Radiological findings in the primary hyperparathyroid case with multiple brown tumors: a case report

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Abstract. Primary hyperparathyroidism is a disease characterized with the excess secretion of parathormone. During the course of this disease, bone loss occurs, particularly depending on resorption in the skeletal system. One of the complications of primary hyperparathyroidism is fibrotic, cystic bone pathology which is called brown tumor. We have planned to present a 15-year-old Turkish female patient with primary hyperparathyroidism and multiple brown tumors depending on parathyroid adenoma and to discuss the radiological changes in the presentation article.

Key words: Brown tumor, Primary hyperparathyroidism, Parathyroid adenoma

1. Introduction

Hyperparathyroidism is a table characterized with increase of parathormone secretion. As a result of parathormone hypersecretion, excess calcium reabsorbtion from kidneys, phosphaturia, increased vitamin D synthesis and bone resorbtion occur. Parathormone causes osteoclastic activity in the bones. While renal calculi has been reported in 10-25% of primary parathyroid cases, frequency of bone disease has been reported as 10-20%. Brown tumor is a localized bone cyst and is a histologically benign lesion. Although bone findings in primary hyperparathyroidism are rarely seen, they are seen frequently in the carcinomas or in serious secondary hyperparathyroidism. Brown tumor may cause swelling, pathological fracture, and bone pain in the skeletal system. Multiple brown tumors depending on primary hyperparathyroidism are very rare (1). Only six cases had been reported in the English literature (2-7).

In this article, we presented the radiological and clinical properties of a case with primary hyperparathyroidism caused by parathyroid adenoma.

2. Case presentation

A 15-year-old female patient applied orthopedy clinic with painful swelling in the 2^{nd} and 5^{th} fingers of left hand, left forearm and right limb for a duration of one year.

On physical examination of the patient, painful tumoral lesions were determined on distal left forearm, proximal phalanges of 2^{nd} and 5^{th} fingers, and distal right tibia. On the left inferior thyroid lobe, a 1x1 cm sized palpable nodule with soft consistency was noted. Findings from other system examination were normal.

In the laboratory analysis, serum level of calcium was 12.4 mg/dl (normal 8.4-10.7 mg/dl), serum albumin level was 5 g/dl (normal 3.4-4.8 g/dl), serum alkaline phosphatase level was 3182 IU/L (normal 50-240 IU/L), serum acide phosphatase level was 11.36 IU/L (normal 0-5.5 IU/L), and serum parathyroid hormone level was 656 pg/ml (normal 7-53 pg/ml).

Large cystic lesions causing fractures on the cortex of proximal and distal ulna were determined on the x-ray of left hand and forearm.

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Large cystic lesions were observed in the 3rd metacarpal bone of the left hand, and proximal



Fig. 1. Appearance of multiple brown tumors in left forearm x-ray of the patient.



Fig. 2. Right cruris x-ray shows brown tumor in lateral malleolus.

phalanges of the 2nd and 5th fingers. These appearances were found compatible with brown tumor (Fig. 1). Thinning and fractures of the

calcaneal and distal fibula cortex were observed on the x-rays of right foot and cruris (Fig. 2). On lateral cranium graphy, granular deossifications were seen more distinct at the 1/3 peripheric area (Fig. 3). On the left wrist computed tomography (CT) examination, a mass at the distal of left radius causing bone deformation was observed (Fig. 4).



Fig. 3. Lateral cranium x-ray shows granular deossification



Fig. 4. Left wrist CT image shows expansion of tumor in left distal radius.

Increased activity compatible with brown tumor was observed in the bone scintigraphy (Fig. 5). Ultrasonography (USG) and magnetic resonance imaging (MRI) examinations revealed a 1cm sized mass in the left parathyroid gland. It was thought to be compatible with parathyroid adenoma (Fig. 6,7). Performed parathyroid scintigraphy did not contain any finding in terms of parathyroid adenoma.

Parathyroidectomy was applied to the patient and histopathological evaluation confirmed the diagnosis as parathyroid adenoma.



Fig. 5. Bone scintigraphy.



Fig. 6. Left parathyroid ultrasonography shows parathyroid adenoma.



Fig. 7. Imageng of neck MR shows parathyroid adenoma in left parathyroid gland.

3. Discussion

Primary hyperparathyroidism is a table characterized by hyperfunction of parathyroid glands. Female/male ratio is 5/2. In our literature study, multiple brown tumor cases depending on primary hyperparathyroidism was firstly reported by Joyce et al. in 1994 (2,4). Also five more cases have been observed in the literature since then.

Although Brown tumor is generally seen more frequently in the seriously secondary hyperparathyroidism, it is fairly characteristic for primary hyperparathyroidism. Brown tumors may be observed on facial bones, pelvis, costa, manus bones and femur. In the hyperparathyroidism early findings are seen on hands. They may be multiple in terminal the stage of hyperparathyroidism or in the carcinomas. They may cause swelling, pathological fracture and bone pain (1).

Generally, high resolution USG, radionuclide imaging, CT and MRI combinations are used for imaging of parathyroid pathology (8).

High resolution USG is one of the most common imaging methods used for neck evaluation and it is practically the first option in the primary hyperparathyroidism assessment. On USG, parathyroid adenoma is seen typically as round or oval homogenous, hypoecoic nodule localized behind the thyroid gland and at the lower aspect of paratracheal or paraeosophageal region. It is clearly separated from thyroid gland due to its capsule. Morphological differences such as hyperecoic component, cystic changes and calcification may be seen particularly in large adenomas. More than 90% of parathyroid adenomas include intraparanchimal hypervascular pattern in the color flow imaging (8).

In the radionuclide parathyroid imaging; Technetium (Tc) 99m marked 2-methoxyisobutylisonitrile (sestamibi) scintigraphy and 201-99m pertechnetat thallium and Tc are predominant to subtraction imaging due to their short half time, giving more image quality and low radiation risk. Sensitivity more of scintigraphic findings is low in lesions that are smaller than 1 cm (8).

Thin section contrast CT is usable for localization of parathyroid adenoma. Its sensitivity range changes between 46-87%. One of the advantages of CT on USG is its ability to determine particularly ectopic parathyroid adenomas in the mediastinum (9).

Sensitivity of MR in the determination of parathyroid adenoma varies between 65-80%. Most common appearance of hyperfunctional parathyroid gland in T1 weighted images is isodens and increased intensity after intravenous gadolinium injection (10).

Brown tumor may cause various complications. Pathological fracture risk is higher in the brown tumors with hemorrhagic and/or cystic component in weight-bearing bones. MRI is important for determination of hemorrhage, cystic component and indirectly estimation of fracture risk in brown tumor (11).

Multiple Brown tumors and other multiple bone primary depending changes on hyperparathyroidism also may appear in the parathyroid adenomas like in our case. Modern imaging methods play an important role in the diagnosis of primary adenoma. Radiological findings in the skeletal system cannot differentiate parathyroid adenoma and parathyroid cancer. Determination of preoperative localization is important for security, surgical efficiency and particularly for invasive surgery. Even if USG is used for beginning evaluation, radionuclide imaging combination performed by usage of USG and Tc 99m-sestamibi is useful for determination of preoperative localization of parathyroid gland with hyperfunction. When doubtful results found in the USG and Tc 99msestamibi combination, CT and MRI may be practical. Radiological images are insufficient to differentiate hyperplasia, adenoma and

carcinoma. Histopathology is necessary for definitive diagnosis.

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