follow an antecedent infection,[3] and OMAS has occasionally been reported in temporal proximity to vaccination, although a causal relationship with vaccination has not been established in the literature.[4] We report a toddler whose OMAS began within days of receiving multiple routine vaccines and who had a relapsing course on intermittent corticosteroids before referral; thorough screening excluded neuroblastoma, and escalation to combination immunotherapy produced sustained clinical recovery, illustrating both the autoimmune basis of OMAS and the importance of distinguishing a temporal association from causation when symptom onset follows a recent vaccination.

Clinical description

A previously healthy, developmentally normal 1-year-11-month-old male child received the diphtheria-pertussis-tetanus (DPT) booster, the second dose of measles-rubella (MR-2), and the second dose of Japanese encephalitis (JE-2) vaccine at 16 months of age, having tolerated the earlier, primary doses of each of these same vaccines uneventfully in infancy. He developed fever on day one after vaccination, and over the following 10 days went on to develop gait difficulty, regression of speech to monosyllables, abnormal eye movements, and involuntary movements of all four limbs, along with irritability, fearfulness, and disturbed sleep. There was no history of seizures, altered sensorium, cranial nerve involvement, or focal motor or sensory deficit. He was unable to sit, stand, or walk independently because of the involuntary movements affecting all four limbs. Over the following six months, he received intermittent courses of oral corticosteroids elsewhere, with partial symptomatic improvement on each occasion followed by recurrence of symptoms during tapering. He presented to us six months after symptom onset. On examination, he had opsoclonus, characterised by rapid, chaotic, multidirectional conjugate eye movements; truncal ataxia with titubation while sitting with support; and action tremor of all four limbs that worsened with voluntary movement. The remainder of the neurological and systemic examination was unremarkable.

Management and outcome

Given the classical clinical triad and a history of partial response to corticosteroids with relapse on tapering, OMAS was suspected, and a stepwise evaluation was undertaken to exclude structural, demyelinating, autoimmune, and paraneoplastic causes. Magnetic resonance imaging (MRI) of the brain, performed within 2 weeks of his initial presentation and repeated two months later because of concern for a demyelinating disorder, was normal on both occasions. Cerebrospinal fluid and serum autoimmune encephalitis panels, including anti-N-methyl-D-aspartate receptor antibodies, were negative, excluding antibody-mediated encephalitis. Because an occult neuroblastic tumour underlies up to half of paediatric OMAS cases,[1] ultrasonography of the abdomen and a whole-body positron emission tomography-computed tomography (PET-CT) scan were performed; neither revealed a tumour. With structural, demyelinating, and paraneoplastic causes excluded, a clinical diagnosis of OMAS, of probable autoimmune origin, was

made. Given his relapsing course on corticosteroid monotherapy elsewhere, he was escalated to combination immunotherapy comprising intravenous dexamethasone, intravenous immunoglobulin (IVIg), and rituximab (Table 1), rather than a further course of steroid tapering alone, a multimodal regimen recently reported to reduce relapse and long-term neurological sequelae compared with corticosteroid monotherapy.[5,6] Following this regimen, he showed significant clinical improvement, with recovery of gait, resolution of opsoclonus, and reduction in limb tremor.

Table 1: Combination immunotherapy protocol.

Treatment	Month 1 Day1	Day2	Day3	Month 2 Day1	Day2	Day3	Month 3 Day1	Day2	Day3
Intravenous dexamethasone (every month), 20 mg/m²/day OD IV	Dose 1	Dose 2	Dose 3	Dose 1	Dose 2	Dose 3	Dose 1	Dose 2	Dose 3
Intravenous immunoglobulin (every month), 1 g/kg/day for 2 days	Infusion	Infusion	–	Infusion	Infusion	–	Infusion	Infusion	–
Intravenous rituximab (every 3 months), 375 mg/m² IV with premedication	Dose 1	–	–	–	–	–	–	–	–

DISCUSSION

Although classically regarded as paraneoplastic, OMAS is now understood primarily as an autoimmune neuroinflammatory disorder rather than a direct tumour effect. Within neuroblastic tumours associated with OMAS, lymphoid follicles rich in CD20-positive B lymphocytes, CD3-positive T lymphocytes, and follicular dendritic cells have been demonstrated, together with elevated cerebrospinal B-cell-activating cytokines and clinical response to B-cell-depleting therapy; these findings collectively support an antibody- and B-cell-mediated mechanism, even though a single confirmatory pathogenic autoantibody has not yet been identified.[7] In children without an identifiable tumour, an antecedent infection is thought to trigger cross-reactive immunity against shared neuronal and pathogen antigens (molecular mimicry), or bystander activation of pre-existing autoreactive lymphocytes, producing a clinical phenotype indistinguishable from paraneoplastic OMAS.[3] This shared immunopathology explains why immunomodulatory therapy is effective regardless of the presence of an underlying tumour or a clearly identified antecedent trigger.

Reports of OMAS in temporal proximity to immunisation remain few, and in nearly all instances, as in ours, a causal relationship cannot be established. A 2-month-old infant has been reported to develop OMAS after routine vaccination, with recurrence of symptoms during steroid tapering and again following a subsequent vaccination, raising the possibility that vaccine-related immune activation could precipitate or re-trigger disease in some children.[4] Similar isolated reports of OMAS in temporal relation to other childhood vaccines exist in the literature, underscoring that such occurrences are rare and not specific to any one vaccine.[4] However, attributing causation is inherently difficult given the rarity of OMAS relative to near-universal childhood immunisation coverage, the absence of any established biological marker linking a specific vaccine antigen to disease, and the fact that vaccination is frequently accompanied by a minor febrile illness that could equally be the trigger. This difficulty is further underscored in our patient by the fact that he had previously received the primary doses of the same DPT, measles-rubella, and Japanese encephalitis

vaccines without any adverse neurological event, arguing against a simple, dose-independent causal link to these vaccine antigens and supporting a temporal, rather than causal, association in this case. Indian tertiary-care case series have similarly documented vaccination as an antecedent event in a small proportion of children with OMAS,[8] alongside a separate series describing post-infectious OMAS following scrub typhus,[9] reinforcing that an antecedent immunological stimulus, whether infectious or merely coincident with vaccination, is a recognised feature of non-paraneoplastic OMAS, while a true cause-and-effect relationship with any individual vaccine remains unproven.

The single most important determinant of long-term outcome in OMAS is the timeliness of diagnosis and the adequacy of immunosuppression. Diagnostic delay, whether due to misdiagnosis as acute cerebellar ataxia or another acute encephalopathy, or due to symptomatic improvement on intermittent corticosteroids masking an underlying relapsing-remitting course, as occurred in our patient, has been associated with greater residual motor, behavioural, and cognitive sequelae.[2,10] His pattern of partial improvement followed by relapse on tapering is well recognised in OMAS treated with corticosteroid monotherapy and is itself an indication for escalation to combination immunotherapy.[1,5] Recognition of this relapsing pattern, together with completion of neuroblastoma screening, prompted initiation of multimodal therapy with IVIg and rituximab in addition to corticosteroids, consistent with current international recommendations favouring combination regimens over corticosteroid monotherapy to reduce relapse and improve developmental outcome.[1,5] This case illustrates that a relapsing course on corticosteroids should prompt early escalation of therapy, and that a temporal association with vaccination, while worth documenting, should not be assumed to be causal or allowed to delay standard diagnostic evaluation and combination immunotherapy.

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