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Case Report

Premenopausal Osteoporosis in A Patient with Autoimmune Polyglandular Syndrome: A Case Report

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Abstract

Osteoporosis is a skeletal disease characterized by low bone mass associated with decreased bone strength and increased risk of fractures. Low bone mass in premenopausal women is less common than in postmenopausal women, and bone loss in premenopausal women is usually due to secondary causes such as estrogen deficiency, glucocorticoid exposure, malabsorption, thyroid disorders, and hyperparathyroidism. In women with premenopausal osteoporosis, treatment should be planned according to the underlying secondary causes. In this case report, the importance of investigating the secondary causes leading to premenopausal osteoporosis and its treatment are discussed.

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Introduction

Osteoporosis is a skeletal disease characterized by low bone mass associated with decreased bone strength and increased risk of fractures.[1] Low bone mass may be associated with insufficient peak bone mass acquisition and/or ongoing bone loss. Peak bone mass which is acquired 90% of its total adult value by the age of eighteen, occurs totally around the age of 30.[2] Most of peak bone mass formation is determined by family history, gender and race while nutrition and exercise are responsible for 25% of peak bone mass formation. Physiological bone loss that starts after the age of 35 in women accelerates with menopause. Osteoporosis is most commonly seen in postmenopausal women. Low bone mass is less common in young premenopausal women.[3,4] Bone loss in the premenopausal period may be due to secondary causes such as estrogen deficiency, glucocorticoid exposure, malabsorption, thyroid disorders, and hyperparathyroidism, as well as idiopathic factors. Bone mineral densitometry measurements alone should not be used to define premenopausal osteoporosis. Premenopausal osteoporosis in a young woman can be defined as low bone mineral density (BMD) for age (Z score ≤-2.0) together with the presence of fractures in

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Table 1. Secondary causes of osteoporosis in premenopausal women

Anorexia nervosa
Gastrointestinal malabsorption (celiac disease, postoperative states)
Diabetes mellitus
Vitamin D and/or calcium deficiency
Hyperthyroidism
Hyperparathyroidism
Cushing's syndrome
Hypogonadism
Hypercalciuria
Rheumatoid arthritis and other inflammatory conditions
Alcoholism
Renal disease
Liver disease
Homocystinuria
Hereditary hemochromatosis
Hematologic disorders (systemic mastocytosis, Gaucher disease, thalassemia major)
Medications (glucocorticoids, immunosuppressants,antiseizure medications [phenobarbital and phenytoin], GnRH agonists, heparin, chemotherapeutics, thiazolidinediones, depot medroxyprogesterone acetate, excess thyroid hormone, selective serotonin reuptake inhibitors, proton pump inhibitors

bones or presence of any risk factor for secondary osteoporosis (Table 1).

Case Report

Our case was a 21-year-old female patient who was diagnosed with autoimmune polyglandular syndrome due to presence of type 1 diabetes mellitus, autoimmune thyroiditis, autoimmune primary adrenal insufficiency, Sjögren's disease, fibromyalgia, primary biliary cholangitis and autoimmune hepatitis. After long-term steroid use, she applied to another clinic with the complaint of diffuse bone pain and was diagnosed with osteoporosis after clinical evaluation and bone mineral densitometric measurements. The patient was then admitted to our endocrinology outpatient clinic with complaints of recurrent bone pain and amenorrhea. In the current bone mineral density measurement, total Z score, T score and BMD were -3.1, -3.1, and 0.620 g/cm^2 for femur and -2.2, -2.5, and 0.892 g/cm^2 for L1-L4, respectively. The patient was scheduled for further diagnostic tests for secondary causes of premenopausal osteoporosis. Blood chemistry showed a calcium level of 8.2 mg/dL [normal range (NR): 8.4-10.2] with a phosphorus level of 2.9 mg/dL (NR: 2.4-4.4). 25-OH vitamin D level was 13.3 µg/L (NR: 20-50), while parathormone level was 186 ng/L (NR: 15-68.3) and ALP level was 88 U/L (NR: 40-150). Laboratory results for sex hormones included a FSH level of 0.73 IU/L (NR: 1.38- 5.47) with a LH level of 0.21 IU/L (NR: 0.56-14.0), an estrogen level of 24 ng/L (NR: 21- 312) and a progesterone level of 1 ng/mL (NR: 1- 4.5). Pertinent blood work for thyroid function resulted with normal TSH 2.17 mU/L (NR: 0.35-4.94) and free T4 levels 1.08 ng/dL (NR: 0.7-1.48) under replacement treatment. Further tests revealed high blood glucose level 133 mg/dL (NR: 70-100) and a HbA1c level of 7.5% (NR: 4-6%). AST level was 14 U/L (NR: 11-25), while ALT level was 17 U/L (NR: 7-28) and creatinine level was 0.67 mg/dL (NR: 0.56-0.85). Anti-tissue transglutaminase IgA level was detected to be 0.7 U/mL (NR: 0-10).

The patient was hospitalized and an esophagogastroduodenoscopy (EGD) was performed because she complained of nausea that did not regress with antiemetic therapy. Grade A esophagitis, laxity in the lower esophageal sphincter, food residues in the stomach and scalloped duodenal folds were detected during EGD. Gastroparesis secondary to diabetes mellitus was suspected in the patient who had food residues in the stomach during endoscopy after 12 hours of fasting. A biopsy was taken from the scalloped duodenal folds with a prediagnosis of celiac

disease, however no pathological finding other than chronic mucosal inflammation was reported. The patient was diagnosed with autoimmune polyglandular insufficiency. Combined estrogen and progesterone treatment was initiated for her amenorrhea. Vitamin D and calcium replacement therapy was given for vitamin D deficiency and secondary hyperparathyroidism. The dosage of steroid treatment for her autoimmune hepatitis and adrenal insufficiency was rearranged. Bisphosphonate treatment was not planned because she was in reproductive age. Thyroid hormone and insulin replacement therapies were continued.

Discussion

The diagnosis and treatment approaches for osteoporosis differ in premenopausal and postmenopausal women. The relationship between BMD and fracture risk in premenopausal women is not correlated, and the prevalence of fracture is much lower than in postmenopausal women. Since the relationship between bone mass and fracture risk in premenopausal women is not the same as postmenopausal women, guidelines for the diagnosis and treatment of postmenopausal osteoporosis do not apply to women diagnosed with premenopausal osteoporosis. In women diagnosed with premenopausal osteoporosis, treatment should primarily address the underlying cause. Lifestyle changes as well as a pharmacological treatment with calcium (1000 mg/day) and cholecalciferol (800-1500 IU/day) are recommended in premenopausal osteoporosis. Lifestyle modification includes regular load-bearing exercises (such as walking), cessation of smoking and alcohol consumption and limiting caffeine intake. Estrogen replacement has beneficial effects on bone mass in women with premenopausal osteoporosis secondary to hypogonadism.[5] Although there are publications showing that bisphosphonates are beneficial in women diagnosed with osteoporosis secondary to glucocorticoid usage, their use is not recommended in childbearing age women.[6] As in our case, the treatment plan in premenopausal osteoporosis should be tailored according to the patient, and if there is an underlying secondary cause, the treatment should be directed primarily to this etiology.

Conflict of Interests

Authors declare that there are none.

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References

1. Kanis JA, Melton LJ 3rd, Christiansen C, Johnston CC, Khaltaev N. The diagnosis of osteoporosis. J Bone Miner Res. 1994 Aug;9(8):1137-41. doi: 10.1002/jbmr.5650090802.
2. Baxter-Jones AD, Faulkner RA, Forwood MR, Mirwald RL, Bailey DA. Bone mineral accrual from 8 to 30 years of age: an estimation of peak bone mass. J Bone Miner Res 2011 Aug;26(8):1729-39. doi: 10.1002/jbmr.412.
3. Lauder TD, Dixit S, Pezzin LE, Williams MV, Campbell CS, Davis GD. The relation between stress fractures and bone mineral density: evidence from active-duty Army women. Arch Phys Med Rehabil. 2000 Jan;81(1):73-9. doi: 10.1016/s0003-9993(00)90225-9.
4. Jamal SA, Ridout R, Chase C, Fielding L, Rubin LA, Hawker GA. Bone mineral density testing and osteoporosis education improve lifestyle behaviors in premenopausal women: a prospective study. J Bone Miner Res. 1999 Dec;14(12):2143-9. doi: 10.1359/jbmr.1999.14.12.2143.
5. Liu SL, Lebrun CM. Effect of oral contraceptives and hormone replacement therapy on bone mineral density in premenopausal and perimenopausal women: a systematic review. Br J Sports Med. 2006 Jan;40(1):11-24. doi: 10.1136/bjsm.2005.020065.
6. McNicholl DM, Heaney LG. The safety of bisphosphonate use in pre-menopausal women on corticosteroids. Curr Drug Saf. 2010 Apr;5(2):182-7. doi: 10.2174/157488610790936178.