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Epidemiology and Prevention

Primary hyperparathyroidism presenting as slipped capital femoral epiphysis; a patient assessment and literature review

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Abstract

Primary hyperparathyroidism associated with slipped capital femoral epiphysis (SCFE) is rare. We report a case of a 14-year-old girl who presented with limping of gait and was diagnosed to have bilateral SCFE and underwent screw fixation of both hips. She had pain involving multiple joints and tenderness on further evaluation and was found to have biochemical hyperparathyroidism. The patient was initially managed medically, and further investigations revealed a parathyroid lesion in the right superior location. Workup for multiple endocrine neoplasia was negative. She underwent right superior parathyroid adenoma excision, and post-operatively, she was cured of the disease. On follow-up, she is asymptomatic, and her calcium is within normal limits. A literature review showed that SCFE is associated with severe hyperparathyroidism in adolescents.

Keywords: Primary hyperparathyroidism, Slipped capital femoral epiphysis, parathyroid adenoma

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Introduction

Primary hyperparathyroidism is rare in children. Most of the cases are associated with single parathyroid adenoma. These patients have symptomatic hypercalcemia or complications like renal stones, abdominal pain, and skeletal fragility. Primary hyperparathyroidism is a rare cause of slipped capital femoral epiphysis (SCFE), and only 13 cases have been reported in the literature worldwide. Appropriate management of primary hyperparathyroidism in children and adolescents requires distinguishing between familial hypocalciuric hypercalcemia, which generally requires no specific treatment, and other causes of primary hyperparathyroidism that requires parathyroidectomy.

We present the case of a 12-year-old girl who complained of hip pain and limping of gait, persisting for several months. Upon evaluation in the orthopedic department, bilateral SCFE was detected through radiological assessment (Figure 1A). After ruling out any secondary causes of SCFE, the patient underwent bilateral cancellous screw fixation. However, her bone pain and fatigue did not improve post-surgery, prompting a referral to the endocrine surgery department.

During a physical examination, the patient was alert and had a slender build with a body mass index (BMI) of 17.3 kg/m^2. Bony tenderness was observed over multiple bones, and she displayed normal height and pubertal development. Additionally, a swelling on the right side of her neck moved with deglutition (swallowing). Routine investigations, including renal and liver function tests (Table 1), yielded normal results. However, biochemical analysis revealed hypercalcemia with a calcium level of 16.3 mg/dL (normal range; 8.5-10.2 mg/dL) and hypophosphatemia with a phosphorus level of 2.1 mg/dL (normal range; 3.5-4.5 mg/dL). Her vitamin D level was within the normal range at 38.4 ng/mL (normal range; 30-50 ng/mL). However, she had a significantly elevated parathyroid hormone (PTH) level of 1147.9 pg/mL (normal range; 25-67 pg/mL) and an elevated alkaline phosphatase (ALP) level of 850 IU/L (normal range; 30–120 IU/L). Meanwhile, X-ray of the spine exhibited a rugger jersey appearance (Figure 1B).

Considering the primary hyperparathyroidism indicated by the biochemical findings, further imaging studies were conducted, including ultrasound of the neck (Figure 2A), parathyroid scintigraphy [Technetium-99m sestamibi (or MIBI)] scan (Figure 2B), and magnetic resonance imaging (MRI) of the neck (Figure 3A). These imaging studies revealed a 40×20×13 mm lesion located posterior to the right lobe of the thyroid gland, most likely corresponding to the right superior parathyroid lesion. Notably, the parathyroid scintigraphy did not show any

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Implication for health policy/practice/research/ medical education

Primary hyperparathyroidism is rare in children, and only 13 cases are reported in the literature, as presented by SCFE. Parathyroidectomy is the best treatment for primary hyperparathyroidism with a localized parathyroid lesion.

focal MIBI concentrating lesion.

Given the occurrence of primary hyperparathyroidism at a young age, multiple endocrine neoplasia type 1 was ruled out based on a detailed history and normal serum prolactin levels. Due to the patient's short QT interval and extremely high calcium levels, initial management involved intravenous fluids, loop diuretics, and calcitonin injections.

Subsequently, the patient underwent a right superior parathyroidectomy (Figure 3B). During the surgery, intra-operative PTH measurement showed a significant decrease in serum PTH levels, dropping to 38.6 pg/mL from the preoperative level of 1147.9 pg/mL, indicating successful removal of the diseased parathyroid gland. Following the surgery, her serum calcium returned to the normal range. The patient received calcium and activated vitamin D supplements to manage postoperative hypocalcemia for one month. Histopathological analysis of the excised tissue

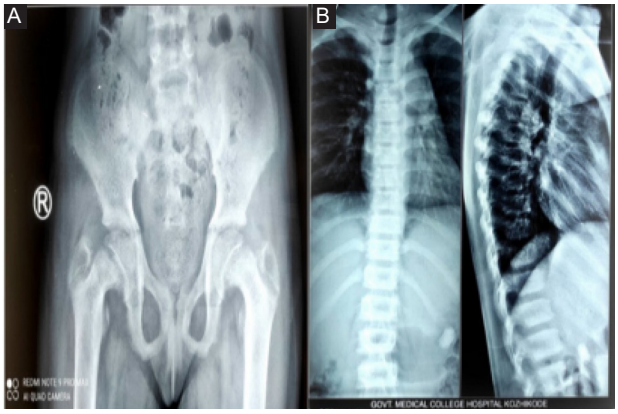

Figure 1. (A) X-Ray of hip showing bilateral slipped capital femoral epiphysis. (B) X-ray of the spine showing Rugger jersey appearance.

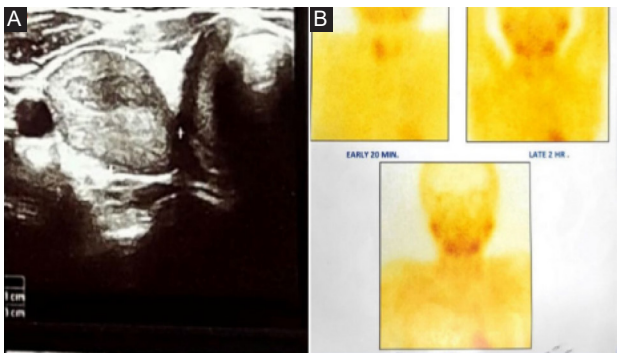

Figure 2. (A) Ultrasound of neck showing parathyroid lesion. (B) Sestamibi showing no tracer uptake in the delayed phase.

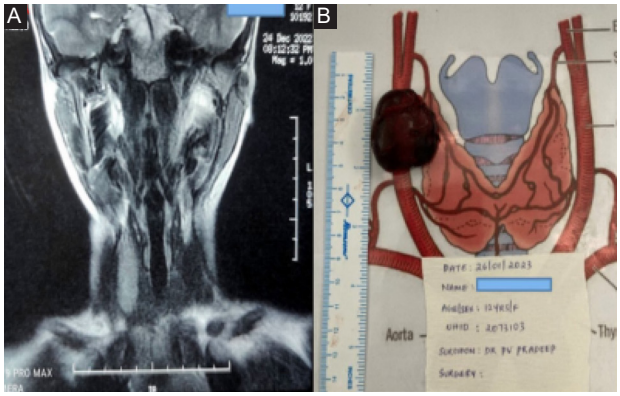

Figure 3. (A) MRI Neck showing right-sided parathyroid lesion.(B) Specimen photograph of parathyroid adenoma excision.

revealed an adenoma in the right superior parathyroid gland. Fortunately, her symptoms improved immediately after the surgery, and her gait returned to normal after one month. She is currently under regular follow-up, and her calcium and phosphorus levels are normal, with no requirement for calcium supplements.

Among teenagers, the most common hip disorder is SCFE. Our case illustrated SCFE in a young teenager due to primary hyperparathyroidism, a rare cause of SCFE (1). The most common endocrine cause of SCFE is hypothyroidism. Other endocrine causes include hypogonadism, hypopituitarism, growth hormone treatment, and rarely primary hyperparathyroidism (2). SCFE associated with primary hyperparathyroidism has been reported in 13 cases in the literature until now (Table 2). Pathophysiology of SCFE with primary hyperparathyroidism is due to abnormal PTH levels, which may lead to abnormal cartilage mineralization, which delays the epiphyseal fusion (3). Additionally, PTH receptors are present in the cells of hypertrophied cartilage zone of the epiphyseal plate- the same zone

Table 1. Laboratory investigations

	Hormone	Normal range
Serum calcium (corrected)	16.3 mg/dL	8.6-10 mg/dL
Serum phosphorus	2.1 mg/dL	3.5-4.5 mg/dL
Alkaline phosphatase	850 IU/L	35-129 IU/L
Serum creatinine	0.5 mg/dL	0.55-1.02 mg/dL
Vitamin D	38.66 ng/mL	20-30 ng/mL
PTH	1147.9 pg/mL	10-75 pg/mL
Post op PTH 10 min post excision	38.6 pg/mL	10-75 pg/mL
Serum calcium post-op day 1 (corrected)	8.7 mg/dL	8.6-10 mg/dL
Serum calcium post-op day 2 (corrected)	8.2 mg/dL	8.6-10 mg/dL
Serum calcium post-op day 30 (corrected)	8.4mg/dL	8.6-10 mg/dL
TSH	3.5 mIU/L	0.5-4.5 mIU/L
Prolactin	4.72 ng/mL	<25 ng/mL

PTH, Parathyroid hormone; TSH; Thyroid stimulating hormone.

Table 2. Case reports of slipped capital femoral epiphysis presented as primary hyperparathyroidism

Author	Age/gender	Calcium mg/dL	S.PTH pg/mL	Histopathology	Remarks
Chiroff et al (2)	11/Male	4.0 (Ionized)	-	Single adenoma	B/L SCFE
Bone et al (8)	13/Female	6.0 (Ionized)	451	Single adenoma	Spontaneous resolution after parathyroidectomy
Kinoshita et al (9)	16/Male	11.5	340	Single adenoma	B/L SCFE and Parathyroid surgery in the same sitting
Yang et al (10)	13/Male			Single adenoma	B/L SCFE and parathyroidectomy 3 months apart
Quadan et al (11)	13/Female	15.6		Single adenoma	Hypercalcemia responded to IV palmidronate
Khiari et al (12)	16/male	3.3 mmol/dL		Single adenoma	B/L SCFE, only parathyroidectomy done
Madeira et al (13)	18/Male	13.6	1529	Single adenoma	Managed conservatively
Alghamdi et al (14)	13/Female	2.91 mmol/dL	2253	Single adenoma	Parathyroidectomy followed by B/L SCFE fixation
El Scheich et al (15)	15/Male	3.52 mmol/dL (Ionized)	172	Single adenoma	Parathyroidectomy followed by B/L SCFE fixation
Bhadada et al (16)	12/Female	10.4	1523	Single adenoma	B/L SCFE fixation followed by Parathyroidectomy
Kim et al (17)	14/Male	11.8	1299	Single adenoma	Simultaneously parathyroidectomy and B/L SCFE fixation
Serrano-Gonzalez et al (18)	14/Female	13.4	1013	Parathyroid carcinoma	Right U/L SCFE fixation followed by parathyroidectomy
George et al (19)	15/Male	17.2	1052	Single adenoma	B/L SCFE fixation followed by Parathyroidectomy
Our case	12/Female	14.4	1148	Single adenoma	B/L SCFE fixation followed by Parathyroidectomy

where the slipping of femoral epiphysis occurs (3). Recent community-based data from Sweden showed that the incidence of SCFE is mildly increased for girls, and obesity is a significant characteristic for boys (4). Hypercalcemia, alkaline phosphate, and high PTH levels were seen in most of the cases. Higher the biochemical parameters, the severity will be high, and the chance of SCFE is high.

Adolescents with unstable SCFE should be managed judiciously, as joint avascular necrosis is possible. However, the optimal time for fixing the joint is controversial—the treatment aimed to prevent further slippage and complications like avascular necrosis and chondrolysis (5). In case of emergency, in situ fixation of SCFE was conducted with pins or screws through the pelvis.

Severe hypercalcemia and its manifestations should also be considered. In these cases, use medical management to stabilize the calcium levels, as done in our case, and then plan for parathyroidectomy. In the literature, 11 cases are due to single parathyroid adenoma, whereas in one case, it is parathyroid carcinoma. In 11 cases of SCFE with primary hyperparathyroidism, only one case has ectopic mediastinal parathyroid adenoma (6).

In the pediatric age group with primary hyperparathyroidism, genetic testing is advisable as it relates to genetic syndromes in 50% of patients, including the calcium-sensing receptor and parathyroid cell proliferation pathways. Genetic testing is warranted even if our patient has no significant family history of parathyroid disease or any other endocrine tumor (7,8). Nevertheless, the family declined genetic testing due to financial constraints.

Conclusion

Though SCFE is rare with primary hyperparathyroidism, it should be considered if there are symptoms of musculoskeletal involvement. Pathogenesis may be related to high PTH levels. In the evaluation of SCFE, treatable causes should be eliminated.

Authors' contribution

Conceptualization: Pradeep Puthen Veetil.
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Methodology: Shikhil Puzhakkal.
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Project administration: Pradeep Puthen Veetil.
Supervision: Pradeep Puthen Veetil.
Validation: Farhana Chathoth Kannoli
Visualization: Shikhil Puzhakkal.
Writing–original draft: Shikhil Puzhakkal.
Writing–review and editing: Pradeep Puthen Veetil.

Conflicts of interest

The authors declare that they have no competing interest.

Ethical issues

The authors have adhered to ethical guidelines, including the World Medical Association Declaration of Helsinki, in conducting this case report. The patient has provided written informed consent for the publication of this report, and the authors have also ensured ethical considerations, including avoiding plagiarism, data fabrication, and double publication.

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References

1. Roizen J, Levine MA. Primary hyperparathyroidism in children and adolescents. J Chin Med Assoc. 2012;75:425-34. doi: 10.1016/j.jcma.2012.06.012.
2. Chiroff RT, Sears KA, Slaughter WH 3rd. Slipped capital femoral epiphyses and parathyroid adenoma. J Bone Joint Surg Am. 1974;56:1063-7.
3. Crabb ID, O'Keefe RJ, Puzas JE, Rosier RN. Differential effects

of parathyroid hormone on chick growth plate and articular chondrocytes. Calcif Tissue Int. 1992;50:61-6. doi: 10.1007/BF00297299.
4. Herngren B, Stenmarker M, Vavruch L, Hagglund G. Slipped capital femoral epiphysis: a population-based study. BMC Musculoskelet Disord. 2017;18:304. doi: 10.1186/s12891-017-1665-3.
5. Peck D. Slipped capital femoral epiphysis: diagnosis and management. Am Fam Physician. 2010;82:258-62.
6. Vitale RJ, Shieh HF, Modi BP, Gordon RJ. Primary Hyperparathyroidism From Ectopic Parathyroid Adenoma in a 12-Year-Old With Slipped Capital Femoral Epiphysis. J Endocr Soc. 2022;6:bvac071. doi: 10.1210/jendso/bvac071.
7. El Allali Y, Hermetet C, Bacchetta J, Amouroux C, Rothenbuhler A, Porquet-Bordes V, Champigny MA, et al. Presenting features and molecular genetics of primary hyperparathyroidism in the paediatric population. Eur J Endocrinol. 2021;184:347-355. doi: 10.1530/EJE-20-1119.
8. Bone LB, Roach JW, Ward WT, Worthen HG. Slipped capital femoral epiphysis associated with hyperparathyroidism. J Pediatr Orthop. 1985;5:589-92. doi: 10.1097/01241398-198509000-00017.
9. Kinoshita J, Kaneda K, Matsuno T, Hosokawa Y, Nagashio R. Slipped capital femoral epiphysis associated with hyperparathyroidism. A case report. Int Orthop. 1995;19:245-7. doi: 10.1007/BF00185232.
10. Choung EW, Yang F. Slipped capital femoral epiphysis in an obese teenager. Phys Sportsmed. 2003;31:39-45. doi: 10.3810/psm.2003.07.437.
11. Qadan L, Al-Quaimi M, Ahmad A. Slipped capital femoral epiphysis associated with primary hyperparathyroidism and severe hypercalcemia. Clin Pediatr (Phila). 2003;42:439-41. doi: 10.1177/000992280304200509.
12. Khiari K, Cherif L, Ben Abdallah N, Maazoun I, Hadj Ali I, Bentaarit C, et al. Slipped capital femoral epiphysis associated with hyperparathyroidism. A case report. Ann Med Interne (Paris). 2003;154:544-6.
13. Pinheiro PM, Santos VD, Madeira CS. Valgus Slipped Capital Femoral Epiphysis (SCFE) Managed by Plaster Cast Immobilization Treatment: Seven Case Reports and Review of The Literature. J Exerc Sports Orthop. 2019; 6:1-13. doi: 10.15226/2374-6904/6/1/00178.
14. Alghamdi AA, Ahmad MM, Almalki MH. Slipped Capital Femoral Epiphysis and Primary Hyperparathyroidism: A Case Report. Clin Med Insights Endocrinol Diabetes. 2016;9:73-76. doi: 10.4137/CMED.S40895.
15. El Scheich T, Marquard J, Westhoff B, Krauspe R, Schneider A, Cupisti K, Oh J, et al. Approach to the management of slipped capital femoral epiphysis and primary hyperparathyroidism. J Pediatr Endocrinol Metab. 2012;25:407-12.
16. Bhadada SK, Menon AS, Bhansali A, Das S, Gill SS, Behera A. An unusual cause of limping in primary hyperparathyroidism. The Endocrinologist. 2009;19:122-3
17. Kim T-S, Choi W-S. Bilateral slipped capital femoral epiphysis due to primary hyperparathyroidism. J Korean Orthop Assoc. 2009;44:565–9
18. Serrano-Gonzalez M, Shay S, Austin J, Maceri DR, Pitukcheewanont P. A germline mutation of HRPT2/CDC73 (70 G>T) in an adolescent female with parathyroid carcinoma: first case report and a review of the literature. J Pediatr Endocrinol Metab. 2016;29:1005-12. doi: 10.1515/jpem-2016-0109.
19. George GS, Raizada N, Jabbar PK, Chellamma J, Nair A. Slipped Capital Femoral Epiphysis in Primary Hyperparathyroidism - Case Report with Literature Review. Indian J Endocrinol Metab. 2019;23:491-494. doi: 10.4103/ijem.IJEM_306_19.