

Parathyroid adenoma presenting with multiple Brown tumors in an adolescent patient

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ABSTRACT

Parathyroid glands are endocrine glands that secrete parathyroid hormone (PTH) and regulate calcium and phosphorus metabolism. The overexpression of PTH is called hyperparathyroidism (HPT) and is classified as primary, secondary and tertiary. Primary HPT (PHPT) is the most common cause of parathyroid adenomas (80–85%). Juvenile PHPT developed during adolescence is quite rare. The incidence of bone lesions in all age groups is reported to be 10–20% in PHPT patients and 5% in juvenile PHPT patients. It is known that in patients with bone lesions, regression occurs in lesions after parathyroidectomy. In this case report, we aimed to present the imaging findings of long bones and left 5th metacarpal bone multiple Brown tumors which is a rare presentation of parathyroid adenoma in adolescent patient and regression after parathyroidectomy.

Keywords: Adolescent; Brown tumor; primary hyperparathyroidism.

Cite this article as: Aslan S., Ceyhan Bilgici M., Bernay R. F., Aydin H. M., Selcuk M. B. Parathyroid adenoma presenting with multiple Brown tumors in an adolescent patient. *North Clin Istanbul*

Parathyroid glands are two endocrine glands, usually located in vicinity of the upper and lower poles of the thyroid gland. Parathyroid glands secrete parathormone (PTH) and regulate calcium - phosphorus metabolism. PTH plays an important role in bone mineral balance. The overexpression of PTH is termed as hyperparathyroidism (HPT) and is classified as primary, secondary and tertiary hyperparathyroidism.

Primary HPT (PHPT) develops as a result of due to adenoma (80–85%), hyperplasia (10–15%) and parathyroid carcinoma (<0.5%) [1]. Secondary HPT is also due to overexpression of PTH secondary to hypocalcemia or vitamin D deficiency. Tertiary HPT develops as a result of autonomy acquired by the parathyroid glands as a result of long-lasting secondary HPT [2]. Juvenile PHPT in the adolescent period is very rarely seen, and a limited number of cases have been reported in the literature [3].

The incidence of bone lesions in PHPT was reported to be 10–20%, and in juvenile PHPT as 5% [4]. The most commonly identified bone lesions are bone resorption, bone cysts, Brown tumors, and generalized osteopenia. Bone lesions are often seen in facial bones, ribs and pelvic bones, and they are rare in long bones. Brown tumors are reactive lesions that result in disruption of the balance between bone formation and destruction in favor of bone resorption due to the direct effect of PHT. Diagnosis is difficult and radiologically confused with giant cell tumor, bone metastasis or multiple myeloma [5].

In this case report, we aimed to present the imaging findings of a large number of Brown tumors in the long bones and the fifth metacarp of the left hand, which have a rare manifestation of parathyroid adenoma in the adolescent patient that regresses after parathyroidectomy.



Received: October 08, 2017 *Accepted:* January 02, 2017 *Online:* August 09, 2018

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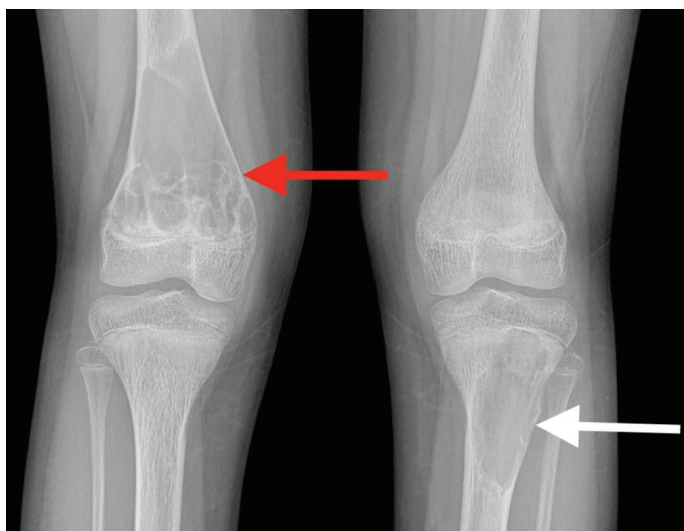


FIGURE 1. A bone lesion measuring 71x30 mm (white arrow) on the proximal metaphysis of the left tibia and the other one measuring 87x44 mm in size (red arrow) in the distal metaphysis of the right femur are observed.



FIGURE 2. A bone lesion measuring 66x30 mm (white arrow) in the proximal metaphysis of the left tibia and the other one measuring 80x36 mm in the distal metaphysis of the right femur (red arrow) are observed.

CASE REPORT

A 14-year-old girl was admitted to the outpatient orthopedic clinic with complaints of increasing pain and

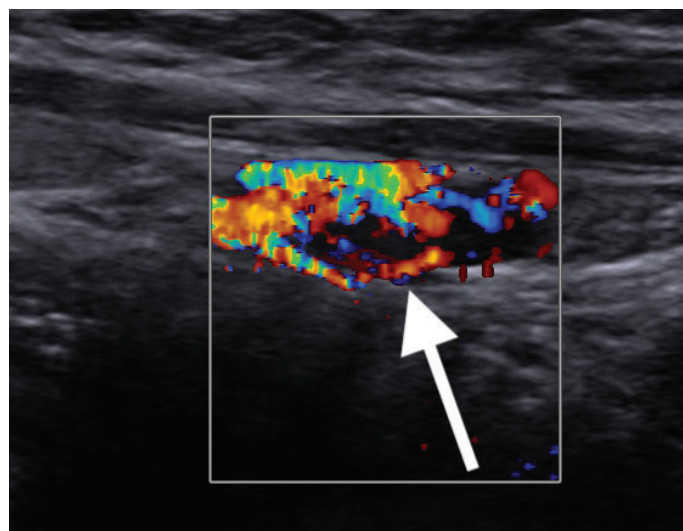


FIGURE 3. Parathyroid adenoma-compatible lesion (white arrow) in the size of 11,5x5,5x19,5 mm is seen in the inferior lobe of the left thyroid gland.

fatigue on her left knee for one month. On physical examination, the patient's weight (40 kg (<3p)) and height 146 cm (<3p) were measured. Her left knee movements minimally limited. Bilateral knee x-ray, and lower extremity long bone radiograms were obtained and the patient was referred to us. Radiograms obtained revealed radiolucent bone lesions proximal to the left tibia and distal to the right femur and extensive decrease in bone densities (Figure 1). In other bones, bone scanning was performed to detect the presence of bone lesion. A bone lesion was observed in distal part of the fifth metacarp of the left hand with similar appearance. Low-dose computed tomography (CT) of the both lower extremities to aid in differential diagnosis. Likewise CT demonstrated expansile, hypodense bone lesions at a location proximal to the left tibia and distal to the right femur (Figure 2). Remarkable laboratory findings were as follows: calcium, 13.4 mg / dL (8.6–10 mg / dL); phosphorus, 2.22 mg / dL (2.6–4 mg / dL); alkaline phosphatase 636 IU / L (73–189 IU / L); 25-OH Vitamin D3 1.25 µg / L (10–55 µg / L), and PTH, 1441 pg / mL (10–65 pg / mL)

With available findings, lesions were thought to be Brown tumors due to PHPT. Neck ultrasonography (USG) and parathyroid scintigraphy (Tc 99m sestamibi) were performed to investigate the etiology. In the neck US, a lesion compatible with parathyroid adenoma was observed in the vicinity of the lower left thyroid gland (Figure 3). Scintigraphy also showed findings consistent with parathyroid adenoma. The patient was diagnosed

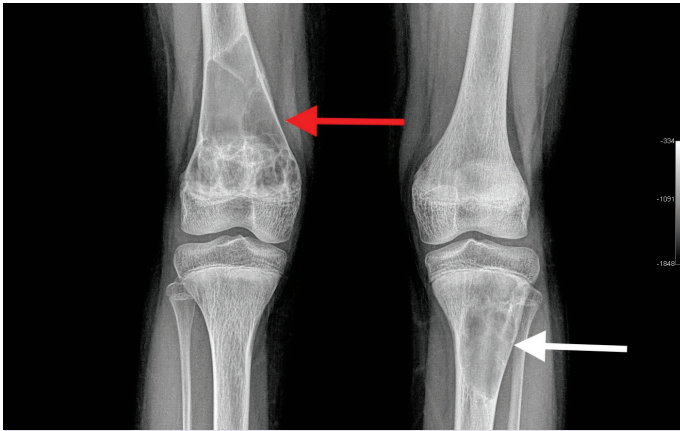


FIGURE 4. Decreases in the dimensions of the Brown tumors localized in the tibia and femur are observed on the control knee radiograms.

with parathyroid adenoma and Brown tumor developed secondary to parathyroid adenoma.

Parathyroid adenoma was removed by the department of surgery. Calcium and PTH levels were normal after parathyroidectomy. The patient had no complaints at 1 month follow-up and calcium, PTH and phosphorus levels were normal. The size of the lesions seen in the knee radiogram also decreased (Figure 4). Thus, without the need for biopsy it was proved that the lesions were Brown tumor and developed due to parathyroid adenoma.

DISCUSSION

We presented an adolescent patient who presented with left knee pain, and had multiple Brown tumors in the long bones of the lower extremities. Radiographically detected bone lesions and laboratory findings supporting PHPT primarily suggested us the presence of Brown tumor. Normalization of PTH after parathyroidectomy and decrease in the size of the lesions in the control imaging performed one month later confirmed our diagnosis without the necessity of biopsy or surgical intervention.

The Brown tumor is a slowly growing, occasionally painful, and locally aggressive bone lesion without metastatic potential. The incidence of Brown tumor in PHPT is reported to range between 1.5–1.7% [6]. It is frequently seen in adults over the age of 50, but the appearance of Brown tumors secondary to parathyroid adenoma in the adolescence period is very rare [7]. The imaging findings of the Brown tumor are not specific.

They are frequently seen as bone lesions with lytic or sclerotic contours, that do not cause cortical erosion, inflammatory changes or periosteal reaction [8]. They are rarely localized in long bones, and they are seldom seen in multiple numbers. It may be confused with giant cell tumor, aneurysmal bone cyst, giant cell granuloma, cherubism when it develops in the mandible, and it may be confused also with metastasis when it is seen in multiple numbers [6].

In the literature, most of the cases of the Brown tumor have been presented in a single bone and mostly in the mandibula and palate [9]. It is difficult to make the diagnosis of Brown tumor in biopsy materials histopathologically, and diagnosis can be frequent indicated if clinical manifestations of HPT are found.

In cases of suspect Brown tumor, the presence of parathyroid adenoma should be investigated before resorting to biopsy and surgical intervention.

If parathyroid adenoma is detected, parathyroidectomy is the most appropriate option. With parathyroidectomy, PTH levels are kept under control, and also existing Brown tumors are treated [4, 10]. Tumors that grow despite treatment or do not change in size during the 6-month period after treatment, and lesions that impair the functions of the affected bone, or those are associated with fractures require interventional treatment [10].

In conclusion, Brown tumor is a rare bone lesion that develops secondary to PHPT in adolescents and it can be anticipated in cases with prolonged PHT elevation. Imaging methods are important diagnostic tools. When suspect lesions are revealed by imaging modalities, the patients should not be immediately directed to biopsy, differential diagnosis should be considered, biochemical values should be reviewed, and the presence of parathyroid adenoma should be investigated.

Informed Consent: Written informed consent was obtained from the patient who participated in this study.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study has received no financial support.

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