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Early Spondyloarthritis Presenting as Premenopausal Osteoporosis and Multiple Vertebral Fractures: a Case Report

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Abstract

We report a 30-year-old woman who presented during pregnancy with mechanical type backache due to non-traumatic multiple vertebral fractures. In depth assessment revealed multiple risk factors for osteoporosis, including adolescent steroid use for asthma, prolonged breastfeeding and vitamin D deficiency. She also had persistently elevated inflammatory markers with radiological evidence of early inflammatory sacro-ileitis. The rheumatoid factor and HLA B27 were negative. She was successfully managed with NSAIDS, zolindronic acid and calcium supplementation. This is an example of early inflammatory arthritis in the background of multiple risk factors causing osteoporosis severe enough to cause fragility fractures. This signifies the importance of screening for multiple risk factors in pre-menopausal osteoporosis even in the presence of one or two identified risk factors.

Keywords: Premenopausal Osteoporosis; Fragility Fractures; Inflammatory Spondyloarthritis; Vitamin D Deficiency

Introduction

Inflammatory arthritis including Rheumatoid Arthritis (RA) and Ankylosing Spondylitis (AS) are established risk factors for early osteoporosis [1,2]. Pathogenesis of osteoporosis in these patients is multifactorial, contributed by increased inflammatory cytokines (E.g. interleukene-6, tumor necrosis factor alfa), immobility, glucocorticoid treatment and clinically silent bowel disease [3].

Case Report

A 30-year-old lady, 2 weeks after the delivery of her second child presented with non-traumatic vertebral wedge fractures at multiple levels. She was referred to the medical ward for further investigations.

She had no ongoing medical problems and her first pregnancy was uncomplicated. She has electively delayed the second pregnancy for 8 years and used hormonal contraceptive methods. In her second pregnancy, she developed lower backache during early second trimester, which was initially attributed to the pregnancy itself. Retrospective symptom analysis revealed that she had morning stiffness lasting for 2 hours, severe nocturnal pain and pain on movements. These symptoms progressively worsened forcing her to be almost bedbound by the mid-3rd trimester. By this time, she developed a visible deformity of her back and she lost height.

She denied involvement of other joints, skin rashes or systemic symptoms of autoimmune rheumatic diseases. Her bowel habits were unaltered. On further inquiry into risk factors for osteoporosis, she revealed that she had childhood asthma and was given systemic steroids frequently. She took oral and inhalational preparations on and off from 8 years to 16 years of age. She breastfed her first child for 5 years. The only other medication she had taken was the oral contraceptive pill, which she took for 8 years.

On examination she had a visible kyphosis of the dorsal spine. She was in considerable pain. There were no other clinical features of nutritional deficiencies. She had multiple tender points along the spine and sacroiliac joints bilaterally. She had limited chest expansion (2cm); modified Schober's test 3cm; occiput to wall distance of 18cm. Findings of investigations at presentation are summarized in Table 1.

Investigation	Value	Normal range
White cell count (*10^3/uL)	13.8	4.00-10.00
Neutrophils (%)	82%	50-70
Lymphocytes (%)	16%	20-40
Eosinophils (%)	0.3%	0.5-5
Platelets (10*^{3}/uL)	393	150-400
Haemoglobin (g/dL)	12.0	11.0-16.0
Erythrocyte sedimentation rate (mm/1st hour)	97	<20
C-Reactive Protein (mg/dl)	35	<5
AST (U/L)	15	10-35
ALT (U/L)	13	10-40
Albumin (g/L)	38	36-48
Globulin (g/L)	39	22-40
Total bilirubin (umol/L)	8	5-21
Alkaline phosphatase (U/L)	217	100-360
Ionized calcium (mmol/L)	0.97	1.0-1.3
Magnesium	0.80	0.8-1.1
Phosphorus	0.80	0.8-1.5
Para Thyroid Hormone (pg/mL)	37.8	11-62
Vitamin D level (ng/ml)	18.98	50-70

Table 1: Key investigation findings

Contrast Enhanced Computer Tomography (CECT) of the spine showed osteopenia and multiple stable wedge fractures in the vertebral bodies at multiple levels (T8-T11) and erosions of the sacroiliac (SI) joints. This was compatible with chronic erosive sacroilitis. No other intra-abdominal pathology was noted in the CECT. These findings were confirmed by Magnetic Resonance Imaging (MRI) of the spine and SI joints. It also excluded involvement of the spinal cord. DEXA scan (performed using a HOLOGIC Discovery A S/N 80600 device (Hologic Inc., Waltham, MA) showed a Z score of -3.0 each in the lumbar vertebral area and -1.6 each in the hip joints.

She was further investigated for a cause of elevated inflammatory markers. The blood picture was suggestive of an inflammatory state with toxic changes in the neutrophils. Other cell lines were normal. Further investigations to exclude secondary inflammatory conditions and chronic infections were negative (Table 2).

Investigation	Result
Serum protein electrophoresis	Polyclonal gamma globulin band
Anti Nuclear Antibody	negative
Rheumatoid factor	negative
Chest X ray	normal lung fields
Sputum for tuberculosis Tuberculin skin sensitivity test	Microscopy (acid fast staining) and culture negative *3 times, Negative
Urine full report and culture	Normal
Blood culture	Negative (twice)
Bone marrow examination	Increase in granulocyte precursors
Bone marrow culture	Negative bacterial, fungal and tuberculosis culture
STD screening (HIV, VDRL, TPHA)	Negative
HLA B27	Negative
Brucella serology	Negative

Table 2: Results of investigations to look for a secondary cause for osteoporosis

While in the ward she developed unilateral anterior uveitis. This responded to steroid eye drops prescribed after consultation with a specialist ophthalmologist. Recurrent episodes of uveitis did not occur and her vision was not affected.

She was treated with sulfasalazine, calcium-vitamin D supplements and zolendronic acid. She was advised to stop breastfeeding. She was prescribed a lumbar corset. Over a period of two months, her symptoms improved. Repeat DEXA scan performed using the same machine and technique in 6 months demonstrated improved bone mineral density (Z scores for lumbar vertebral area -2.4 and -1.3 for hip joints).

Discussion

A secondary cause can be identified in almost two thirds of patients with premenopausal osteoporosis [4]. Common etiologies include use of drugs (steroids, anticonvulsants), endocrinopathies, nutritional deficiencies and chronic inflammatory conditions [5,6]. Depending on the age at onset and duration, these insults will result in either failure to achieve expected normal bone mass (low baseline bone mass) or loss of pre-formed or newly forming bone. In this patient there were multiple risk factors, acting in unison to produce severe osteoporosis.

Measuring Bone Mineral Density (BMD) using DEXA scan is the most widely used and standardized method for diagnosis of post-menopausal osteoporosis [7,8]. Although commonly used among post-menopausal women, DEXA scan is only indicated in special circumstances among premenopausal women. The indications include history of fragility fractures, having predisposing condition for secondary osteoporosis, or if treatment for osteoporosis is considered [7]. The patient in this case discussion was investigated due to presence of multiple fragility fractures.

The T-score obtained by DEXA scan can only be used for post-menopausal women and men over the age of 50 years [8]. In other categories of patients it is recommended to use the Z score (which indicates the BMD compared with age matched controls) [8].

The FRAX score is an algorithm developed by the World Health Organization to determine fracture risk due to osteoporosis. Although this was initially developed for post-menopausal women and men above the age of 50 years, it is currently validated for females and males between the ages 40-90 years. As our patient is not within this age group, FRAX score could not be applied [8].

Once low bone density is confirmed, especially in a pre-menopausal woman, further investigations are warranted to exclude secondary causes for osteoporosis. This is particularly applicable for patients with BMD Z score <-2.0 or has fragility fractures. A detailed history and examination can identify or exclude most of these conditions and can guide investigations [4]. This case report signifies the importance of carrying out a comprehensive evaluation even if one or more risk factors are identified early in the evaluation. The patient discussed in this case study, had multiple risk factors including possible low baseline bone density due to childhood steroid use, vitamin D deficiency and having a history of prolonged breastfeeding. Recent onset of inflammatory arthritis and pregnancy were the triggers that led to sudden deterioration. Although rare, pregnancy and breastfeeding itself can cause severe osteoporosis, especially in the presence of other risk factors [9,10].

This patient had spondyloartritis with involvement of the sacroiliac joints and early involvement of the spine. Of the spondyloarthropathies, ankylosing spondylitis (AS) is well known to cause osteoporosis even in early or mild stage of the disease [11,12]. Although this patient did not have clear criteria to diagnose AS, this may be the initial presentation where her disease may later evolve into one of inflammatory arthropathies.

Conclusion

This case study is an example of early inflammatory arthritis presenting with pre-menopausal osteoporosis, severe enough to cause multiple vertebral fractures. Pre-menopausal Osteoporosis can be multi factorial and hence it is important to fully evaluate the patient even in the presence of one of more clearly identifiable risk factors. Treating all identified risk factors will facilitate a rapid response and prevention of further complications.

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