Pathologic Fractures, Anemia, Hypercalcemia and Hypocalciuria: an Association between Celiac Disease and Hyperparathyroidism

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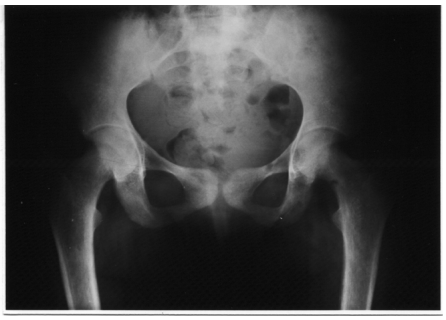
Hyperparathyroidism associated with celiac disease is extremely rare. We report on such an association and its diagnostic complications.

Case Description

A 31-year-old woman of Jewish-Yemenite origin was admitted because of left hip pain that had persisted for 3 months, associated with severe anemia. The patient denied fever, weight loss, bloating, abdominal pain, diarrhea, blood loss, or muskuloskeletal pain in other sites. On physical examination she looked extremely thin and pale. The only pertinent finding was severe pain evoked by left thigh abduction.

Laboratory investigation revealed severe iron deficiency anemia (hemoglobin 56 g/L and ferritin 2.2 μg/L), hypercalcemia (2.75 mmol/L), hypophosphatemia (0.58 mmol/L), high alkaline phosphatase (706 U/L), metabolic acidosis (bicarbonate 12 mmol/L), hypocalciuria (7 mmol/L) and hypophosphaturia (150 mmol/L). Radiological examination of the pelvis [Figure] revealed two radiolucent foci, one in the left femoral neck, presenting as an incomplete fracture, and the other in the pubis. At this point, serum parathyroid hormone was very high (48.8 pmol/L, normal values 1.0-7.6 pmol/L). A further evaluation of the iron deficiency anemia revealed normal radiological studies of the gastrointestinal tract. A small bowel biopsy demonstrated duodenal mucosa with subtotal villous atrophy and moderate lymphoplasmacytic infiltrate in the lamina propria. The anti-endomysial antibodies titer was highly positive. The patient was put on a gluten-free diet. After a parathyroid ^{99m}Tc MIBI scan that had demonstrated an increased uptake in the area of the left thyroid lobe, a 1.1 g parathyroid adenoma was resected.



Radiological anteroposterior view of the pelvis, showing two radiolucent foci — one in the left femoral neck, presenting as an incomplete fracture, and the other in the pubis.

Six weeks after the operation the patient gained weight and could walk easily. Marked radiological improvement was seen on repeated X-ray examination of the pelvis. Repeated laboratory results showed a correction of the anemia and the iron deficiency state. The serum PTH, calcium, phosphorus, chloride and bicarbonate returned to normal range, as did the urinary calcium excretion. A recent examination, 2 years after surgery, revealed the patient to be clinically healthy, after a normal pregnancy and birth.

PTH = parathyroid hormone

Comment

A young patient with imminent pathological fracture associated with hypercalcemia, hypocalciuria and iron deficiency anemia is described. We suggest that this unique presentation was the result of the co-occurrence of occult celiac disease, manifested as isolated iron deficiency anemia (and a probable vitamin D malabsorption), and hyperparathyroidism induced by a parathyroid adenoma. The diagnosis of celiac disease, which had a very unusual presentation in this case, was established by the combination of iron deficiency anemia, typical findings in a small bowel biopsy, the presence of high titer anti-endomysial antibodies, and the excellent stable response to the gluten-free diet.

The possible association of celiac disease and hyperparathyroidism has not been emphasized, and only two previous cases coexistent with celiac disease have been reported to date [1,2]. A distinct feature of the present case was the severe metabolic bone disease, evidenced by the X-ray studies and associated with high levels of PTH, but only mild hypercalcemia and hypocalciuria. Primary hyperparathyroidism, in contrast, usually

presents with hypercalcemia and hypercalciuria, although calcium clearance is reduced [3].

We therefore suggest that a combination of hyperparathyroidism and vitamin D deficiency, caused by the celiac disease, induced this unique metabolic state. Whether the celiac disease was the primary event, inducing secondary and eventually "tertiary hyperparathyroidism," as previously described [1,2], remains an interesting speculation.

References

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