A Neglected Case of Bilateral Hip Fractures and Other Fractures with Delayed Diagnosis of Hyperparathyroidism as Cause

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Abstract

Introduction: Primary hyperparathyroidism is a disease characterized by hypercalcemia attributable to autonomous overproduction of PTH ParaThyroid Hormone.Primary hyperparathyroidism PHPT is prevalent in approximately 1% of adult population with F;M 3;1. The disease affects multiple systems –Musculosketal, CVS, G.Urinary, Abdomen and Endocrine. Primary hyperparathyroidism present with classic signs and symptoms of hypercalcaemia. Non-specific symptoms may include muscle weakness, thirst, polyuria, anorexia and weight loss along with pathological fractures.The treatment of choice is parathroidectomy after proper investigation.

Case Report: A 40 yrs Male presented with Pain, Swelling and Deformity over Bilateral Hip, Right Shoulder, Right Hand and Right Clavicular Region. Unable to bear weight since 1 year. Along with its General Complaint of Weakness, Joint Pain, Abdominal Pain with clinical syptoms and operated right sided hip after proper investigation patient was diagnosed Hyperparathyridism with B/L Hip fracture {unusual presentation }multiple fractures with implant failure .After Parathyroidectomy and regular follow fracture fixation was done later with calcium supplementation the outcome was satisfactory.

Conclusions: Repeatedly multiple fractures must be investigated with appropriate and precise routine serum biochemical along with PTH Vitamin D Thyroid profile to diagnose endocrine disorder as in this case diagnosed as Hyperparathyroidism.With Clinical examination of neck as specific site with extremities, deformities and systemic examination is essential. USG Neck as routine investigation for this disease. After parathyroidectomy and regular follow up is done. Afterwards fracture fixation is done as secondary procedure with adequate calcium and vitamin D correction give good prognosis better union with best possible outcome. This is rare presentation of PHPT with B/L Hip fracture and other fractures deformities is an excellent example of an endocrine disease that is best managed by a multidisciplinary approach and long term patient follow up.

Keywords: Primary Hyperparathroidism PHPT, Pathological fracture, PTH Parathyroid Hormone.

Introduction

Primary hyperparathyroidism is a disease characterized by hypercalcemia attributable to autonomous overproduction of PTH ParaThyroid Hormone [1].Primary hyperparathyroidism PHPT is prevalent in approximately 1% of adult population with F;M 3;1[2] HPT Hyperparathyroidism can be classified into due to benign parathyroid adenomas (80-85%). The majority of remaining cases are due to hyperplasia of multiple parathyroid glands 10-15% with parathyroid cancer being a very rare cause, indeed (<1%) [3, 4]. In Primary hyperparathyroidism PHPT serum levels of calcium are elevated and serum levels of phosphate are decreased the excessive secretion of parathyroid hormone results in different effects in cortical and trabecular bone. In cortical bone, bone turnover increases, resulting in increased resorption at the endosteal envelope, increased cortical porosity, and thinned cortical bone. This condition is contrasted to cancellous bone, where there is reduced osteoclastic resorption and osteoblastic bone formation at individual bone multicellular units, resulting in diminished erosion depth, decreased bone formation, and decreased thickness of bone structural units. Secondary Hyperparathyroidism Hypocalcemia stimulates parathyroid hormone secretion and chronic conditions stimulate parathyroid gland hyperplasia resulting from end-organ resistance to parathyroid hormone. This can be caused by several different mechanisms including intestinal causes, impaired PTH action, and loss of calcium from the extracellular compartment. It is most commonly caused by renal disease. Tertiary Hyperparathyroidism that occurs in situations of long-standing secondary hyperparathyroidism is a condition in which the cause for secondary hyperparathyroidism has been corrected The fourth type of HPT has been recognized, which occurs due to increased PTH levels synthesized in patients with malignant diseases. The main effect of parathyroid hormone (PTH) is to increase the concentration of plasma calcium level by increasing the release of calcium and

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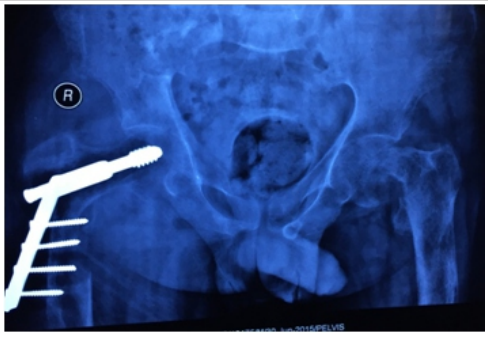

Figure 1. At time of presentation Right Intertrochanteric Fractures with failed implant with Left Hip Neck of Femur

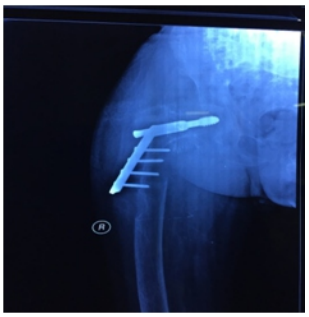

Figure 2. At time of presentation Right Intertrochanteric Fractures with failed implant

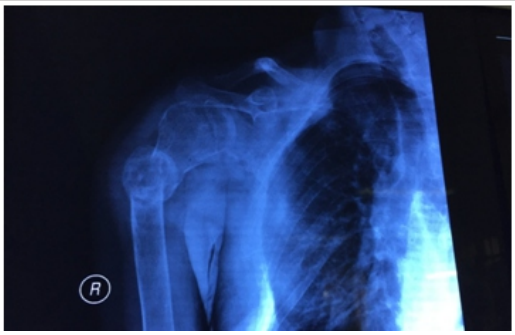

Figure 3. At time of presentation Maluinted Proximal humerus Shaft

phosphate from the bone matrix, increasing calcium reabsorption by the kidney and increasing renal production of 1.25 dihydroxy vitamin D-3 (Calcitriol) which elevates level of plasma calcium. PTH also causes phosphaturia, thereby decreasing serum phosphate level. The severe deficiency of Vit D produces lack of mineralization of bone matrix resulting in bone softening and deformity leading to pathological fracture. The disease affects multiple systems –Musculosketal, CVS, G.Urinary, Abdomen and Endocrine [5]. Primary hyperparathyroidism present with classic signs and symptoms of hypercalcaemia. Non-specific symptoms may include muscle weakness, thirst, polyuria, and anorexia and weight loss. However, the diagnosis is often made after blood investigation clinical evaluation with hypercalcaemia in an asymptomatic or symptomatic individual, with raised PTH with some cases may have normal calcium [6]. Radiologically visible as subperiosteal bone resorption , pathological fractures , cysts and brown tumor.

The persistent PHPT is mainly due to delayed diagnosis, lack of localization of ectopic adenomas, and inadequate tumor resection, which prolong the illness and complicate the clinical course [7].

Case Report

A 40 yrs Male presented with Pain,Swelling and Deformity over Bilateral Hip, Right Shoulder, Right Handand RightClavicular Region.Unable to bear weight since 1 year. Along with its General Complaint of Weakness,Joint Pain, Abdominal Pain with clinical syptoms. Past History – Patient alleged history of fall 1 year back and sustained injury. He simultaneously started developing deformities over limbs. One year back he was diagnosed Intertrochanteric fracture Right side and got operated DHS Dynamic Hip Screw in other hospital. The right hip pain persists he was not mobilized simultaneously developed left hip pain with other deformity but patient was never previously investigated for progressive deformity and cause of implant failure with other clinical symptoms in follow up. Later patient presented to us with above complaint with undiagnosed bilateral hip pain and implant failure with other clinical symptoms. Patient was investigated Blood-Routine Throid profile PTH X Rays of involved extremity which show pathological fracture multiple osteolytic lesion subperiosteal erosions in distal phanges .CT Pelvis USG AbdomenUSG ThyroidHomogenous Hypo echoic lesion in midline of lower neck inferior thyroid gland has mild flow USG Abdomenright Kidney show Calculus 13.5 mm. PARATHROID SCAN – an area increased Tc 99m concentration is seen below the lower pole of the left lobe of thyroid Hyperfunctioning Parathyroid (Fig. 1, 2, 3) General surgery unit diagnosed the case as Parathroid Adenoma and they planned for Surgery Patient underwent Parathroidectomy Inferior Lobeof parathyroid. Then postoperative patient was kept in SICU for monitoring. The complication known as bone hungrysyndrome was treated 3 Days intravenous Calcium was given then oral supplementation. Later after 5-7 days patient was discharged on traction lower limbs with calcium supplement calcitriol. Patient was admitted again after 3 months and investigation was done this are the reports (Table 1) Table Blood Report Then patient was operated for Right side - Failed Implant DHS on right with Malunion - Implant removalof DHS revision again with DHS with new implant and bone grafting was done. Left side with Fracture NOF Neck of Femur Hemiarthroplasty was done. Physiotherapy started Patient was allowed delayed weight bearing because he was having fracture of Proximal Humerus which was treated conservativelyalready malunited. (Fig. 4, 5) After 6 Weeks he started walking on walker and outcome was satisfactory with good supplementation of calcium high protein diet. The patient was able ambulate with normal gait and without pain. The last follow up visit which was 6 month to 1 year showed marked improvement in patient clinical and radiological finding. The patient was undiagnosed at previous surgery what the cause of multiple fractures and progressive deformity (Fig. 6, 7).

Table 1: a. Patient was admitted again after 3 mnths and investigation was done this are the Report

CBC	LFT	KFT	THYOID PARATHYROID	OTHERS
Hb 14	S Protein 7.4	S.Cret .4	TSH 7.34	Platelet c 3.57
PCV 44	Bilirubin .55	S.Phos 1.8	T4 11.18	
TLC 9600		S.Calcium 11.5	T3 6.26	
ESR 8	Alk Phos - 2542	I.Calcium 6.1	PTH 1803	25-OH -VIT D < 8.1
				PSA .29 ngml

Table 1: b. After 3 Months Report After Parathyriodectomy and Calcium Supplementation

CBC	LFT	KFT	THYROID	OTHERS
Hb 8.7 --> 10	S Protein 7.1	S.Cret .7		Platelet 6.17
PCV 31	Bilirubin .76	S.Phos 2.2		
TLC 7300		S.Calcium 9		
ESR 32	Alk Phos - 1159	I.Calcium 5.1	PTH 400	25-OH -VIT D 35
RBS 86				

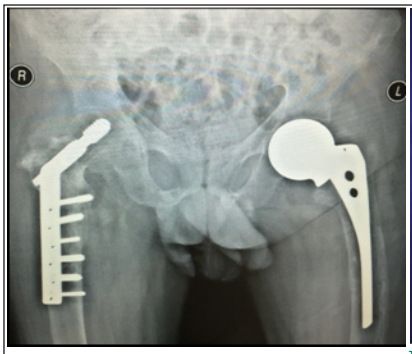

Figure 4. Post Operative X Rays after 10 Days

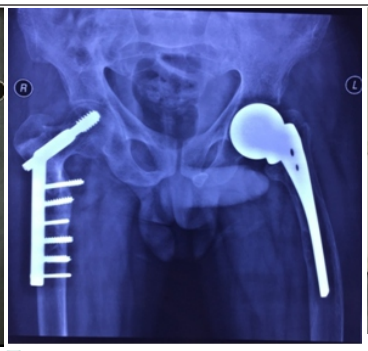

Figure 5. Post Operative X Rays after 3 Months Union

Figure 6. Post operative Partial weight Bearing

Figure 7. Post operative Full weight Bearing

Discussion

In 1891, Von Recklinghousen described the classic bone disease termed osteitis fibrosa cystic. In 1925, the Viennese surgeon Mandl performed the first parathyroid exploration and adenoma resection. Mandl noted improvement of the patient's severe skeletal abnormalities post operatively, thereby linking HPT with bone disease. Albright later in 1930s described the clinical entity of classic primary hyperparathyroidism [8,9]. This condition is more common in females. Peak age incidence is between 30 to 50 years. It is endocrine disease mostly commonly due to solitary adenoma it remain undiagnosed in developing country instead of so many cases because limited diagnostic facilities. The late presentationsof hyperparathyroidism as multiple fractures is because of dietary deficiency, especially deficiencies in protein, vitamin D and calcium. Because 50% of serum calcium is bound to albumin, protein deficiency can mask hypercalcaemia, even if a correction is made for hypoalbuminaemia e i.e. PHPT can present with normocalcaemia in protein deficient patients and clinicians should aware of this. With increased awareness, knowledge of the possible presentations ofPHPT and the availability of diagnostic facilities, late presentations can be avoided. With metabolic bone disease and multiple fractures testing of intact PTH level is core diagnosis of hyperparathyroidism increase PTH serum ionized calcium level is diagnosis of PHPT. A principal test at present is the 'Immunoassay' for PTH 1-84 as it distinguishes the hypercalcaemia of malignancy from that of hyperparathyroidism [10]. Disease results from excessive secretion of parathyroid hormone either due to solitary (50-85%) or multiple (10%) adenomas, hyperplasia (10-40%), or rarely due to a carcinoma of single parathyroid gland.Two distinct types of bone lesions are seen [11] the slowly progressive type- leads to cortical thinning and osteoporosis and other is rapidly progressive type. Pathological fractures may occur through a cyst or in a weakened long bone. The most typical manifestation of PHPT is the cystic fibrous osteitis, which is characterized clinically by bone pain and radiographically by subperiosteal bone resorption in the distal third of the clavicles ,phalanges and radiological appearance of "salt and pepper" in the skull. Although bone cysts and brown tumors can occur anywhere, they mainly affect the ribs, humerus, and jaw [12]. Greater than 50% of patients with hyperparathyroidism have renal symptoms nephrolithiasis and nephrocalcinosis. Abdominal Recurrent acute pancreatitis, Muscle weakness, particularly in the proximal extremity muscles, together with progressive fatigue malaise, withCNS depression, nervousness,and cognitive dysfunction CVS Hypertension may occur in PHPT.This Nonspecific system signs and symptoms can mislead the physician and cause significant delay in diagnosis. As you see in the present case, he had complained from those symptoms for more than 1 year and one previous surgery without definite diagnosis during last six months. USG is one of the most common imaging methods used for neck evaluation and it is practically the fi rst option in the primary hyperparathyroidism assessment High Resolution Ultrasound Scan (HRUS) very diagnostic.On USG, parathyroid adenoma is seen typically as a round or oval homogenous, hypoechoic nodule localized behind the thyroid gland and at the lower aspect of paratracheal or paraeosophageal region. CT scan and MRI are more sensitive to assess ectopic sites. Preoperative parathyroid and thyroid imaging using technetium Tc 99m sestamibi scintigraphy-single-photon emission computed tomography (Tc 99m MIBI SPECT) and technetium Tc 99m sodium pertechnetate, suitable for minimally invasive radio-guided surgery [13, 14]. Parathyroidectomy is the treatment of choice in PHPT. In the last 10 to 15 years surgery of primary hyperparathyroidism (PHPT) moved from the wide bilateral neck exploration to minimally invasive approaches as the minimally invasive radioguided parathyroidectomy. Excised parathyroid gland has to be subjected to histopathological examination to confirm and differentiate adenoma, hyperplasia and malignancy.Sudden, post-operative hypocalcaemia may be a major complication of parathyroidectomy. The incidence of this'hungry bone syndrome' (i.e. sudden, unbalanced osteoblastic activity, lowering serum calcium levels) is likely to be higher in the developing world, due to associated deficiencies in pre-operative dietary calcium and vitamin D, than in the western world. This potential complication should be anticipated and aggressive nutritional support given. Our experience with fractures in primary hyperparathyroidism revealed that these take longer to heal and are prone to malunion unless immobilsedand Nonunion of fractures is rare. Bone histology returns to normal within 5-6 week. Brown tumours usually resolve with increase in bone density and sclerosis after parathyroidectomy. The extensive skeletal involvement due to hyperparathyroidism has rarely been reported. The substantial improvement in bone density, in promotion of fracture healing and in preventing pathological fractures after successful parathyroid adenectomy has been seen our case [15,16]. Our experience say every

patient should be investigated routine serum biochemical along with PTH, Vitamin D, Thyroid profile in Repeatedlymultiple fractures with progressively deformity for diagnosing Endocrine disorder as diagnosed in this case asHyperparathyroidism which was undiagnosed initially only treated as intertrochanteric fracture of hip which was failed and simultaneously develop other side hip pain with progressively deformity .this investigation also required for better prognosis of fracture union with assessing clinical symptoms.

This case is rare because both Hip was involvedfracture and patient was immobilsed for one year because remained undiagnosed.

Conclusion

Repeatedly multiple fractures must beinvestigated with appropriate and precise routine serum biochemicalalong with PTH Vitamin D Thyroid profile to diagnose endocrine disorder as in this case diagnosed as Hyperparathyroidism. With Clinical examination of neck as specific site with extremities ,deformities and systemic examination is essential . USG Neck as routine investigation for this disease. After parathyroidectomy and regular follow up is done. Afterwards fracture fixation is done as secondary procedure with adequate calcium and vitamin D correction give good prognosis better union with best possible outcome. This is rare presentation of PHPT with B/L Hip fracture and other fractures deformities is an excellent example of an endocrine disease that is best managed by a multidisciplinary approach and long term patient follow up.

Clinical Message

Repeatedly multiple fractures must be investigated with appropriate and precise method with detail clinical examination for diagnosing Endocrine disorder with very small cases as they present..Before proceeding for fracture it should be diagnosed causes of multiple fractures or implant failure as in this case diagnosed as Hyperparathyriodism. It is essential to correct prior metabolic disorder along with fracture fixation for better prognosis and good union.

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Conflict of Interest: NIL
Source of Support: NIL

How to Cite this Article

Chhawra S, Kambar AN, Gupta R. A Neglected Case of Bilateral Hip Fractures and Other Fractures with Delayed Diagnosis of Hyperparathyroidism as Cause. Trauma International May - Aug 2018;4(1):38-41.