

Infantile Cortical Hyperostosis of Scapula Presenting as Pseudoparalysis in an Infant

Caffey disease or Infantile cortical hyperostosis, is a rare disorder of unknown etiology, characterized by cortical hyperostosis with inflammation of contiguous fascia and muscle [1]. It is self-limiting and usually affects young infants. The diagnosis may be delayed as it mimics a wide range of conditions like osteomyelitis, hypervitaminosis A, bone tumor and child abuse. We present an infant with pseudoparalysis, who was initially considered to have osteomyelitis of scapula, but was later diagnosed to have Caffey disease.

A 75-day-old boy with uneventful antenatal and neonatal period, presented to us with complaints of paucity of movement of left upper limb for one week. There was no history of fever, intramuscular injection or trauma, lethargy or seizures. He was not on any vitamin supplementation. On examination, he was irritable. Neurological examination revealed paucity of movement of left upper limb. Deep tendon reflexes were preserved. Detailed general examination showed mild soft tissue swelling over the body of left scapula. Left shoulder movement was painful.



FIG. 1 MRI showing altered signal intensity in the body of left scapula with periosteal reaction.

There was no other bony swelling or tenderness. After consulting Orthopedician, chest X-ray and magnetic resonance imaging (MRI) of left shoulder with upper limb was done. MRI (**Fig. 1**) showed altered signal intensity in the body of left scapula with periosteal reaction and adjacent soft tissue edema, features favoring osteomyelitis. Chest X-ray (**Fig. 2**) showed slightly thickened and sclerotic left scapula suggestive of Caffey disease. Laboratory tests showed leukocytosis, anemia (Hb 8.4 g/dL), thrombocytosis, positive C-reactive protein (25.1 mg/L), and high ESR (20 mm/hr). Blood culture was sterile. Infant was treated with Ibuprofen. Left upper limb movement improved and infant is doing well on follow-up.

Though MRI was reported as osteomyelitis of scapula in our infant, the absence of underlying risk factors for bone infection made us suspect an alternative diagnosis. The presence of irritability, soft tissue swelling and hyperostosis on X-ray helped us clinch the diagnosis. Thrombocytosis has also been reported in Caffey disease [2]. Though Mandible is the most commonly involved bone, scapular involvement presenting as Erb's palsy has been earlier reported [3]. A high index of clinical suspicion is required to diagnose Caffey disease and avoid unnecessary investigations and intervention.

Acknowledgement: Dr S Muralinath for radiology assistance.

***VENKATESWARI RAMESH AND JANANI SANKAR**

Department of Pediatrics, CHILDS Trust Medical Research Foundation, Kanchi Kamakoti CHILDS Trust Hospital, Chennai, India

**venka80@gmail.com*



FIG. 2 X-ray showing showing thickened and sclerotic left scapula.

REFERENCES

1. Horton WA, Hecht JT. Disorders for which defects are poorly understood or unknown. *In: Nelson Textbook of Pediatrics*, 20th edn. Eds. Kliegman RM, Stanton BF, St. Gema JW, Schor NF, Behrman RE. Philadelphia: WB

Saunders Co, 2011. p.3379.

2. Krishnamurthy S, Srinivasan S. Severe thrombocytosis as initial manifestation of Caffey disease in a 4 month old infant. *Pediatr Blood Cancer*. 2012;59:345-6.
3. Holtzman D. Infantile cortical hyperostosis of scapula presenting as Erb's palsy. *J Pediatr*. 1972;81:785-8.

The Conundrum of Optimal Drug Dosing in Obese Children

In 2013, WHO estimated that 42 million children under 5 years of age were overweight, with 75% of them living in developing countries; and the prevalence has increased from 4.2% in 1990 to 6.7% in 2010 [1]. More obese children are being encountered in clinical practice, including those requiring chronic drug therapy. These patients are at risk for iatrogenic drug dose related complications as dosages are based on body weight. We report a case of early onset hypertension following systemic steroid therapy in a 10-year-old obese child with rheumatic carditis.

A 10-year-old boy presented with fever, migratory joint pain, headache and generalized severe myalgia for a week. On examination, he had fever and signs suggestive of congestive cardiac failure. Body mass index (BMI) was 23.63 (>97th percentile). ESR was elevated (90 mm/hr) with a positive CRP and ASLO. Cardiac ultrasound showed rheumatic carditis with left ventricular dysfunction. He was treated with oral prednisolone (60 mg/day as 2 mg/kg crossed the adult maximum dose) [2]. He developed hypertension (BP >95th centile) within 3 days which required intravenous furosemide for 3 days followed by oral nifedipine. Steroids were stopped and aspirin started. His blood pressure decreased progressively to the normal range within 14 days following which nifedipine was discontinued.

Adverse effects of prednisone usually, develops after prolonged use of doses in excess of the normal physiological requirement, often after a week of usage [3]. As our patient presented with early-onset drug-induced hypertension, it probably was iatrogenic due to inappropriate dosage. A thorough literature search revealed the lack of evidence for ideal drug dosing in obese children and no stipulated guidelines [4,5].

Excess body weight alters the pharmacokinetics in overweight and obese children leading to higher risk of toxicity or reduced therapeutic efficacy [4]. There is limited data about the pharmacokinetics and drug dosing in obese children as compared to adults. All children with weight above adult maximum dosage would receive the same dose of drug which, logically cannot be right (For example, three 10-year-olds weighing 35 kg, 45 kg and 55 kg respectively, would receive same adult maximum dosage). The pharmacokinetics of adult maximum doses will certainly vary based on the weight, and can lead to both toxicity and sub-therapeutic doses.

In view of continuing childhood obesity epidemic, there is a need for further research regarding drug dosages in these children. In spite of limited evidence, it is essential to have a practice guideline on drug dosages for obese children.

PODHINI JEGADEESAN AND

***THIRUNAVUKKARASU ARUN BABU**

Department of Pediatrics,

Indira Gandhi Medical College & Research Institute,

Puducherry, India

**babuarun@yahoo.com*

REFERENCES

1. World Health Organization. Childhood Overweight and Obesity, 2013. Available From: <http://www.who.int/dietphysicalactivity/childhood/en>. Accessed July 12, 2015.
2. Kumar RK, Tandon R. Rheumatic fever and rheumatic heart disease: The last 50 years. *Indian J Med Res*. 2013;137:643-58.
3. Liu D, Ahmet A, Ward L, Krishnamoorthy P, Mandelcorn ED, Leigh R, *et al*. A practical guide to the monitoring and management of the complications of systemic corticosteroid therapy. *Allergy Asthma Clin Immunol*. 2013;9:30.
4. Kendrick JG, Carr RR, Ensom MH. Pediatric obesity: Pharmacokinetics and implications for drug dosing. *Clin Ther*. 2015;37:1897-923.
5. Mulla H, Johnson TN. Dosing dilemmas in obese children. *Arch Dis Child Educ Pract Ed*. 2010;95:112-7.