



Recurrent Primary Hyperparathyroidism after Autotransplantation of an Excised Parathyroid Adenoma

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Abstract

Introduction: We present a patient with recurrent primary hyperparathyroidism after re-implantation of a portion of an excised parathyroid adenoma. To our knowledge, there has been only one other case reported in the literature describing a re-implanted parathyroid adenoma as the culprit for recurrent primary hyperparathyroidism.

Case Description: A 65-year-old female presented with recurrent primary hyperparathyroidism. She had undergone a left superior parathyroidectomy for hyperparathyroidism eight years ago and previously had undergone a total thyroidectomy. The intraoperative intact parathyroid hormone (iPTH) level at the time of her arathyroidectomy reportedly dropped from a baseline of 93.8 pg/mL to 11.7 pg/mL at twenty minutes post-excision. Due to concern for a lack of functioning parathyroid tissue on the contralateral side, a portion of the excised gland was re-implanted into the left sternocleidomastoid (SCM) muscle. Over the following seven years, her bone density declined to osteoporosis, and she was found to have an elevated serum calcium level of 10.8 mg/dL (reference: 8.6 mg/dL to 10.4 mg/dL) as well as an elevated iPTH level of 79 pg/mL (reference: 10 pg/mL to 65 pg/mL). Urinary calcium was additionally elevated at 273 mg/24 h (reference: 35 mg/24 h to 250 mg/24 h). A CT scan of the neck demonstrated a 2 cm left-sided mass within the inferior portion of the left SCM. The patient therefore underwent a re-operative neck exploration. Baseline iPTH was 75 pg/mL. Intra-operative findings revealed abnormal parathyroid tissue within the left SCM muscle. Post-excision iPTH level was 47 pg/mL. The surgical pathology demonstrated a 2.4 cm mass composed of hyper-cellular parathyroid tissue and skeletal muscle.

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Introduction

We present a patient with recurrent primary hyperparathyroidism after re-implantation of a portion of an excised parathyroid adenoma. To our knowledge, there has been only one other case reported in the literature describing a re-implanted parathyroid adenoma as the culprit for recurrent primary hyperparathyroidism.

Case Presentation

A 65-year-old female presented with recurrent primary hyperparathyroidism. She had a history significant for papillary thyroid carcinoma and had undergone total thyroidectomy 14 years prior. Subsequently (8 years prior), during workup for osteopenia, she was diagnosed with hyperparathyroidism and underwent a left superior parathyroidectomy. As per the operative record, intraoperative intact parathyroid hormone (iPTH) level dropped from a baseline of 93.8 pg/mL to 11.7 pg/mL at twenty minutes post-excision. Due to concern for a lack of functioning parathyroid tissue on the contralateral side, a portion of the excised gland was re-implanted into the left sternocleidomastoid (SCM) muscle. Pathology was interpreted as cellular parathyroid, weighing 318 mg.

Over the following seven years her bone density declined to osteoporosis. She denied nephrolithiasis or fractures, however was found to have an elevated serum calcium level of 10.8 mg/dL (reference: 8.6 mg/dL to 10.4 mg/dL), as well as an elevated iPTH level of 79 pg/mL (reference: 10 pg/mL to 65 pg/mL). Urinary calcium was additionally elevated at 273 mg/24 h (reference: 35 mg/24 h to 250 mg/24 h).

A CT scan of the neck demonstrated a 2 cm left-sided mass within the inferior portion of the left SCM muscle. SPECT CT confirmed a focus of radiotracer at this site which was felt to most likely represent a parathyroid adenoma.

The patient was taken to the operating room for a re-operative neck exploration and planned excision of the left SCM muscle parathyroid adenoma. Baseline iPTH was 75 pg/mL. A left sided incision was made in a skin crease overlying the SCM muscle. Sub-platysmal flaps were elevated allowing for exposure of the SCM muscle. Several clips were identified along the anterior surface of the muscle. Following dissection, there was evident abnormal parathyroid tissue posterior to the clips. A wide excision of this tissue was performed to include the clips as well as some adjacent muscle. A 1.2 cm × 0.8 cm × 0.5 cm portion of the specimen was sent for frozen section which confirmed hyper-cellular parathyroid tissue and histologically unremarkable skeletal muscle, weighing 180 mg. An additional portion was sent for cryopreservation. Post-excision iPTH level was 56 pg/mL at 15 minutes. A repeat iPTH was 47 pg/mL approximately 1 h post-op.

Final pathology of the remainder of the specimen demonstrated a 2.4 cm × 1.2 cm × 0.8 cm mass composed of hyper-cellular parathyroid tissue and histologically unremarkable skeletal muscle, weighing 920 mg. Follow-up labs at two weeks post-op revealed a serum calcium level of 9.8 mg/dL and iPTH of 40 mg/dL, and at six months post-op 10.3 mg/dL and 44 mg/dL, respectively.

Discussion

Recurrent primary hyperparathyroidism has been well described in the literature. This has been cited to most commonly be secondary to missed parathyroid adenomas during prior surgery (either in their normal or ectopic locations), re-growth of a single adenoma due to incomplete resection, or failure to resect multiple abnormal glands [1]. The phenomenon of a re-implanted parathyroid gland resulting in recurrent hyperparathyroidism is also well reported in the literature; however, this has typically been associated with cases of secondary hyperparathyroidism. Recurrence in these cases is thought to be due to hyper-function of either the transplanted hyperplastic parathyroid tissue (after four gland parathyroidectomy with autotransplantation) or the hyperplastic parathyroid remnant (in the instance of a 3½ gland resection) [2]. To our knowledge, there has only been one other case reported in the literature of recurrent primary hyperparathyroidism secondary to an auto transplanted parathyroid adenoma [3].

The apparent infrequency of parathyroid adenoma auto transplantation is most likely a result of a conscious effort on the surgeon's behalf to prevent the anticipated recurrence of primary hyperparathyroidism. It has historically been felt that parathyroid adenomas are largely monotypic entities arising from clonal expansion of a single transformed progenitor cell [4]. Autotransplantation of such tissue would therefore be expected to continue monoclonal expansion, resulting in a recurrence of the adenoma.

Thus, when considering autotransplantation of a parathyroid adenoma, the surgeon must weigh the probable risk of recurrence against the expected risk of rendering the patient with permanent hypoparathyroidism.

It is generally accepted that a decrease in intraoperative iPTH of at least 50% from baseline and into the normal range, or to a final

level of <40 pg/mL is indicative of successful parathyroid surgery [5]. Furthermore, it has been suggested that patients with final intraoperative iPTH <40 pg/mL are felt to have lower rates of recurrence [6]. At what final iPTH level, however, should one be concerned about the patient developing permanent hypoparathyroidism? Currently, there are no established guidelines for which autotransplantation is recommended in the setting of preventing permanent hypoparathyroidism after parathyroidectomy. Most of the literature describing indications for parathyroid re-implantation has been in the setting of preventing permanent hypoparathyroidism after total thyroidectomy. An intraoperative iPTH level <10 ng/L at 10 min to 20 min after total thyroidectomy has been suggested as a threshold for autotransplantation to decrease the risk of permanent postoperative hypoparathyroidism [7]. While this recommendation may be applied to parathyroidectomy, there remains a need to establish set guidelines through future clinical trials.

Finally, cryopreservation of the excised parathyroid tissue is an alternative one might consider to immediate autotransplantation. This allows for the potential to re implant this tissue at a later time should the patient prove to become persistently hypoparathyroid. This has been reported in the literature to be successful after as long as eighteen months of cryopreservation [8].

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