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Primary hyperparathyroidism as a cause of multiple long bone fractures in a young Nigerian adult

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Summary

Primary hyperparathyroidism is a recognized though relatively rare cause of multiple limbs fractures. We report a case in a young Nigerian adult. It is emphasized that primary hyperparathyroidism may be asymptomatic and therefore under-recognized in this environment because it is very uncommon.

Keywords: Primary hyperparathyroidism, spontaneous femoral fractures.

Résumé

L'hyperparathyroïdisme primaire est reconnu, quoique rare être la cause des fractures multiples des membres. Nous reportons ici le cas d'un jeune Nigérian Adult. Il a été insisté sur le fait que l'hyperthyroïdisme primaire pourrait être asymptomatique et par conséquent pas très bien connu dans notre environnement, parce qu'elle n'est pas très commune.

Introduction

Primary hyperparathyroidism is said to be an uncommon condition in Sub-Saharan Africa. This may either be due to misdiagnosis or non-recognition of the disease as few affected individuals present with overt or florid clinical features. The introduction of the multi-channel autoanalyser in measuring serum calcium concentrations has led to a dramatic change in the clinical recognition and diagnosis of hyperparathyroidism [1,2,3,4].

We report a case of a young man who had multiple femoral fractures with no preceding history of trauma or other systemic manifestations of hypercalcaemia.

Case Report

A 36-year-old man presented with few weeks' history of right thigh pain with no history of trauma or fever. There was no obvious swelling in the thigh or associated deformity. Radiograph of the right femur revealed an osteolytic lesion in the right femur (Fig. 1). He developed a spontaneous fracture of the right femur while in bed and was placed on skeletal traction pending further investigations. Few days after, he developed pain and swelling in the left thigh and radiograph of the left thigh revealed osteolytic lesion in the left femur with an insufficiency fracture (Fig. 2).

Biochemical blood analysis revealed parathyroid hormone of 779 pg/ml (normal = 13-54 pg/ml) and elevated calcium level of 13.5 mg/dl (normal = 8.5-10.2 mg/dl) with

serum phosphate level of 0.9 mg/dl (normal = 2.5-4 mg/dl). Skeletal survey showed multiple lytic lesions in both humerus, skull, pelvis and subperiosteal erosion in the phalanges. The diagnosis of primary hyperparathyroidism with severe bony manifestation was made.

Further evaluation of the patient with ultrasonography demonstrated parathyroid adenoma and he is presently being worked up for parathyroidectomy.



Fig. 1:



Fig. 2

Discussion

Primary hyperparathyroidism is an endocrine disorder characterised by excessive secretion of parathyroid hormone, which causes a set increase in the serum calcium level. This disease condition is commoner than is generally thought because patients are often asymptomatic and diagnosis is suspected on the incidental finding of hypercalcaemia [3,4]. Hypercalcaemia has been detected in an increased number of persons in recent years mainly on account of the increased awareness and screening facilities available for serum calcium estimation [1,2,3,4,5].

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Various studies since 1969 indicate a prevalence of primary hyperparathyroidism of 100-200 cases per 100,000 in various clinic and hospital populations [1,2]. It is unusual in children and most common in the fifth and sixth decades [1,2]. This disorder is two to three times more common in women than in men, and this difference increases with age to nine times in female compared to male patients over 70 years [1,2,3,4]. This is worrisome because of the strong possibility that primary hyperparathyroidism may cause or aggravate osteoporosis [2].

The precise aetiology of primary hyperparathyroidism is unknown but its genetic basis is demonstrated by its occurrence in familial syndromes (e.g., MEN1, 2a) as well as the increase of parathyroid adenomas many years after exposure of the head and neck to radiation [1,2,6]. A single parathyroid adenoma or parathyroid cysts are uncommon and parathyroid carcinoma is very rare [4,7,8,9].

The effect of primary hyperparathyroidism on bone is well noted and the disease was first identified in association with a skeletal disorder called osteitis fibrosa generalisata [9]. This is characterised radiologically by subperiosteal resorption of cortical bone, grossly abnormal bone architecture, bone cyst and "brown tumours" with deformity and multiple fractures in severe cases [9,10,11,12]. The incidence of bone lesions in this disease is reported to range from 10 to 15% to as high as 30-40% [12]. There is a tendency for asymptomatic patients to be older than those with symptoms as the natural history of mild primary hyperparathyroidism can be benign.

Albright and Reifstein [12] emphasized that urologic abnormalities particularly nephrolithiasis are more common than skeletal abnormalities and patients with bone disease have a much more rapid course, with early renal failure and nephrocalcinosis but this was not the case in our patient. The occurrence of nephrolithiasis in primary hyperparathyroidism has decreased greatly because more symptom-free patients may first present with tiredness and muscle weakness that primarily affect the lower extremities and is associated with neuropathic atrophy. Other patients have symptoms that constitute a poorly characterised neuropsychiatric syndrome from decreased attentiveness and ability to concentrate to severe depression [1,9].

Primary hyperparathyroidism and malignancy account for more than 90% of cases of hypercalcaemia. Hypercalcaemia is the biochemical hallmark of primary hyperparathyroidism and is an essential diagnostic criterion [1,9].

The serum phosphate concentration is usually in the low or low/normal range because of depressed renal-tubular re-absorption. The circulating parathyroid hormone is high in primary hyperparathyroidism as can be seen in this patient. Radiologic and imaging techniques like skeletal survey, abdominal and anterior neck ultrasonography, CT scan and/or MRI will help in demonstrating skeletal and renal lesions and localisation of the parathyroid gland pathology.

Most patients with primary hyperparathyroidism benefit from surgery. Indications for surgery as

recommended by the National Institute of Health Consensus Development Conference [1,13], include: [1] total serum calcium concentration of more than 11.0 mg/dl (0.25 mmol/L) above the upper limit of normal range [2] if there is evidence of overt bone disease (e.g. osteitis fibrosa cystica); [3] if cortical bone mineral density is more than 2SD below the adjusted mean for age and sex; (4) if there is reduced renal function; (5) if there is renal stone diseases (nephrolithiasis or nephrocalcinosis); [6] significant hypercalcaemia (400 mg (9.98 mmol) per day); [7] if there has been an episode of acute primary hyperparathyroidism; and [8] patients who are young (50 years) should be considered for surgery even in the absence of other indications since younger patients will have primary hyperparathyroidism for longer than older people and therefore have a greater theoretical risk of complications.

Surgical complications are rare and include hypoparathyroidism and injury to the recurrent laryngeal nerves. Most patients experience mild; and transient postoperative hypocalcaemia, but those with extensive and florid skeletal disease can develop protracted hypocalcaemia that is due to remineralisation of "hungry bones" that will in most cases require aggressive replacement therapy with calcium, magnesium and active vitamin D [1,7,8,9,14].

Patients who are symptomless and do not meet criteria for surgery can be treated conservatively. Regular reassessment and follow-up is essential to detect complications that would justify prompt surgery. Medical management is also appropriate for patients who have symptoms but are unfit for surgery or have inoperable parathyroid cancer.

Urgent medical treatment of hypercalcaemia is often necessary to stabilise patients before surgery. Agents in current use include oral phosphate supplement, which lowers calcium concentration in the serum and urinary calcium excretion. Oestrogen therapy especially in postmenopausal women with primary hyperparathyroidism is known to inhibit parathyroid hormone-mediated bone resorption. Bisphosphonates and calcitonin are effective in the acute management of hypercalcaemia.

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