

There are previous descriptions of the association between X monosomy and JIA in the literature (Table 1). Most articles on the subject correspond to single case reports,¹⁻³ although there are also case series^{4,5} of up to 18 patients.⁶ The largest case series corresponds to a multi-centre study with participation of 28 rheumatology centres in Europe and North America and followup of approximately 15 000 patients with JIA. Based on the number of identified patients, the authors estimated that the prevalence of the association of TS and JIA was six times greater than expected.⁶ A more recent study using the Danish Cytogenetic Central Register that reviewed a cohort of 798 women with TS followed up for 12,461 person-years concluded that these patients had double the risk of developing an autoimmune disease compared to the general population, with a standardized incidence ratio for JIA (ratio of the observed number of cases to the expected number of cases) of 4.4 (Jørgensen et al., *Arthritis Rheum.* 2010;62:658–666).

The presentation of arthritis in JIA may be oligoarticular (up to 4 affected joints) or polyarticular (5 or more affected joints), with variability in the detection of ANA and HLA-B27 antigen and the presence of bone erosion. Additional autoimmune disorders are frequently reported, especially inflammatory bowel disease in HLA-B27-positive patients.

In conclusion, the prevalence of JIA in girls with TS is greater than expected for a random association. This must be taken into account in the evaluation of patients with TS that develop musculoskeletal manifestations suggestive of arthritis (joint swelling, limping or morning stiffness).

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Replacing “Apparent Life Threatening Event” (ALTE) with “Brief Resolved Unexplained Event” (BRUE). A retrospective review of the ALTEs that meet the criteria of a BRUE[☆]



Sustitución de ALTE por BRUE: revisión retrospectiva de los ALTE que cumplen criterios de BRUE

To the Editor:

In 2016, the American Academy of Pediatrics (AAP) published a guideline recommending replacing the term *apparent life-threatening event* (ALTE) with the term *brief resolved unexplained event* (BRUE) with the aim of identifying lower-risk patients (with a low probability of a repeat event or of severe underlying disease) that do not require hospital admission or further investigation.¹

The authors of the guideline considered that a more specific term was necessary because ALTE is a concept

that encompasses a broad range of disorders (from periodic breathing to sepsis), which can generate a feeling of uncertainty in the clinician and thus compel performance of unnecessary tests.² The alternative term, BRUE, is intended to reflect the transient nature and lack of clear aetiology of such events and remove the “life-threatening” notion that is the source of such anxiety. The guideline proposed that the evaluation of each infant should be based on the level of risk of the event and established recommendations for the management of low-risk cases.³

Based on this change, we carried out a study with the aim of retrospectively reviewing the cases of patients admitted with a diagnosis of ALTE that met the criteria for BRUE.⁴ Then, we determined which cases in the group meeting the criteria for BRUE would qualify as higher or lower risk, which would allow us to determine the number of infants in who performance of tests and hospital admission may have been deemed unnecessary. Until May 2018, our hospital had a protocol for management of ALTE that called for admission of all patients with ALTE.^{5,6}

Between January 2013 and December 2017, there were 194 patients with an admitting diagnosis code of ALTE. During this period, there was no diagnosis code for BRUE. We excluded 52 infants for who we did not find data allowing us to determine whether the event would qualify as a BRUE. The final sample included 142 patients, 68 female and 74 male, with a median age at admission of 37 days (range, 2–461 days), 95 of who were aged less than 2 months.

Of the 142 patients, 68 (48%) did not meet the criteria for BRUE because a cause for the event had been identified during the history-taking or physical examination (cold symptoms, fever, vomiting, abnormal breath sounds on aus-

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cultation), the event had not resolved at the time of arrival to the emergency department, or the patient was aged more than 1 year.

Of the 74 cases (52%) that met the criteria for BRUE, 69 (93%) qualified as higher-risk BRUE. The most frequent reasons for classification as higher-risk BRUE were age less than 2 months or corrected age less than 45 weeks (51 cases), repeat event (18 cases), event duration greater than 1 min (16 cases), concerning social assessment (5 cases) and family history of sudden death (4 cases). Several patients met more than one higher-risk criterion. None of the infants was classified as higher-risk based on need for cardiopulmonary resuscitation (CPR) by a trained medical provider.

Of the 5 infants that met the criteria for lower-risk BRUE, 3 underwent diagnostic tests (blood tests, cranial ultrasound, echocardiogram, electroencephalogram). All test results were normal, save for the incidental finding in the echocardiogram of a haemodynamically insignificant aorto-pulmonary collateral arteries and patent foramen ovale in 1 patient. In these infants, there were no abnormalities in the vital signs during the hospital stay, repeat episodes or diagnosis of severe underlying disease. The mean length of stay in these 5 patients was 1.8 days.

In the group of 69 patients that met the criteria for higher-risk BRUE, diagnostic tests were performed in 48% in the emergency department and in 74% during the hospital stay. Ten percent of these patients experienced a repeat event during the stay. Abnormal test results or relevant diagnoses from testing included diagnosis of convulsive seizures in 1 infant, 1 case of congenital hypothyroidism (the results of the newborn screening for metabolic diseases became available during the hospital stay), 1 case of respiratory infection by respiratory syncytial virus, 1 case of meningitis caused by enterovirus and detection of haemodynamically insignificant aorto-pulmonary collateral arteries in 1 infant. The mean length of stay in this group was 2.6 days.

Recent guidelines recommend educating parents on how to perform CPR. Such training was only delivered in 3 cases (2%): all 3 met the criteria for higher-risk BRUE, and 2 were managed with home cardiorespiratory monitoring.

There are limitations to our study, chief of which is its retrospective design.

Only half of the infants admitted due to ALTE met the criteria for BRUE. Most infants that experienced these events were aged less than 2 months, which made them qualify

as higher risk on account of the age criterion. Only 7% of patients with BRUE met the criteria for lower risk BRUE, so testing and hospital admission could only have been avoided in this percentage of the total.

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Home monitoring of sodium in children with adipsic diabetes insipidus*



Monitorización domiciliaria de sodio en niños con diabetes insípida y adipsia

Dear Editor,

Children with diabetes insipidus associated with the lack of thirst (adipsia) may experience severe oscillations in their serum sodium levels requiring frequent and prolonged hospitalizations. On one hand, they are

at risk of hyponatraemia resulting from excessive fluid intake due to absolute or relative overdosage of desmopressin. On the other hand, due to the absence of thirst, they are also at risk of hypernatraemia due to fluid deprivation if lost fluids are not properly replenished.^{1,2}

We present the cases of 3 children with panhypopituitarism with antidiuretic hormone deficiency (central diabetes insipidus) and adipsia as immediate complications of surgical removal of large hypothalamic tumours. After a protracted postoperative hospital stay due to countless episodes of hyponatraemia and hypernatraemia, patients could be discharged home and managed at the outpatient level by providing parents with a portable system for measuring capillary blood sodium levels at home.

The patients were 2 girls and 1 boy aged 5.5, 15 and 9 years, respectively, that had undergone surgery for treatment of craniopharyngioma, optic nerve glioma and mixed germ cell tumour and were hospitalised for

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