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Case report

Hyperostosis around the bilateral acetabulum associated with hypoparathyroidism: A case report

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【Abstract】

We report the case of a 57-year-old woman with hyperostosis around the bilateral acetabulum associated with untreated secondary hypoparathyroidism. She presented with gait disturbance and inability to walk. Radiographs showed abnormal ossification around her hips. We resected the ossifications to improve joint function. One year after surgery, radiographs show no recurrence of ossification. When radiographs show excessive hyperostosis, it is important to exclude the presence of metabolic bone disease.

[Introduction]

Hypoparathyroidism occurs when the parathyroid glands are unable to maintain calcium homeostasis. Known features of hypoparathyroidism include soft tissue ectopic calcifications and skeletal abnormalities [1].

In this case report, our patient developed hyperostosis around the bilateral acetabulum due to untreated secondary hypoparathyroidism, which in turn caused a gait disturbance that dramatically improved following resection of the ossifications.

Our patient gave informed consent for this report, and the Kanazawa University hospital investigational review board approved the research protocol.

[Case Report]

A 57-year-old woman presented to our hospital with chief complaints of limited range of motion of bilateral hip, gait disturbance, and bilateral leg cramps. She had noticed limited range of motion of bilateral hip since 40 years of age, but had not sought medical assistance because it did not cause pain. Over time, however, the range of motion of her hip had become progressively more limited, and she then become aware of a gait disturbance.

Her medical history included strumectomy at 17 years of age, laminoplasty for cervical ossification of the posterior longitudinal ligament (OPLL) at 49 years of age, and hormone replacement therapy for hypothyroidism starting at 55 years of age.

On physical examination, the range of motion of both hips was severely limited (right hip: flex 40°, ext -10°, abd 10°, add 10°, external rotation 5°, internal rotation 0°; left hip: flex 50°, ext -5°, abd 10°, add 10°, external rotation 5°, internal rotation 0°). The patient was walking with bilateral T canes and could not put on her socks or clip her toenails by herself.

Pelvic radiography showed abnormal ossification extending along both hip joint capsules (Fig. 1). Computed tomography showed bony sclerosis around both hip joints with preservation of joint spaces; osteoarthritic changes were not seen (Fig.2).

Blood testing revealed very low serum levels of calcium (5.7 mg/dl : 8.0-10.5 mg/dl) and elevated serum phosphate (4.8 mg/dl : 2.5-4.5 mg/dl). Additional hormonal examinations revealed that despite the low calcium level, parathyroid hormone was at the lower limits of normal (16.1 pg/ml : 10.3-65.9 pg/ml). On the basis of these findings, the endocrinologist made a diagnosis of hypoparathyroidism. Additional testing failed to detect any areas of abnormal ossification other than both hip joints.

Following diagnosis, the patient was started on alfacalcidol, an analogue of vitamin D, which quickly improved her leg cramps. Based on the clinical history, blood examination data, and results of medication, we concluded that the hyperostosis around her hips was associated with hypoparathyroidism. We decided to perform a two-stage resection of the ossification areas to improve joint function: right side first and then the left side.

Postoperatively, the patient's gait disturbance improved such that she can now walk without needing a T-cane. The range of motion of the bilateral hips has dramatically improved (right hip: flex 90°, ext 0°, abd 15°, add 10°, external rotation 30°, internal rotation 0°; left hip: flex 100°, ext 0°, abd 25°, add 10°, external rotation 30°, internal rotation 5°). Radiographic examinations one year after surgery showed remodeling of the excised ossification edges and no recurrent ossification around the both hips. The vitamin D replacement therapy is being continued.

[Discussion]

Hypoparathyroidism is caused by either decreased secretion of parathyroid hormone or decreased responsiveness to parathyroid hormone, leading in turn to hypocalcemia or hyperphosphatemia. Our patient was diagnosed with hypoparathyroidism secondary to her thyroidectomy at age 17. Because hypoparathyroidism is most commonly seen as a complication of surgery, specifically thyroidectomy, parathyroidectomy, or radical

neck surgery [2], taking a detailed medical history is important to arriving at a correct diagnosis in cases of suspected hypoparathyroidism.

Hypoparathyroidism can exhibit diverse signs related to hypocalcaemia, including neuromuscular symptoms (*e.g.* muscle cramps, convulsions or tetanic contractions), circumoral and/or acral numbness, and paresthesias, [3,4]. Hypoparathyroidism sometimes causes ossification of spinal ligaments, but the frequency of bony lesions due to hypoparathyroidism is relatively rare in comparison with hyperthyroidism generally. Hyperostosis around the hips and shoulders is one of the calcium metabolism-related lesions of hypoparathyroidism [1]. Hypoparathyroidism also has been associated with extraosseous calcification, such as basal ganglia calcification and cataract [4]. However, imaging findings of hyperostosis due to hypoparathyroidism closely resemble imaging findings of several other disease entities, such as diffuse idiopathic systemic hyperostosis (DISH) or ankylosing spondylitis (AS) [5-7]. One published case report involves a patient with idiopathic hypoparathyroidism who had been misdiagnosed as having AS and thus was undergoing immune-modulating treatment before being correctly diagnosed [7]. Our patient had undergone laminoplasty for cervical OPLL when she was age 49, but her hypoparathyroidism remained undiagnosed and untreated for a long time. These reports highlight the importance of excluding metabolic bone disease when radiography shows excessive hyperostosis.

To our knowledge, few reports have made reference to resecting ossifications due to hypoparathyroidism, and long-term results are thus unknown. One case report described the resection of a bilateral osseous bridge across the acetabulum due to entrapment [8]. One year after surgery, plain radiographic examination showed no evidence of recurrence of our patient's ossified lesions, and she has maintained a good range of motion in her hip. Ongoing treatment of her hypoparathyroidism with vitamin D appears to be helping prevent recurrence. Nonetheless, careful follow up is essential in patients with hypoparathyroidism.

Conflict of interest statement None.

References

- 1) Steinberg H, Waldron BR. Idiopathic hypoparathyroidism: analysis of 52 cases, including report of new case. *Medicine* 1952 ; 31 : 133-38
- 2) Harris VW, De Beur SJ. Postoperative hypoparathyroidism : medical and surgical therapeutic options. *THYROID* 2009 ; 19 : 967-73
- 3) Marx SJ. Hyperparathyroid and hypoparathyroid disorders. *N Engl J Med* 2000 ; 343 :1863-75
- 4) Shoback D. Clinical practice. Hypoparathyroidism. *N Engl J Med* 2008 ; 359 :391-403
- 5) Goswami R, Ray D, Sharmat R, Tomar N, Gupta R, Gupta N, et al. Presence of spondyloarthropathy and its clinical profile in patients with hypoparathyroidism. *Clinical Endocrinology*. 2008 ; 68 : 258-63.
- 6) Lambert RGW, Becker EJ. Diffuse skeletal hyperostosis in idiopathic hypoparathyroidism. *Clinical Radiology*. 1989 ; 40 : 212-15
- 7) Korkmaz C, Yasar S, Binboga A. Hypoparathyroidism simulating ankylosing spondylitis. *Joint Bone Spine*. 2005 ; 72 : 89-91
- 8) Tsuji S, Tomita T, Inaoka M, Higashiyama M. Psoriatic erythroderma with bilateral osseous bridge across the acetabulum. *Clin Orthop Relate Res*. 2010 ; 468 : 1173-77

Figure legend

Fig.1

Bilateral ossification around the acetabulum was observed on plain radiography.

Fig.2

Computed tomography showed severe ossification extending around the bilateral acetabulum.



