

**Case Report**

# Parathyroid Adenoma Arising from Autotransplanted Parathyroid Tissue in Sternocleidomastoid Muscle: A Case Report with Review of the Literature

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**Total parathyroidectomy followed by autotransplantation in patients with renal hyperparathyroidism to prevent hypoparathyroidism is a relatively common surgical procedure. However, parathyroid adenoma arising in autotransplanted parathyroid tissue in patients with secondary hyperparathyroidism is very rare. To date, less than 20 such cases have been reported in the English literature. Here we report a case of a 47-year-old African-American male with a history of end stage renal disease on dialysis undergoing a total parathyroidectomy and autotransplantation in 2001, presenting with severe hyperparathyroidism and enlarged transplanted parathyroid tissue 6 years later. The palpable mass at the site of autotransplantation was excised. Grossly, the cut surface of the mass appeared brown-tan. Microscopically, the mass was well circumscribed and composed of sheets of small, round and relatively uniform cells, morphologically consistent with chief cells. No adipose tissue or oxyphil cells were found inside the mass. Cellular atypia was not identified. Based on the above morphological features and the patient's history, the diagnosis of parathyroid adenoma arising from autotransplanted tissue was established. This case illustrates that parathyroid adenoma arising from autograph can cause hyperparathyroidism.**

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**Key Words:** hyperparathyroidism, parathyroid adenoma, autotransplant

## INTRODUCTION

Parathyroid adenoma arising in autotransplanted tissue in patients with recurrent secondary hyperparathyroidism (rSHPT) is very rare. Since its first description in 1968, autotransplantation of parathyroid tissue into forearm or the sternocleidomastoid muscle to prevent hypoparathyroidism after total parathyroidectomy gained more and more attention and interest.<sup>1</sup> However, rSHPT arising from the autotransplant represents a major diagnostic and therapeutic challenge.<sup>2</sup> Here we report a case of parathyroid adenoma arising from autotransplanted parathyroid tissue into the sternocleidomastoid muscle in a 47-year-old African-American male. In this report, we describe the clinical course, pathological features and further review of the literature.

## CASE REPORT

The patient was a 47-year-old African-American male with a history of parathyroid hyperplasia of four parathyroid glands secondary to renal failure undergoing total parathyroidectomy and autotransplant of a portion of parathyroid tissue into the sternocleidomastoid muscle in 2001. The patient was normocalcemic after the surgery. He presented with severe hyperparathyroidism and increasing

the size of autotransplant in 2007. The palpable mass at the site of autotransplantation was excised and sent for pathological evaluation. His past medical history was significant for polycystic kidney disease, end-stage renal disease with hemodialysis for 14 years, and clear cell renal cell carcinoma of kidney status post right nephrectomy in 2006.

## PATHOLOGIC EXAMINATION

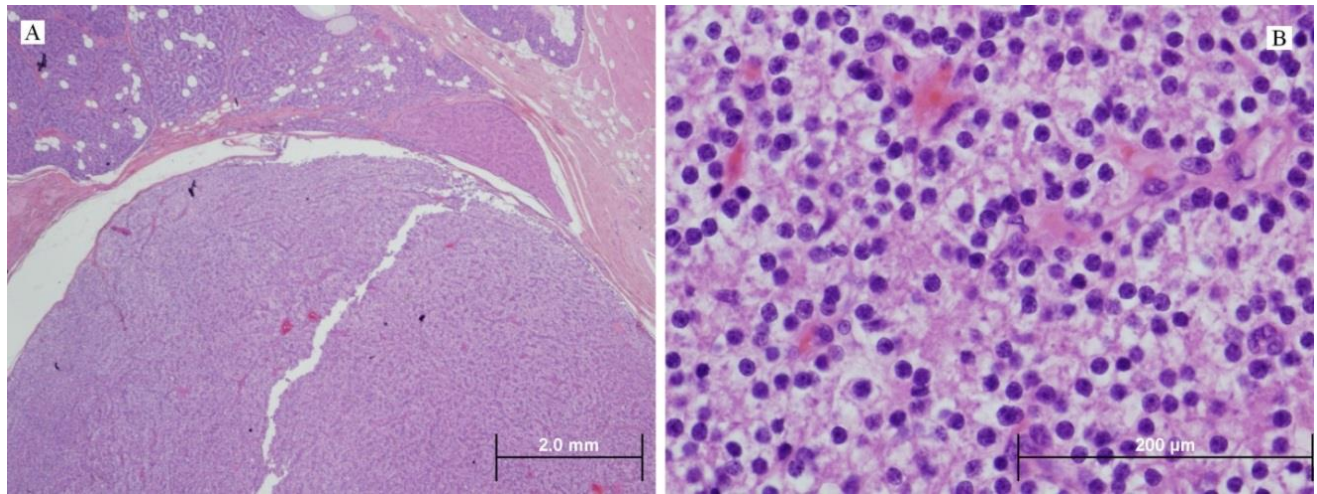
**Gross appearance:** Two pieces of tan-pink to brown firm soft tissue was received, weighing 2.7 grams and measuring 2 x 1.9 x 1.2 cm and 1.9 x 0.7 x 0.4 cm. The specimen is serially sectioned to reveal a tan-pink to brown homogenous cut surface. The specimen is submitted entirely for microscopic examination.

**Microscopic examination:** The mass was well circumscribed and encapsulated (**Figure 1A**). A rim of normal parathyroid tissue was present at the periphery of the tissue. The mass was composed of sheets of small, round and relatively uniform cells with pale or clear cytoplasm (**Figure 1B**). These cells were morphologically consistent with chief cells. No oxyphil cells were present. In addition, no adipose tissue could be identified inside the mass. No cellular atypia was appreciated.

**Pathologic diagnosis:** Parathyroid adenoma arising from autotransplanted tissue.

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**Figure 1.** Parathyroid adenoma arising from autotransplanted parathyroid tissue.

**A.** The mass was well circumscribed and encapsulated with fibrous tissue. No adipose tissue was present. A rim of normal parathyroid tissue admixed with adipocytes was compressed to the periphery (Hematoxylin-eosin stain: original magnification x100).

**B.** The mass was composed of sheets of small, round and relatively uniform cells, morphologically consistent with chief cells (Hematoxylin-eosin stain: original magnification x400).

## DISCUSSION

Renal insufficiency is a common cause of secondary hyperparathyroidism (SHPT). Surgical treatment for SHPT is required in 2.5-8% of patient with dialysis.<sup>3</sup> In general, two highly competitive surgical treatment options are being used. Subtotal parathyroidectomy with leaving a portion of normal parathyroid gland is performed, especially in younger patient with the potential for kidney transplantation. On the other hand, total parathyroidectomy with autotransplant has gained more attention and become a relative common procedure. However, given the pathophysiology, rSHPT arising from the autotransplanted parathyroid tissue are substantial and the recurrence rate is around 10%.<sup>2</sup> The histopathological feature of the autotransplant tissue after rSHPT is usually consistent with hyperplasia. However, adenoma arising in autotransplanted tissue can also occur with 4% incidence rate.<sup>2,4</sup> To date, less than 20 such cases have been reported in the English literature. These patients usually develop rSHPT 3-10 years after the initial total parathyroidectomy with autotransplantation and require surgical therapy.

The three main criteria to histologically diagnosis of adenoma are the rim of normal parathyroid tissue around the capsule, diffuse chief cell growth and absence of adipose tissue inside the mass. The lesion in our case is consistent with the parathyroid adenoma morphologically. However, the distinction between adenoma and nodular hyperplasia has not been conclusive with histological, immunohistochemical and DNA examination.<sup>5</sup> With patient's history of autotransplant with hyperplastic parathyroid tissue, the possibility of nodular hyperplasia cannot be totally ruled out. However, the etiology of original hyperplastic tissue developing into adenoma is unknown.

The graft dependent rSHPT represents a major diagnostic, therapeutic challenge and disadvantage. Factor involved in the recurrence of hyperparathyroidism has been suggested to be nodular hyperplasia, as graft hyperfunction is significant higher in patient received nodular gland than diffuse gland.<sup>6</sup> In addition, a recent study indicated that intra-operative tissue selection of parathyroid tissue with low proliferative potential will lower the rate of recurrence.<sup>7</sup>

The high incidence of rSHPT argues for some alternative surgical methods for patient with secondary hyperparathyroidism from renal failure, especially in patient with dialysis. Brennan et al reported low recurrence rate of their patients with autotransplantation of cryopreserved tissue that has been frozen from 45 days to 18 months.<sup>8</sup> Agha et al also proposed an alternative method of treatment without autotransplantation after the initial surgery of total parathyroidectomy.<sup>2</sup>

In conclusion, we demonstrated the clinical course and histopathologic features of a rare case of parathyroid adenoma arising from autotransplant tissue in a patient with secondary hyperparathyroidism.

## CONFLICT OF INTEREST

Authors have no conflict interest to report.

## ETHICAL APPROVAL

This work meets all the ethical guidelines.

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