Primary hyperparathyroidism (PHPT) is an uncommon condition in children and adolescents. However, rapid growth spurt during puberty may result in unmasking and development of certain skeletal manifestations of PHPT. We present three cases of PHPT associated with rare skeletal manifestations of rickets. All three patients had radiological evidence of rickets with primary hyperparathyroidism. All the three patients had single gland adenoma. Literature is sparse regarding reversal of features of rickets following parathyroidectomy. In all three patients of our series, there was a complete resolution of bone/joint pain. However, in two children only the genu valgum persisted but their growth was normal and they had no proximal muscle weakness. In another child multiple corrective surgeries were done to correct the deformities.

Keywords: Primary hyperparathyroidism, rickets, skeletal manifestations
concordant with the ultrasonography (USG) finding of a left superior parathyroid adenoma. Bilateral neck exploration was performed with intraoperative parathyroid hormone (IOPTH) monitoring. At operation, both right parathyroid glands were found to be grossly normal; a left superior parathyroid adenoma was found which was excised (2.2 × 1.6 × 1.6 mm, weight 2.2 gm). However, left inferior parathyroid gland could not be found. IOPTH according to Miami criteria was curative, as there was >50% drop from the pre-excision value of 2130 to 231 at 10 minutes. Histological examination was suggestive of parathyroid adenoma. At the last follow up, in 2016, her biochemical parameters including calcium and iPTH were within normal limits and she was free of bone pains, though her genu valgum persisted.

**Case 2**

A 15-year-aged young adolescent girl presented with history of pain in both legs since 3 years followed by stiffness in both knees with limping gait and dislocation of hip joint with deformities in both hands (widenings of the wrist) and chest (kyphosis and rachitic rosary) [Figures 5-9]. There was history of fracture left clavicle and left hand by trivial trauma. Since last 1 year she was bedridden. She consulted an orthopaedic when it was detected that she had high serum alkaline phosphatase levels and low vitamin D levels. She was put on vitamin D and calcium supplements but then she developed an episode of pancreatitis which required hospitalization, though she recovered on conservative treatment. She was evaluated during her admission for pancreatitis and serum calcium was found to be raised and hence was referred to our institute. She was found to have raised serum calcium along with high intact PTH of 1250 pg/ml. Her localization investigations were concordant, therefore, she underwent focused parathyroidectomy under cervical block. Postoperatively patient was given prophylactic calcium infusion because of the severe bone disease. Histopathology turned out to be parathyroid adenoma.

**Case 3**

A 11-year-old boy presented with difficulty in walking and bowing of knees since 10 months. Patient also had severe...
proximal muscle weakness with multiple bony swellings over face [Figure 10]. Patient was evaluated with magnetic resonance imaging of knee and given calcium and vitamin D supplements during his evaluation elsewhere with suspicion of rickets. However, when patient did not improve further

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**Figure 5**: Radiograph of pelvis AP view - demonstrates diffuse osteoporosis with coarse trabecular pattern. Coxavea deformity and pathological fractures of bilateral femoral neck

**Figure 6**: Radiograph of bilateral wrists - demonstrates irregularity and widening of growth plate with metaphyseal cupping and fraying of both radius and ulna

**Figure 7**: X-ray bilateral knee demonstrates generalised osteopenia, increased trabecular pattern, genu valgum deformity and increased zone of provisional calcification involving distal metaphyseal ends of bilateral femur

**Figure 8**: Widening of wrists

**Figure 9**: Chest deformity

**Figure 10**: Brown tumor of the mandible
investigations were done. On biochemical evaluation, serum calcium was high on three occasions and intact PTH was 607 pg/ml. Localization studies done revealed concordant findings on USG and MIBI. Patient underwent focused parathyroidectomy. Postoperatively patient developed clinical and biochemical hypocalcemia which was managed with calcium infusion and oral calcium and vitamin D. Histopathology came out to be parathyroid adenoma. Biopsy from bony swellings of face was osteitis fibrosa cystica (OFC).

**DISCUSSION**

Rickets as presenting feature of PHPT in children is extremely rare with only 23 cases reported till date. PHPT is typically a disease of post-menopausal women in Caucasian population. However, in areas of vitamin D deficiency and calcium deficiency, PHPT may be seen in young patients and even in children and adolescents.

Unlike the Western world, the classic presentation of PHPT in the form of OFC and nephrolithiasis is still commonly seen in developing countries and one of the reasons for this has been severe vitamin D deficiency in these regions.

In a review by Pitukcheewanont et al.

common presenting signs and symptoms of PHPT with rickets in children include knock-knees or genu valgum (70%), rachitic rosary (50%), widened wrist and ankle (31.5%), bone/joint pain (37.5%) and anorexia (31.25%). All three children in our report had radiological findings consistent with rickets. It can be speculated that the severe skeletal abnormalities seen in children may be due to synergistic effect of vitamin D deficiency along with deleterious effect of elevated PTH. A state of malabsorption leading to vitamin D deficiency is unlikely because there were no overt gastrointestinal symptoms in any of the patients.

Two radiological findings in rickets with PHPT require special mention. Genu valgum can also manifest in vitamin D deficiency states and these cases may be thought of having tertiary HPT, but all of the 23 cases mentioned in literature including ours had a single gland enlargement, whereas the classic picture of tertiary HPT is of parathyroid hyperplasia. Most of the cases of PHPT with genu valgum described so far have occurred in the period of adolescence when there is a rapid growth spurt combined with direct effect of elevated PTH on the growth plate.

Another finding is of slipped upper femoral epiphysis (SUFE). In the report by Pitukcheewanont et al. 13 children had hypercalcaemia and only three children were normocalcaemic at presentation but who became hypercalcaemic after vitamin D supplementation. In a review by Roizen and Levin it was reported that young PHPT has greater incidence of hypercalcaemia and hypercalciuria as compared to adult PHPT at similar levels of PTH. The highest recorded PTH was 1659 pg/ml while in one child the PTH level was still higher 1967 pg/ml.

In the report by Ganie et al. all four children had single adenoma; in the series by Pitukcheewanont et al. 13 out of 14 children had adenoma and only one child had multiglandular disease. In the case reported by Dutta et al. their patient also had a single adenoma. In all our three children there was a single adenoma. The PHPT in developing countries is also associated with large and heavy adenomas with a higher mean weight.

Literature is sparse regarding reversal of features of rickets following parathyroidectomy. In all three patients of our series, there was complete resolution of bone/joint pain. However, in two children only the genu valgum persisted but their growth was normal and they had no proximal muscle weakness. In another child multiple corrective surgeries were done to correct the deformities.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

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