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The role of the orthodontist in diagnosis of hyperparathyroidism : rare case with general root resorption

Bong-Kuen Cha[1] · Suk-Kuen Lee[2]

Excessive production of parathyroid hormone causes bony disorder such as periosteal bone resorption and bone pain due to excessive skeletal demineralization. A Class III facial deformity case with generalized root resorption presented here was turned out to be due to hyperparathyroidism. Clinical and cephalometric analysis revealed a straight skeletal profile with a retruded maxilla and a prognathic mandible. The x-ray findings demonstrated generalized root resorption of entire dentition to different degree. There also appeared osteoporosis like immature trabecular structure with the evidence of ground glass appearance. Serum test showed elevated level of parathyroid hormone and growth hormone. Change of cranial growth by hyperparathyroidism can be dependent upon a decreased bone apposition in viscerocranial growth site and abnormalities in cranial suture growth. It is possible to hypothesize that growth retardation of maxilla at least partially be accounted for hyperparathyroidism. Therefore, regarding to the definite etiology of skeletal Class III and orthodontic treatment planning considering root resorption and osteoporosis, the early diagnosis for the hyperparathyroidism should be carefully carried by clinical and laboratory studies.

Key words : Class III malocclusion, hyperparathyroidism, osteoporosis, root resorption

Hyperparathyrodism is an uncommon disease and the presenting symptoms may occur in the jaws with generalized osteoporosis manifesting as loss of lamina dura around the teeth or production of osteolytic areas known as 'Brown tumor'.[1-12] But disease process accompanying the generalized root resorption and malocclusion is extremely rare. A Class III facial deformity case with generalized root resorption presented here was turned out to be due to hyperparathyroidism. This study is going to analyze and describe the deformity of craniofacial structure and diagnostic characteristics of this rare case.

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CASE

In August 1998, 16-year-old female was referred to Department of Orthodontics at Kangnung National University Dental Hospital because of generalized teeth mobility, and making counsel for the possibility of surgicoorthodontic treatment of skeletal Class III malocclusion by general dentist. Previously she was examined in other hospital for consultation of her general growth

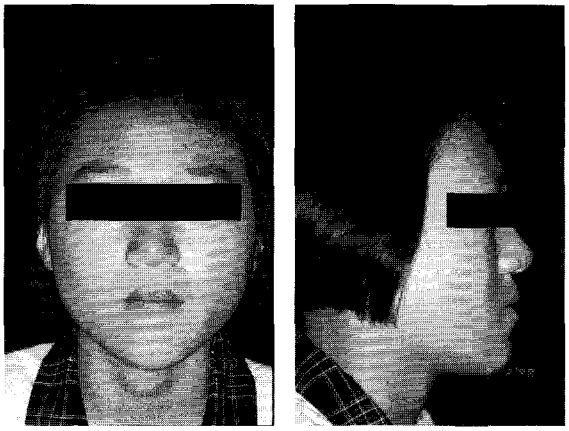


Fig. 1. Extraoral photos

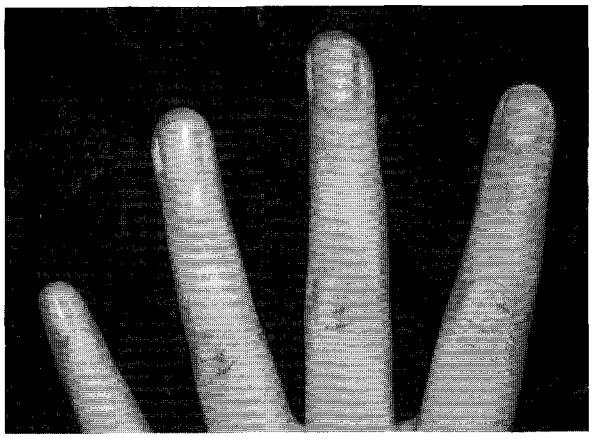


Fig. 2. Photo of fingers

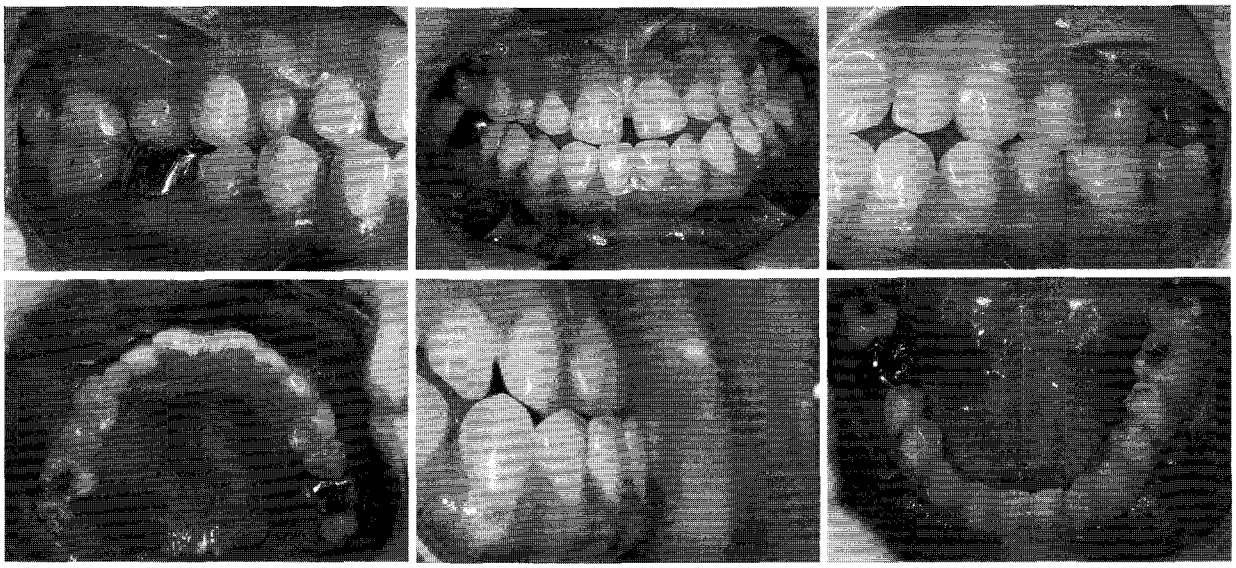


Fig. 3. Intraoral photos

2 years ago.

The patient has begun her menstruation since summer, 1997. The patient had a slightly asymmetrical face with a protrusive mandible and hypoplastic maxilla and showed a straight profile(Fig. 1). Her finger nail was thin and fragile(Fig. 2).

Intraoral inspection revealed that the size of her teeth was relative small and especially upper and lower third molars are very tiny. In upper dentition, there was a large anterior diastema with heavy labial frenum and a retained upper right primary canine. Lower dentition was well aligned and severely dental compensated. The mandibular dental midline was shifted 2mm to the left of the maxillary dental midline. The patient had a Class III malocclusion with a reverse overjet of 4mm, an overbite of 0mm and posterior crossbite bilaterally (Fig. 3).

The panoramic and standard radiographs of mandible and maxilla demonstrated impacted right upper canine and generalized root resorption of entire dentition with different degree. There also appeared immature trabecular structure with evidence of ground glass appearance or osteoporotic sign with minute disappearance of the lamina dura around the teeth (Fig. 4, 5).

Hand-wrist analysis by Hägg revealed that her ske

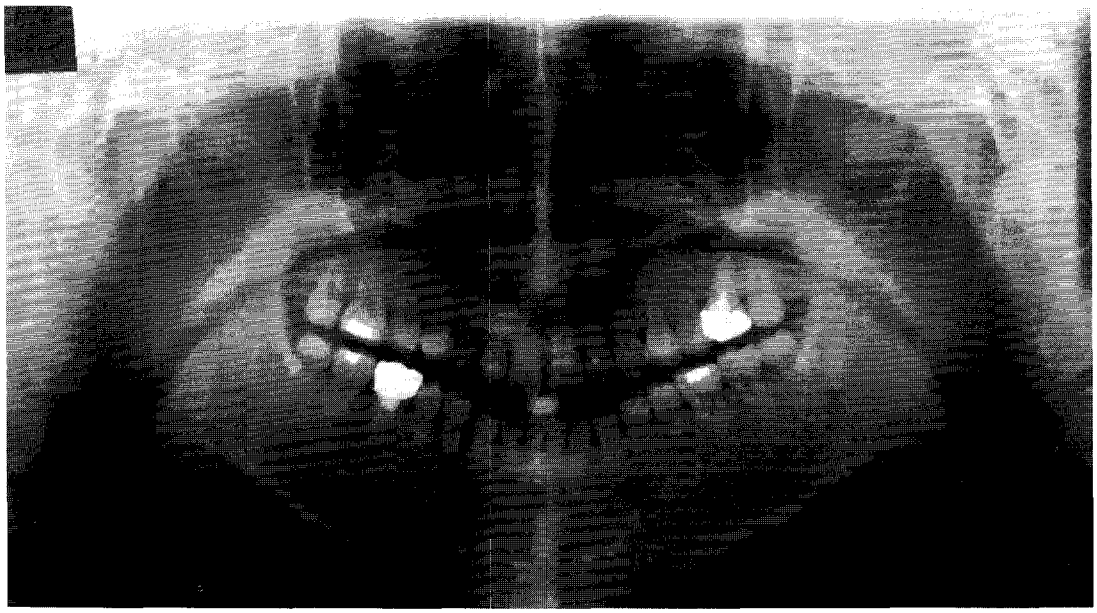


Fig. 4. Panoramic radiograph

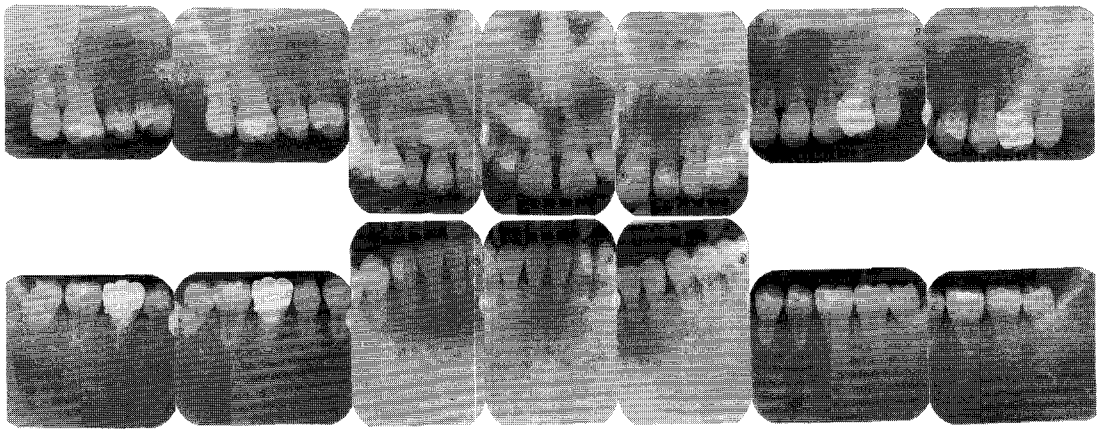


Fig. 5. Full mouth standard radiographs

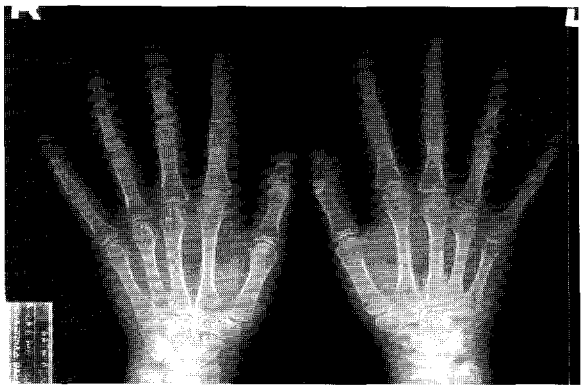
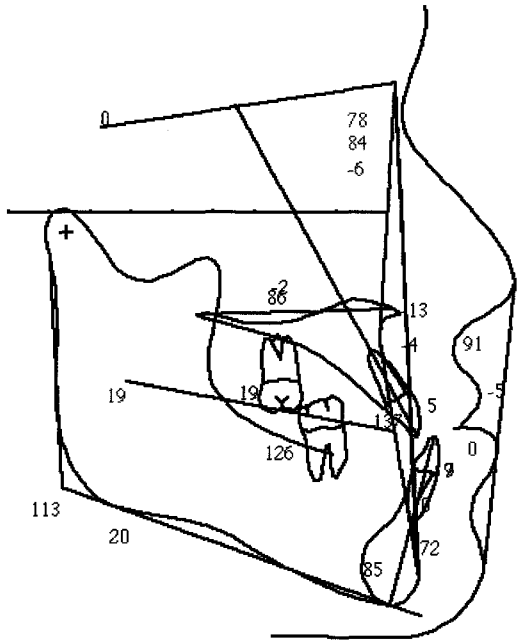


Fig. 6. Hand-wrist radiograph

letal maturation stage was MP3-I stage that fusion of the epiphysis and metaphysis had begun and R-IJ stage that fusion of the distal epiphysis of the radius had almost completed but there was a small gap at one or both margins. There was found slight immature trabecular structure in the middle phalanx (Fig. 6).

Cephalometric analysis revealed a straight skeletal profile with a retruded maxilla and a prognathic mandible (SNA 77.9°, SNB 83.8°, Wits appraisal - 6.0mm). The lower incisors were lingually compensated (IMPA



	A	Normal	Clin. Dev.	
SNA (dg)	77.9	82.0	-1.4	*
SNB (dg)	83.8	80.0	1.3	*
ANB (dg)	-6.0	2.0	-4.0	***
Wits Appraisal (mm)	-16.0	1.1	-9.0	***
Mandibular Plane Angle (dg)	19.7	23.8	-0.9	
Mx. 1 – APo (mm)	5.3	6.2	-0.4	
IMPA (dg)	84.7	90.0	-0.9	

Fig. 7. Cephalometric analysis

Table 1. Serum test

Test Item	*Result*	*Reference*
Alkaline phosphatase	256 IU/l	70-250 IU/l
Growth Hormone	16.05	M : 0.50-3.83 ng/mL F : 0.10-7.02 ng/mL
ACTH	41.18	12-76 pg/mL
Calcitonin	Below 5.0	Below 30 pg/mL
25-OH-Vit. D	14.14	9.9-41.5 ng/mL
PTH-intact	84.24	9-55 pg/mL

84.7°). The mandibular plane angle (19.7°) and gonial angle (112°) were both below normal, indicating a brachyfacial tendency (Fig.7).

Serum level of parathyroid and growth hormone were remarkably elevated, 84.24pg/ml(normal range ; 9 ~55pg/ml) and 16.05ng/ml(normal range ; 0.5~3.83ng/ml), while those of ACTH, calcitonin and 25-OH-vitamin D were within normal limit. Alkaline phosphatase level was also somewhat elevated (Table 1).

DISCUSSION

Hyperparathyroidism is a relative rare disease predominantly affecting the middle-aged, which is said to be three times as common in woman as in man.[13]

There are two sources of calcium-diet and skeletal system. When the calcium demand surpasses the dietary intake, calcium is then removed from the bone. This hormone is responsible for maintaining serum calcium-phosphorous homeostasis. In general, the metabolic disturbance that produce retention of phosphate or depletion of calcium will cause increased metabolic activity of the parathyroid gland.[3,5,7,14-17]

Hyperparathyroidism patient may have decreased bone density as well as bone remodeling changes consistent with elevated serum alkaline phosphatase level.[1,14] The presenting symptom may occur in the jaws, with generalized osteoporosis manifesting as loss of lamina dura around the teeth on radiography[1] and production of osteolytic area known as 'Brown tumor'.[3,13,18,19] This patient showed elevated serum level of parathyroid and growth hormone and alkaline phosphatase level (Table 1). Although we couldn't differentiate between primary and secondary type for this

patient, such a laboratory results combined with the radiographic signs as osteoporotic trabecular pattern and ground glass appearance of bone confirmed the diagnosis of hyperparathyroidism.

This patient had considerable tooth mobility of entire dentition due to mainly shortened root. Root resorption by systemic osteolytic disease is not so frequently because tooth doesn't function generally as the reservoir for calcium and phosphate.[17]

Central giant-cell granuloma or cystic bone lesion by hyperparathyroidism, oxalose, Morbus Gaucher and eosinophilic granuloma belong to such a rare systemic disease that could lead to entire root resorption but not always.[19] According to Becks,[20] other possible systemic endocrine problems including hypothyroidism, hypopituitarism, and hyperpituitarism could be related to root resorption. But in that extensive root resorption doesn't occur on all hyperparathyroidism case, some question arising out of this case is not fully answered. In literatures, there are not such a case yet frequently described. Hyperparathyroidism or Paget disease have linked to root resorption in a few anecdotal case reports.[21,22] But recent controlled animal study did not support the hypothesis that hyperparathyroidism is primarily responsible for the increase of root resorption.[9]

As a metabolic disease, oxalose can lead to overproduction of calcium oxalate crystal in tissue and this can give rise to renal failure and secondary type of hyperparathyroidism. A case reported by Bunte et al,[19] in which oxalose lead to hyperparathyroidism with general dentomaxillary destruction, could give a preview of this unique case but not full answer to this problem. Definite etiology of root resorption for this female patient should be a future study assignment. Despite of reports showing no correlation between gender and root resorption,[23,24] some studies suggested female was more susceptible to root resorption and idiopathic root resorption ratio was 3.7 : 1, female to male respectively.[25]

Despite many studies have focused on bone remodeling as a necessary component for tooth movement by orthodontic force, electric current, or circulating monocyte, little has been done to show that systemic effects on bone metabolism can change the rate of tooth movement. The study using beagles fed with a hyperparathyroidism diet, indicated that the hyperparathyroidism animal had significantly decreased bone density and osteoporotic trabecular loss and revealed more rapid tooth movement under orthodontic force.[26] Furthermore the overall regulation of bone turnover and thus local remodeling activity by orthodontic tooth movement can be partially determined by factors controlling the calcium homeostasis such as parathyroid hormone and vitamin D, and in the hypocalcemic situation the increase of root resorption was strongly related to enhanced alveolar bone resorption.[9] So the orthodontist should be careful for treating patient with hyperparathyroidism not only because of shortened root but also possible increased rate of teeth moving and possible alveolar bone resorption.

Lamina dura can be defined as the internal septum that normally presents a thin radiopaque border adjacent to the periodontal ligament and the crest. Changes representing pathosis result in either partial or total loss of the lamina dura. Partial loss or discontinuity around one or more teeth is usually caused by local disorder but sometimes seen in systemic disease like Gaucher, leukemia, and scleroderma or hyperparathyroidism.[1] In this patient the definite loss of lamina dura was not happened in all teeth. Diminutive loss was difficult to discern but by careful reading some discontinuity was found (Fig. 5).

Secondary hyperparathyroidism associated with renal failure often causes osteitis fibrosa resulting in deformity, which is especially prominent in the spine, rib, long bone, and skull. The cortical plate of bone can be expanded in the cystic formation and giant-cell tumors but rarely is the entire maxilla or mandible.[3,18,19] In the case described, the symptom assumed to involve the deformity and malocclusion could be caused possibly by enlargement of mandible. But it is difficult to differentiate the etiology of Class III malocclusion between genetic origin and the pathological enlargement of jaw as usually occurred in hyperparathyroidism patient and furthermore in this patient no sign of osteitis fibrosa was observed.

Aberration of bone turn-over are known to cause

changes in the skull morphology.[8,27-33] Recent studies in vitro have shown that when bone resorption is stimulated by parathyroid hormone, mitogenic factors for bone cells are released from the matrix.[8] Critical roles of parathyroid hormone-related protein (PTHrP) and the PTH/PTHrP receptor in the regulation of endochondral bone formation are well documented.[28,31]

It was found in animal experimental studies by rats that a change of cranial growth pattern induced by hypocalcemia and secondary hyperparathyroidism was dependent upon a decrease of bone apposition in cranial suture growth sites and this alteration of osteogenesis was only induced locally in growth sites determining the morphology of viscerocranium and its relation to the neurocranium.[8]

The cephalometric analysis of this patient indicated a Class III skeletal tendency mainly due to underdevelopment of maxilla and a little overgrowth of mandible. It was possible to speculate that growth retardation of maxilla at least partially be accounted for by hyperparathyroidism.

In addition, plasma growth hormone(GH) levels of this patient remained elevated. It was hypothesized that the elevation of serum PTH level was induced either by the rise of phosphorus levels induced by GH.[33] The fact that mandible prognathism could be caused by growth hormone-relating hormone as acromegaly implied the another possible explanation about the etiology of this type of malocclusion.

However, critical evidence does not yet support this concepts and pathophysiological influence of this hormone on growth modification activity remains to be clarified.

SUMMARY

A case of facial deformity and malocclusion associated with hyperparathyroidism was presented. In a 16 year-old female patient, extensive resorption of entire dentition and retrusion of maxilla was observed which has not often been described.

The physiology of hyperparathyroidism should be of interest to general practitioners and orthodontists because of the fact that the first symptom in this case involves the oral cavity and consequently has a tremendous influence on treatment planning.

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국문초록

부갑상선 기능항진증 진단에서의 교정의사의 역할 : 치근흡수를 동반한 희귀증례

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부갑상선 호르몬이 과도하게 분비되는 경우 전반적인 골의 탈석회화에 의해 골막성 골흡수 및 골의 동통 등의 증상이 발현될 수 있다. 본고에서 소개된 환자의 경우 광범위한 치근 흡수를 동반한 부정교합 환자로, 임상 및 두부 방사선 계측사진 분석에서 상악 열성장과 하악 과성장이 동반된 골격성 III급 부정교합으로 진단되었으며, 병리검사결과 부갑상선 호르몬 및 성장 호르몬의 수치가 증가된 것으로 보아 부갑상선 기능항진증으로 판단되었다. 방사선 사진 분석 결과 광범위한 치근의 흡수를 관찰할 수 있었으며 골다공증과 유사하게 골소주 형태가 매우 성기면서 ground glass 양상을 보였다. 이와 같이 부갑상선 기능항진증에 의해 골질뿐만 아니라 치근이 영향을 받은 것은 매우 드문 경우로 생각된다. 또한 부갑상선 기능항진증에 의해 내장두개의 골침착이 감소될 수 있다는 동물실험 결과를 볼 때 III급 부정교합의 원인이 상악 열성장인 점과 연관하여 매우 흥미로운 점으로 지적된다. 따라서 부갑상선 기능항진증의 다양한 증상 및 양태에 대한 조기진단 및 이에 대한 지식은 교정진단 및 치료에 중요한 역할을 할 수 있다고 생각된다.

주요 단어 : III급 부정교합,부갑상선 기능항진증, 골다공증, 치근흡수