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Hypertrophic osteoarthropathy in a young child with adult respiratory distress syndrome (ARDS) secondary to burns

N. Wadhwa, D. Balsam, P. Ciminera

Department of Radiology, Nassau County Medical Center 2201 Hempstead Turnpike, East Meadow, New York 11554, USA

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Abstract. A case of hypertrophic osteoarthropathy (HOA) is presented in a 14-month-old girl with adult respiratory distress syndrome (ARDS) secondary to severe burn injury. The child developed clubbing during a lengthy and tumultuous hospitalization for her underlying lung disease, and hypertrophic osteoarthropathy was demonstrated radiologically.

Hypertrophic osteoarthropathy in children is a well recognized feature of suppurative lung, congenital heart, and biliary disease but has only sporadically been described with other conditions [1]. We report a case of HOA associated with ARDS, an association not previously described.

Case report

A 14-month-old black female was admitted with 31 % third degree burns. On the second post-admission day she was noted to be in respiratory distress which worsened despite treatment. Clinical and radiological features of ARDS were noted two days later, with a respiratory rate of 75/min and an arterial oxygen saturation of 69–75%. The next twelve weeks were characterized by a turbulent clinical course during which the patient remained confined to a non-oscillatory Bunnel jet ventilator and then to a conventional one.

She sustained two episodes of cardiorespiratory arrest, multiple pneumothoraces resulting in a total of 36 chest tube placements, multiple episodes of pneumomediastinum and several episodes of pneumopericardium requiring drainage on two occasions. Each time one of these events occurred, her oxygen saturation decreased into the 60%–75% range. The patient finally stabilized after a five month intensive care unit stay and was weaned to a tracheostomy collar with oxygen supplementation.

The patient was noted to have marked clubbing and periosteal elevation of the clavicles and limb bones (Figs. 1, 2) in the sixth post-admission week. This peaked at 18–19 weeks post-admission and gradually partially resolved as her clinical condition improved.

Discussion

HOA (Marie-Bamberger's) disease is well known in adults but is rarely encountered in children. Most cases in children are connected with chronic suppurative lung processes, (with and without cystic fibrosis), congenital cyanotic heart disease, biliary atresia/cirrhosis and chronic inflammatory bowel disease [1]. Its exact etiology remains unclear but it is thought that it might result from mediators in the systemic circulation that escape inactivation in the pulmonary capillary bed as a result of bypass of the lung, often associated with conditions causing marked hypoxemia. Since it is associated with disease of visceral organs supplied by the vagus nerve, an associated vagal etiology has also been implicated [2].

The radiographic findings in our case were quite typical and characterized by bilateral periosteal elevation of the clavicles and long bones. These findings, and the associated new onset clubbing, have not previously been described during the course of ARDS. Of particular interest to us is a previous report of clinical

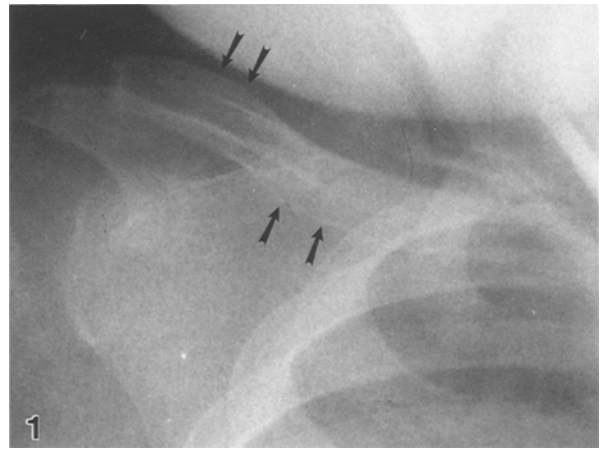

Fig. 1. Right clavicle at 18 weeks postadmission. Note periosteal cloaking of entire bone *(arrows)*. Identical changes were present in the left clavicle

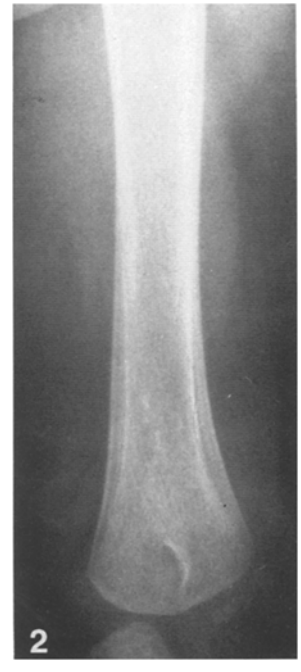

Fig. 2. Left humerus at 19 weeks postadmission. Well formed periosteal new bone is seen. Similar changes were seen in the forearm, and in the long bones of all the other extremities

Correspondence to: Dr. D. Balsam

clubbing in a neonate with severe bronchopulmonary dysplasia [3] but it made no mention of radiological bone changes, if any.

We postulate that severe destruction of our patient's pulmonary capillary bed, as evidenced by her clinical, laboratory and radiological course, led to marked, prolonged hypoxemia and prevented inactivation of an unknown chemical mediator resulting in periosteal new bone formation. While similar changes have been described in children receiving prostaglandins for ductus-dependent heart disease and in burn patients on long term high dose Vitamin A therapy, our case did not receive the former and only a single small dose of Vitamin A was administered early in the course of her disease. Other reported causes of periosteal reaction in children include spontaneous hemorrhage, management on oscillatory ventilation [4], and rickets or hypocalcemia with subsequent healing. None of these factors applied to our patient, and the presence of marked clubbing acquired during the current illness supports the diagnosis of HOA. Therefore, we believe her hypertrophic osteoarthropathy is unrelated to the above and caused solely be the destruction of lung parenchyma by ARDS.

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