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Case Report

Functional cystic parathyroid adenoma with bilateral mandibular brown tumor: a case report ☆

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ABSTRACT

Primary hyperparathyroidism is a disorder of increased parathyroid hormone secretion. Among various causes, parathyroid adenoma is one of the common causes. However, Cystic parathyroid adenoma is a very rare entity. Patients may present with various signs and symptoms related to hypercalcemia, with brown tumors being the end-stage presentation. Although radiological modalities play a central role in diagnosing parathyroid adenomas, histopathology is important to attain a diagnosis. Herein, we present a case of a 53-year-old patient who presented with functional cystic parathyroid adenoma with bilateral mandibular brown tumor.

Introduction

Primary hyperparathyroidism is a disease in which the parathyroid hormone is secreted in high amounts from the parathyroid glands [1]. It can be due to several etiologies with parathyroid adenoma being the most common underlying cause. However, other less common conditions including parathyroid hyperplasia and cancers were identified [1,2]. In addition, it presents with various signs and symptoms of hypercalcemia affecting different organs, including the gastrointestinal tract and kidneys, such as constipation, vomiting, frequent urination, and renal stones [1–3].

Cystic parathyroid adenoma, which can be functional or nonfunctional, is also a very rare pathology of the parathyroid glands [3,4]. Moreover, functional ones compose only around 10% out of 0.01% of neck masses [3]. The clinical features of patients with functional cysts are similar to that experienced by other patients with primary hyperparathyroidism such as gastrointestinal and renal symptoms [3–5]. However, it is less likely to be detected on Tc-99m Sestamibi scintigraphy, which can be explained by parathyroid gland lack of uptake due to compression [2,5]. This can cloak the clinical presentation and change the track away from the diagnosis of this condition [4,5].

This case is unique as it exhibits not only an extremely rare condition, but the first case to report a functional cystic parathyroid adenoma diagnosed alongside with bilateral mandibular brown tumor.

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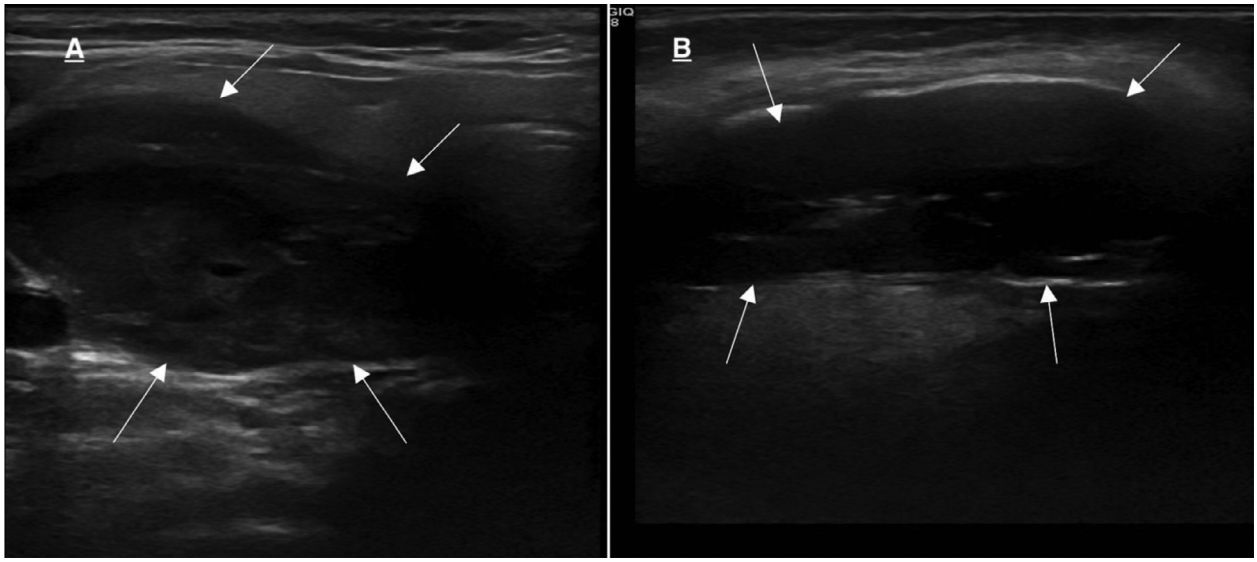

Fig. 1 – An ultrasound image showing: (A) At the level of the right thyroid lobe, a large cystic lesion with thick content causing mass effect on the posterior aspect of the right thyroid lobe and pushes it anteriorly. (B) The right mandibular bone shows a well-defined hypoechoic expansile lesion within the right mandible.

Case presentation

A 53-year-old female patient presented to the clinic complaining of a neck mass. The mass was anterolateral, painless, gradually enlarging, well-circumscribed, doesn't move with swallowing, and had no skin changes. In addition to that, there was a painful mandibular swelling. She was diagnosed with depression and hypertension, and she was confused and had a muscular weakness. She is currently using Candesartan for hypertension and Sertraline for depression. She is a non-smoker and had elevated blood pressure (153/96 mm Hg) and a high body mass index (41 kg/m^2) normal range (18.5-25 kg/m^2). Laboratory tests were ordered which showed elevated serum and urine Calcium levels (11 mg/dL) (8.6-10.3 mg/dL) and an elevated serum Parathyroid hormone (PTH) (986 pg/mL) (10-65 pg/mL). Ultrasound showed a large cystic lesion pushing the right thyroid lobe anteriorly (Fig. 1A). An aspirated sample from the cystic lesion showed a PTH level of (8565 pg/mL). An ultrasound of the mandible swelling showed a well-defined expansile lesion within the right mandible which raised the suspension for a brown tumor according to the clinical scenario (Fig. 1B). A noncontrasted computer tomography (CT) scan of the mandible showed a well-defined expansile lytic lesion on the bilateral sides of the mandible. In addition, a bone window axial CT showed a larger lesion of the right side with evidence of bone destruction at the same side (Fig. 2). An magnetic resonance image showed a moderate enhancement of the mandibular lesion (Fig. 3). Moreover, it showed a well-defined cystic lesion posterior to the right thyroid lobe, thick irregular wall enhancement measuring 5.5 × 3.2 × 8 cm in anteroposterior, transverse, and craniocaudal dimensions, and perilesional edema due to the recent cyst aspiration (Fig. 4). A Bone scintigraphy scan using Tc 99m MDP was ordered to detect other brown tumor locations, which were negative apart from the mandibular lesions (Fig. 5). The patient was referred to an endocrine surgery for surgical excision of the adenoma. The cyst was excised surgically with an eventless postsurgical course. The result of the biopsy showed features consistent with cystic parathyroid adenoma with normal adjacent tissues, without malignant features. Due to the aforementioned findings, she was diagnosed with functional cystic parathyroid adenoma with bilateral mandibular brown tumor. Unfortunately, the patient lost to follow-up.

Discussion

Constituting approximately or even less than 1%-2% of all causes of primary hyperparathyroidism, cystic parathyroid adenomas are considered one of the extremely uncommon pathologies [2,5]. Furthermore, only a handful of studies have tackled the matter of the incidence of cystic parathyroid adenoma which makes up about 0.01-5% of all neck masses. [3,4,6]. Parathyroid cysts are usually discovered around the ages of 30-60 years [5,6]. Moreover, although parathyroid cysts have a higher female-to-male ratio, the functional type of these cysts has a slight male-to-female predominance (1.6:1) [5,6].

Several hypotheses regarding parathyroid cyst's origin are present; one of the theories suggests that they have an embryologic origin from the 3rd and 4th brachial pouches which is supported by the fact that it contains several tissues (adipose, parathyroid, thyroid, etc.) [2,7]. Another theory claims that they arise due to the fusion of former parathyroid microcysts [4,7]. However, one theory states that functional cystic parathyroid adenoma results from the degeneration, infarction, or hemorrhage into parathyroid glands and adenomas

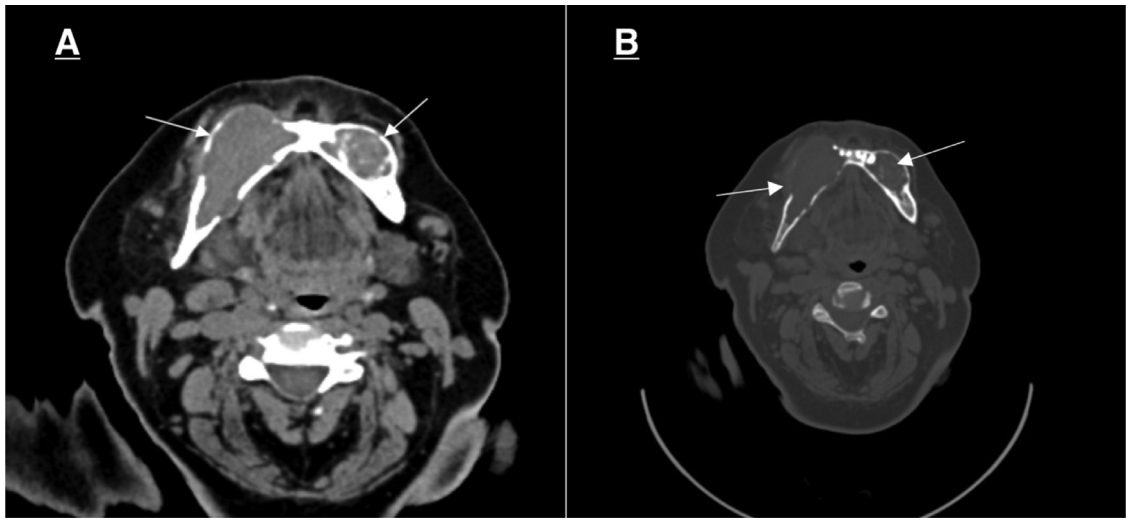

Fig. 2 – (A) A noncontrasted axial CT image of the mandibular bone shows a well-defined expansile lytic lesions of the bilateral sides of the mandibular bone. (B) Bone window axial CT image at the level of the mandible shows two well-defined expansile lytic lesions of the bilateral sides of the mandible, on the right it appears larger and shows bone destruction.

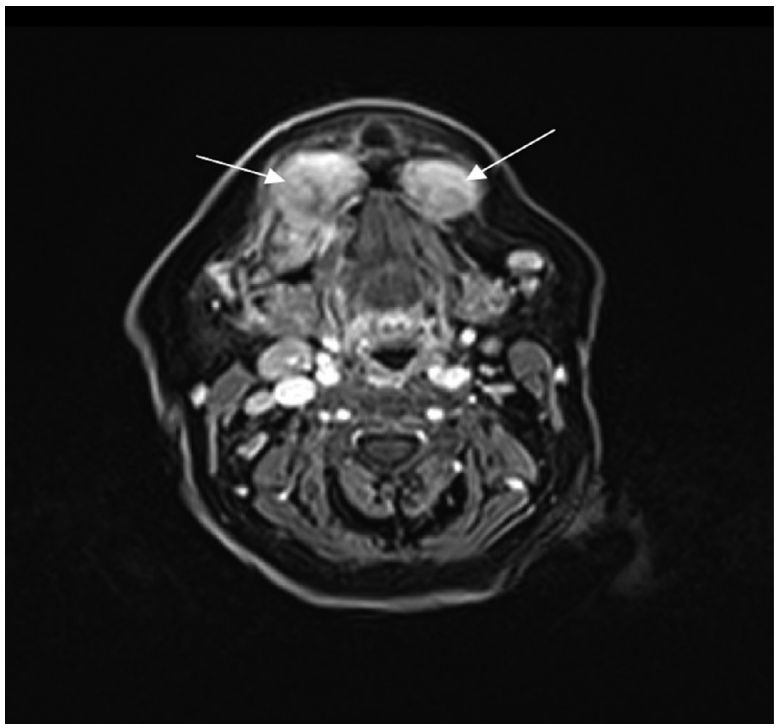

Fig. 3 – An axial T1 fatsat contrast-enhanced MRI of the mandible, showing a moderate enhancement of the mandibular lesions.

[2–4]. In addition, the existence of parathyroid tissue within parathyroid cysts walls is pathognomonic [7].

Due to the substantial differences in parathyroid cyst's location and functional status, they display considerable variety in clinical presentations [5,6]. In addition, although most parathyroid cysts present asymptomatically and are found during surgery or on imaging indicated for other reasons, some larger and more functional cysts may cause symptoms of obstruction or compression such as swallowing and breathing difficulties, change in voice, symptoms of hypercalcemia such as kidney stones, bone pains, and constipation, or even parathyroid crisis [2, 5–7].

Several imaging modalities such as ultrasound, computed tomography, or Tc-99m Sestamibi scintigraphy can be used to detect the presence of functional cystic parathyroid adenomas and parathyroid cysts [4–6]. However, ultrasound is considered

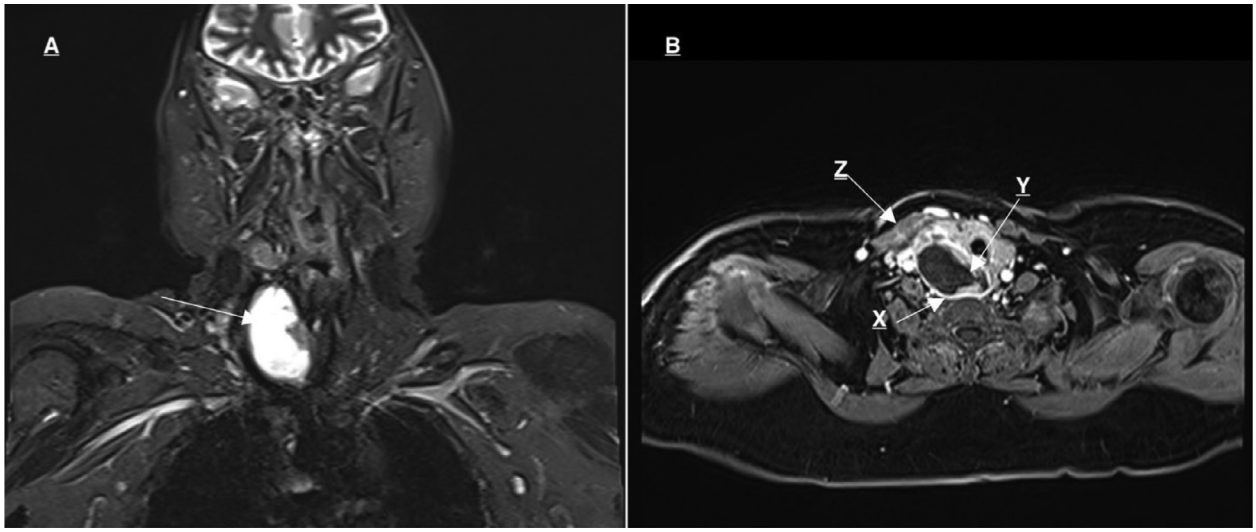

Fig. 4 – An MRI showing: (A) A coronal T2 fatsat image of the neck shows a well-defined cystic lesion with irregular wall. (B) An Axial T1 fatsat contrast enhanced image at the level of the thyroid showing: X: Well defined cystic lesion posterior to the right thyroid lobe pushes it anteriorly. Y: Peripheral irregular thick wall enhancement. Z: peri-lesional Fluid due to recent cyst aspiration.

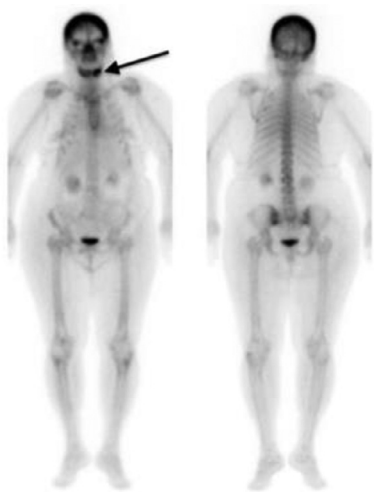

Fig. 5 – Bone scintigraphy using Tc 99m MDP showing an abnormal focal uptake in the left and right sides of the mandible representing the Brown tumors. No other skeletal focal uptake to indicate other Brown tumors.

the best initial modality as its noninvasive, can help in differentiating the cystic nature of the mass, and can be done along with fine needle aspiration, which is of great diagnostic and therapeutic value in such cases [6,7]. It's worthy to mention that Tc99-m Sestamibi scintigraphy may reveal a false negative result due to a lack of uptake of the tracer [2,5]. However, histopathology of the cysts remains crucial for the definitive diagnosis [7].

Furthermore, as functional parathyroid cysts cannot be definitively distinguished from other masses such as thyroid cysts and nodules, fine needle aspiration remains an important diagnostic and a potential therapeutic technique in such cases, which will reflect on the management [3,4]. In parathyroid cysts, the aspirated sample will reveal elevated levels of PTH, in contrast to thyroid cysts which will reveal high levels of thyroid hormones for example [2,3,5]. However, although PTH levels are more elevated in functional parathyroid cysts, it can also be high in nonfunctional cysts [3]. Serum PTH and calcium levels are used to distinguish if the cyst is functional or not in such cases [2,5].

Several options are available for the management of parathyroid cysts; of which simple cystic aspiration and sclerosing therapy, through injecting tetracycline or ethanol, can be considered especially in nonfunctioning and small cysts [5–7]. However, surgical resection remains the best option for the management of functional cysts, large cysts, or whenever there is diagnostic uncertainty or suspicion of malignancy [3,5]. In addition, surgical management is superior as complications like the recurrence of the cyst or fibrotic changes which may affect recurrent laryngeal nerve function are less likely to occur than with other approaches [5–7]. Furthermore, surgical resection of such cysts should be monitored via following up of PTH and calcium levels to make sure of good response to treatment and to avoid hypocalcemia [2].

As our patient was shown to have a bilateral brown tumor of the mandible, it's worthy to mention that the end stage of hyperparathyroidism bony disease is the creation of brown tumors [8]. Brown tumors presenting as the primary manifestation of hyperparathyroidism is a seldom finding, it can be noticed as a palpable swelling in the affected area [8]. With that being said, the reported prevalence of brown tumors is reported to be only 0.1% [9]. In addition, only 3% of primary hyperparathyroidism patients suffer from brown tumors and only 4.5% of 220 hyperparathyroidism patients demonstrated mandibular brown tumors in another study [8,10].

Conclusion

Cystic parathyroid adenoma is a rare entity, which can be functional or nonfunctional. Functional ones increase parathyroid levels in the serum thus calcium levels. As a consequence, those patients will present with signs and symptoms related to increased calcium levels, such as kidney stones, abdominal pain, constipation, compressive symptoms, or even brown tumor, which mandates management of such cases. Moreover, false negative results with Tc-99m Sestamibi and misdiagnosis as thyroid cysts or other lesions mandate the use of more specific diagnostic modalities and a lower threshold for diagnosis since such lesions can be cured completely. Our case provides a radiological illustration of a functional cystic parathyroid adenoma and brown tumor.

Patient consent

Written informed consent for the publication of this case report was obtained from the patient.

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