Generalized pruritus is a well-known complication of primary hyperparathyroidism [1,2]. We present a case of an intractable localized inguinal pruritus secondary to a parathyroid adenoma that completely resolved after subtotal parathyroidectomy. To our knowledge, this is the first case of primary hyperparathyroidism presenting as a localized inguinal pruritus.

REPORT OF THE CASE

A 63-year-old man presented with a severe disabling persistent bilateral inguinal pruritus that started eight months ago and had been resistant to topical corticosteroids and antifungals. Physical examination revealed mild hyperpigmentation with secondary excoriations and no sign of eczema or fungal infection.

The patient underwent extensive work-up including blood exams (inclusive of creatinine blood levels), skin biopsy, bone mineral density (BMD), chest X-ray, pelvis and abdominal ultrasound; all were normal except for mild hypercalcemia (10.6 mg/dl) and hypophosphoremia (2.8 mg/dl).

Further investigations showed normal levels of 25-hydroxyvitamin D, elevated levels of 24-hour urinary calcium excretion (250 mg/24h) and of parathyroid hormone (PTH) (120 pg/ml), as well as a 26 x 6.5 x 13 mm left subthyroid hypoechoogenic nodular mass. A sestamibi parathyroid scintigraphy confirmed the diagnosis of solitary parathyroid adenoma.

Multiple gland disease was found intraoperatively and a subtotal parathyroidectomy was performed; histology confirmed the diagnosis of multiple benign adenomas. Pruritus completely resolved two weeks later with no sign of recurrence on follow-up visits. Patient had no family history of multiple gland disease or hypercalcemia.

DISCUSSION

Primary hyperparathyroidism (HPT) occurs in 0.2% to 0.5% of the population and is due to a hypersecretion of PTH by a parathyroid adenoma or hyperplasia [1-3]. It is the most common cause of hypercalcemia in outpatients and second to cancer in inpatients [3]. Whereas parathyroidectomy is mainly indicated in markedly elevated serum calcium (>12 mg/dl) and for patients with renal, bone and neuromuscular symptoms, cutaneous symptoms alone are still not included in the current indications of parathyroidectomy [1,4-6]. In this case, parathyroidectomy was not indicated by guidelines, but in view of the history and the severe intractable pruritus, parathyroidectomy was the only available therapeutic option. Cutaneous signs, even alone, might therefore be added to the indications of parathyroidectomy in primary HTP, especially that pruritus improved after parathyroidectomy in other studies as well [7].

Cutaneous manifestations of primary HPT are in fact rare and include generalized pruritus and deposition of calcium in the skin [1]; chronic urticaria was also reported once [8]. As in uremic pruritus, the pathophysiology of pruritus in HPT is not known and probably multifactorial. Some studies showed that hyperparathyroidism may induce itch by stimulating mast cells to release histamine and by promoting microprecipitation of calcium.
and magnesium salts in the skin [9]. The role of PTH in the genesis of pruritus in primary and secondary hyperparathyroidism is also unclear; some studies reported significant association between serum PTH levels and incidence and severity of itching [6], while others found no specific relationship between uremic pruritus, hypercalcemia and level of PTH [9,10]. The importance of this case is the fact that pruritus was only localized to the inguinal area, suggesting that local factors (maceration, friction, allergic sensitization...) may also contribute to the pathogenesis of pruritus in HTP. Local factors might indeed trigger release of histamine, of neurotransmitters like substance P and hormones like PTH [10], which facilitate precipitation of salts in the skin [9] or favor pruritogenic cytokine release [11], all contributing to the genesis of pruritus in HPT.

In conclusion, this case discusses the pathogenesis of pruritus in primary and secondary HPT and suggests that intractable cutaneous symptoms like pruritus might be added to the current approved indications of parathyroidectomy in primary HPT. Further studies with larger groups are still needed to better understand the exact mechanisms of pruritus in HPT.

CONFLICT OF INTEREST: None to declare.

REFERENCES


