Introduction

Primary hyperparathyroidism may be caused by giant parathyroid adenoma, a type of large, solitary adenoma. The median weight of adenomas is 0.61 g, and an adenoma in excess of 3.5 g is rare. Primary hyperparathyroidism may also cause osteolytic lesions called brown tumors. Currently, primary hyperparathyroidism is detected via a screening blood test. Therefore, in developed countries, such as North America, Europe, and Japan, it is unusual that a brown tumor would be the first presentation of primary hyperparathyroidism.

Here, we report a rare case of giant parathyroid adenoma presenting with a brown tumor. Because of this rarity, the differentiation between parathyroid adenoma and carcinoma is essential.

Case presentation

A 21-year-old woman presented to the orthopedic clinic with persistent pain in her right knee. She had reported enduring pain in her right knee as well as general fatigue over the last few years. Radiographic examination of her right knee revealed an osteolytic lesion in her right tibia (Fig. 1). A bone biopsy was performed. A histological examination revealed that this lesion consisted of circular-shaped or spindle-shaped mononuclear cells and osteoclast-like giant cells. Cellular density was high with attendant hemorrhaging (Fig. 2). These histological findings were compatible with a giant cell tumor. Her blood laboratory tests revealed that serum calcium and intact parathyroid hormone were increased significantly. Physical examination showed a 3-cm, firm, immovable lump in her neck. Ultrasonography of the patient’s neck revealed a 3.5-cm, well-circumscribed mass with a homogeneous internal texture. Computed tomography also showed a localized tumor with clear margins. The 99mTc-MIBI scintigraphy showed a hot spot in the right inferior gland. Based on these findings we judged that she had giant parathyroid adenoma presenting with a brown tumor. These pre-operative findings suggest that the possibility of carcinoma was low. We performed a parathyroidectomy via a small incision. The excised parathyroid gland weighed 10.3 g. The patient’s postoperative course was unremarkable.

Conclusions: Giant parathyroid adenoma presenting with a brown tumor is rare. Accordingly, the differentiation between giant adenoma and parathyroid carcinoma is crucial. However, we determined that the possibility of carcinoma was low based on imaging studies, including ultrasonography and computed tomography. As a result, we performed successful parathyroidectomy with a small incision.

Key words: brown tumor, giant parathyroid adenoma, primary hyperparathyroidism, case report

was 0.96. Computed tomography showed that a tumor was located near the left lobe of the thyroid (Fig. 4). The tumor was separate from the surrounding structure in both computed tomography and ultrasonography. 99mTc-MIBI parathyroid scintigraphy showed a hot spot in the left inferior gland (Fig. 5).

Based on these findings, we judged that the patient had a brown tumor. The causal lesion was the left inferior parathyroid gland. Although physical examination showed a large, firm, immovable tumor, ultrasonography and computed tomography revealed that this tumor showed few signs of malignancy. Therefore, we diagnosed this lesion as adenoma, clinically. We judged that removal of the tumor with a small incision was possible. However, a larger incision could be made to ensure definitive surgical treatment if any signs of infiltration of the tumor were found intra-operatively. We informed the patient of our treatment strategy, and we obtained informed consent.

We then performed parathyroidectomy via a 3-cm incision. There were no signs of infiltration around the tumor. The surgical specimen was 4.0 × 2.5 × 1.3 cm and weighed 10.3 g (Fig. 6). Microscopic examination showed that the tumor consisted of proliferating tumor cells, which exhibited a solid or follicular arrangement (Fig. 7). Most of the cells were small or medium in size, and the nuclei contained coarse aggregates of chromatin. These tumor cells showed no atypical features suggesting malignant potential, including invasiveness or vessel invasions. The tumor capsule was intact. Although the patient was administered the maximum dose of 6 g calcium lactate hydrate and 2 μg alfacalcidol per day, her serum calcium dropped to 8.1 mg/dL on day 5 after operation. However, she remained asymptomatic.
A giant parathyroid adenoma presenting with a brown tumor

Discussion

Here, we report a rare case of giant parathyroid adenoma presenting with a brown tumor. Because of this rarity, the differentiation between parathyroid adenoma and carcinoma is essential. To achieve this, careful analysis of ultrasonography and computed tomography images was performed, enabling us to perform successful parathyroidectomy with a small incision.

In our case, the excised parathyroid adenoma weighed 10.3 g. Spanheimer et al. investigated a series of 300 consecutive parathyroid adenomas. They revealed that the 95% percentile of weight for these cases was 3.5 g. Therefore, the adenoma from our patient was considered giant parathyroid adenoma.

In cases of giant parathyroid adenoma, differentiation from parathyroid carcinoma is necessary. Parathyroid carcinoma is often accompanied by significantly elevated serum calcium and iPTH, osseous symptoms, and a palpable mass. Our case also exhibited these clinical findings; however, the findings of image studies in our case were incompatible with carcinoma. Our tumor was an oval-shaped mass with a well-circumscribed margin and separation from surrounding tissue. The depth/width ratio was 0.96, which represents adenoma, rather than carcinoma. Although the giant parathyroid adenoma measured 4.0 cm in our case, we were able to successfully

Fig. 5 Images of 99mTe-MIBI parathyroid scintigraphy. Arrows indicate increased radiotracer accumulation in the left lower parathyroid gland.

Fig. 6 Surgical specimen of the left inferior giant parathyroid adenoma. The maximum size was 4.0 cm, and the weight was 10.3 g. The scale denotes 1.0 cm.

Fig. 7 Microscopic findings of the removed parathyroid tumor. Proliferative tumor cells exhibited a solid or follicular arrangement. Most of these cells were small or medium in size, and the nuclei contained coarse aggregates of chromatin. The tumor capsule was intact.
use a minimal excision technique

Parathyroid adenoma and carcinoma can lead to hyperparathyroidism, which may cause osseous lesions, including brown tumors. Hypersecretion of iPTH activates osteoblasts, which can result in the acceleration of bone absorption. Chronic hypersecretion in a localized area can precipitate the formation of a lytic bone lesion. A histological examination shows the appearance of giant multi-nuclear cells, the proliferation of fibroblasts, and the accumulation of hemosiderin. The pigments in hemosiderin cause these lesions to appear brown, hence the name ‘brown tumor’.

The prevalence of symptomatic hyperparathyroidism depends on location, and few Japanese individuals show symptomatic manifestation of hyperparathyroidism. Notably, 2–5% of patients with primary hyperparathyroidism develop this type of tumor. However, asymptomatic hyperparathyroidism increased significantly in the United States of America and the United Kingdom after the institution of multichannel biochemical screening test.

Classical symptomatic hyperparathyroidism is still frequent in some developing countries, and brown tumors are quite rare in patients with hyperparathyroidism in Japan. One Japanese university hospital reported the first manifestation in 100 cases of hyperparathyroidism. In total, 84 cases were asymptomatic, and only 4 cases showed osseous-type tumors; no brown tumors were detected. Currently, brown tumors appear to be the initial presentation in younger patients because younger patients are less likely to receive screening blood tests.

**Conclusion**

We report a rare case of giant parathyroid adenoma presenting with a brown tumor. Because of this rarity, the differentiation between parathyroid adenoma and carcinoma is essential. Accordingly, careful analysis of ultrasonography and computed tomography images was performed, which enabled us to perform successful parathyroidectomy with a small incision.

**List of abbreviations**

iPTH: intact parathyroid hormone

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**Consent for publication**

We obtained written informed consent for publication from the patient.

**COI**

The authors declare no conflicts of interest associated with this manuscript.

**References**