Hypertrophic osteoarthropathy in a child due to thoracic Hodgkin's disease

Eleni E. Drakonaki · K. M. Bradley · Z. A. Khan ·
C. Patel · D. J. Wilson

Received: 21 July 2011 / Revised: 28 September 2011 / Accepted: 19 October 2011 / Published online: 18 November 2011
© ISS 2011

Keywords Hodgkins disease · Osteoarthropathy ·
Radiographs · CT

There is periosteal reaction along the shaft of the long bones of the hand and foot, leading to the diagnosis of hypertrophic osteoarthropathy (HPOA), associated with intrathoracic Hodgkin's disease.

Discussion

Hypertrophic osteoarthropathy in children is rare and is typically considered to be a sign of pulmonary disease [1–3]. However, it may be related to a variety of inflammatory or neoplastic disorders, including congenital heart diseases, pulmonary or cystic fibrosis, infectious airway disease, chronic liver and inflammatory bowel disease as well as various intrathoracic and gastrointestinal tumours [1–3]. While up to 90% of HPOA cases are associated with primary or metastatic pulmonary neoplasms in adults,

intrathoracic malignant disease accounts for only 12% of HPOA cases in children [1–3]. The presence of HPOA has been regarded as a poor prognostic sign, since most of the patients with malignancy and HPOA have showed extensive metastases [2]. Therefore, although HPOA is rare in childhood cancers, its occurrence should alert physicians for the recurrence of disease or intrathoracic involvement during follow-up [2–4].

HPOA has a characteristic radiological appearance [1–4]. Initially, there is periosteal new bone formation forming a thin radiopaque line around the cortex and separated from it by a radiolucent band. In our patient, this pattern is evident at both the X-rays, and the CT. Subsequently, the two layers gradually fuse and lead to poorly demarcated or lamellar periosteal reaction. This appearance may be easily differentiated from bone involvement from the Hodgkin's disease itself, as periosteal infiltration by lymphoma usually affects the anterior surfaces of the vertebral bodies rather than the long bones and is always associated with cortical bone destruction [1–4].

In pediatric Hodgkin's lymphoma, ^{18}F -fluorodeoxyglucose positron emission tomography–computed tomography (FDG PET-CT) is increasingly used for staging the disease and assessing prognosis and response to therapy. Given the prognostic significance of HPOA, depiction of this condition in FDG PET-CT scans of children with Hodgkin's disease is important. FDG PET-CT usually reveals mild or moderately increased FDG uptake in the periosteal of the affected bones, as a result of increased metabolic activity [5]. In our patient, however, although the mediastinal tumour was markedly FDG avid, there was no increase in the FDG uptake at the sites of HPOA, indicating low grade metabolic activity (Figs. 1 and 2). In such cases, HPOA can be easily missed if the CT component of the study is not

The case presentation can be found at doi:10.1007/s00256-011-1316-y.

E. E. Drakonaki · D. J. Wilson
Department of Radiology, Nuffield Orthopaedic Centre, John
Radcliff Hospitals NHS Trust,
Headington,
OX3 7LD Oxford, UK

E. E. Drakonaki (✉) · K. M. Bradley · Z. A. Khan · C. Patel
PET-CT Department, Churchill Cancer Centre,
John Radcliff Hospitals NHS Trust,
Headington,
OX3 7LD Oxford, UK
e-mail: drakonaki@yahoo.gr

Fig. 1 **a** Fused coronal half body FDG PET-CT image showing a markedly FDG avid mediastinal mass, along with subcarinal nodes and direct extension of the disease into the lung parenchyma (stage IVe). **b** Follow-up fused coronal half body FDG PET-CT image acquired after two courses of chemotherapy showing reduction in size and no FDG uptake, in keeping with complete metabolic response

Fig. 2 Fused sagittal FDG PET-CT image acquired with the hands raised including part of the humeri in the study. There is periosteal new bone formation at the humeral shaft, which is non-FDG avid (arrows)

carefully reviewed. In our case, the periosteal reaction was still evident after two cycles of chemotherapy, although the tumour had regressed, showing a delayed response of HPOA to therapy compared to the tumor itself.

In conclusion, HPOA, although extremely rare, may be an indicator of pediatric intrathoracic Hodgkin's disease. Therefore, the depiction of HPOA in a skeletal X-ray should lead to further chest imaging to rule out malignancy. As HPOA can be an incidental finding in staging FDG PET-CT scans for pediatric Hodgkin's disease and given its prognostic significance, radiologists and nuclear medicine physicians should review the CT component of the study very carefully, as HPOA may be FDG negative and could therefore be easily overlooked.

References

1. Yao Q, Altman RD, Brahn E. Periostitis and hypertrophic pulmonary osteoarthropathy: report of 2 cases and review of the literature. *Semin Arthritis Rheum.* 2009;38:458–66.
2. Karadeniz C, Oguz A, Dalgic N. Hypertrophic osteoarthropathy and childhood Hodgkin disease. *Pediatr Hematol Oncol.* 2001;18:79–82.
3. Shapiro RF, Zvaifler NJ. Concurrent intrathoracic Hodgkin's disease and hypertrophic osteoarthropathy. *Chest.* 1973;63:912–6.
4. Cavanaugh JJA, Holinan GH. Hypertrophic osteoarthropathy in childhood. *J Pediatr.* 1965;66:27.
5. Strobel K, Schaefer NG, Husarik DB, et al. Pulmonary hypertrophic osteoarthropathy in a patient with non small cell lung cancer: diagnosis with FDG PET/CT. *Clin Nucl Med.* 2006;31:624–6.