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Pregnancy and Lactation Associated Osteoporosis: A Case Report

Hadil Basma[1]* | Sara Dbouk[1] | Zeinab Issa[2]

***Correspondence:** Hadil Basma

Address: [1]Second year fellowship, Department of Endocrinology and Diabetes, Lebanese University, Faculty of Medical Sciences, Beirut, Lebanon; [2]Endocrinologist, Bahman Hospital, Beirut, Lebanon

E-mail ✉: hadilbasma@hotmail.com

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ABSTRACT

Pregnancy and lactation associated osteoporosis (PLO) is an uncommon condition that may present with fragility fractures occurring during pregnancy or the post-partum period. Here, we report a case of 37-year-old woman who presented for lower back pain that started in her third trimester. Her body mass index (BMI) was 22.4, her menarche was at the age of 14 years, and she had no family history of osteoporosis. She sustained several vertebral fractures in her second month postpartum while lactating and then diagnosed with PLO. She was treated with teriparatide for two years. This is the first case of PLO reported in Lebanon to our knowledge. PLO should be suspected in pregnant women with back pain during pregnancy and the postpartum period. Stopping breastfeeding, supplementation with calcium and vitamin D, and treatment with anti-osteoporotic medications are the mainstays of the management of PLO.

Keywords: *Osteoporosis, Pregnancy, Lactation*

Introduction

Epidemiological data on PLO are limited and its pathophysiology is partially explained. Pregnancy and lactation associated osteoporosis (PLO) is a rare type of osteoporosis that can present with vertebral compression fractures and back pain. PLO is typically diagnosed during the third trimester of pregnancy or in the early postpartum phase when lactating. Nordin and Roper reported it for the first time in 1955. (Nordin and Roper, 1995). The underlying mechanism is not well understood; some suggested genetic predisposition with relevant variants found in women with PLO; others found some risk factors related to the disease; but the majority of case reports found no identifiable cause of secondary osteoporosis (Butscheidt *et al*., 2021).

Systematic reviews addressing this topic are scarce. In addition, guidelines addressing this topic are lacking.

Case Presentation

We report the case of a 37-year-old woman who presented to the emergency department one month post-delivery for upper and lower back pain that started during the third trimester of pregnancy and was considered related to pregnancy itself. She sought neurosurgical consultation, and magnetic resonance imaging (MRI) of the lumbar spine with gadolinium showed:

Acute vertebral fracture of T10 presenting a loss of 50% and acute T12 superior endplate fracture with minimal loss of height of 20% (compression fractures), normal conus medullaris. Bone marrow edema at the L4 inferior endplate is compatible with Modic changes type 1.

A Dorsal CT Scan Done with IV Contrast Showed:

Compression fracture at T10, causing 70% loss of height and 60% anterior wedging; fracture in the superior endplate of the T12 vertebral body, causing 35% loss of height. She was referred to an endocrinology clinic, where a workup for secondary osteoporosis was carried out. CBCD showed anemia (patient has thalassemia trait), low 25 OH vitamin D, normal parathyroid hormone (PTH) and calcium, normal thyroid-stimulating hormone (TSH), and a negative workup for celiac disease, Cushing syndrome, and multiple myeloma.

Table 1: Patient's characteristics summarized.

Patient's Characteristics	
Age	37 years
History of fracture	None
Family history of minimal trauma fracture or osteoporosis	None
Regular exercise	None
Number of previous pregnancies	3
Body mass index (BMI)	22.4 Kg/m^2
Age of menarche	14 years

Table 2: Fracture sites.

Fracture site	Bone loss %
T10	50%
T12	20%

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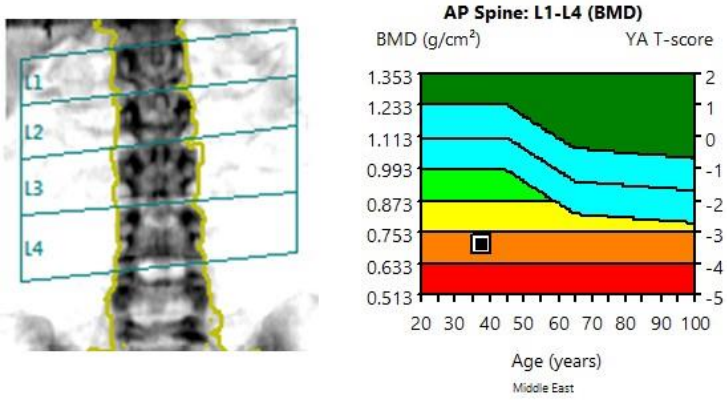
Table 3: Result of Laboratory tests are shown.

Test	unit	Patient' value	Normal range
White blood cells	/µl	7000	11-Apr
Hemoglobin	g/dl	11.8	16-Dec
Hematocrit	%	40.4	37-47
Mean corpuscular volume	fl	62	70-97
Platelets	k/µl	192	140-440
Blood urea nitrogen	mg/dl	14	25-Jul
creatinin	mg/dl	0.62	0.6-1.3
Thyroid stimulating hormone	µIU/mL	1.21	0.27-4.2
Free T4	ng/ml	1.28	0.93-1.71
Antibody anti thyroglobulin	IU/ml	2.07	0-34
Antibody anti thyroperoxidase	IU/ml	3.44	0-12
Vitamin B12	pg/ml	277	187-883
Folic acid	ng/ml	17.1	3.1-20.5
ferritin	ng/ml	25.96	4.63-204
Erythrocytes sedimentation rate	mm/hr	25	0-20
25-hydroxyVitamin D	ng/ml	15.6	Target range:30-40 Toxic level>100
Calcium	mg/dl	9.4	8.4-10.5
phosphorus	mg/dl	4.4	2.5-5
magnesium	mg/dl	2.06	1.6-2.5
Parathyroid hormone	pg/ml	34.38	Oct-69
prolactin	ng/ml	14.2	23-Apr
Plasma electrophoresis	-	normal	-
Serum immunofixation	-	normal	-
Urine cortisol (24 hours urine collection)	µg/24 hrs	25.5	4.3-176/24 hours
Aspartate aminotransferase	IU/L	25	Oct-42
Gamma-glutamyl transferase	IU/L	14	May-37

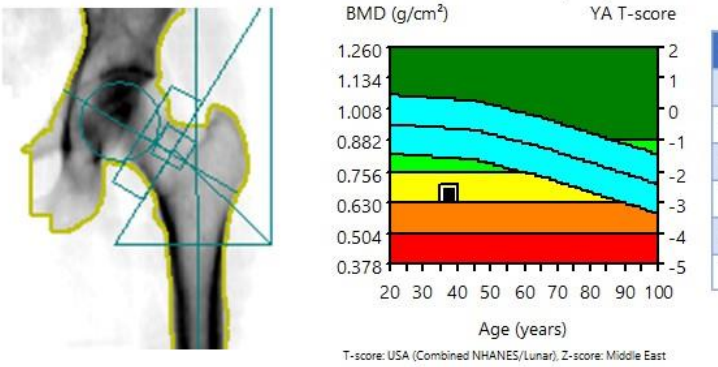
The patient was given calcium and vitamin D replacement (1000 mg calcium and 30000 IU vitamin D per week) and started on teriparatide for two years.

Bone mineral density (BMD) at first visit to clinic in 2018:



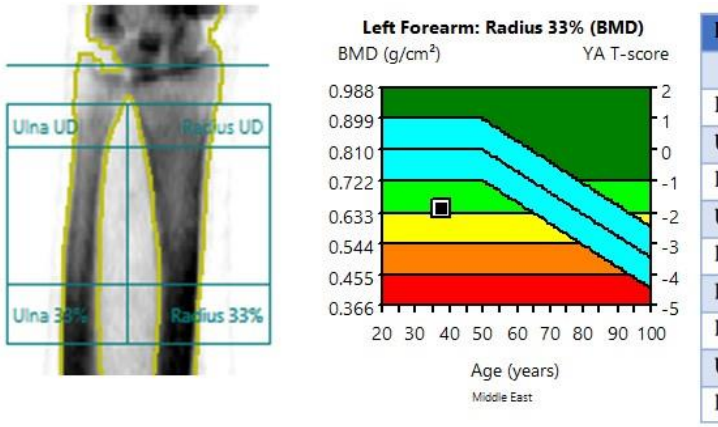
Region	BMD	YA	YA	AM	AM
	(g/cm²)	(%)	T-score	(%)	Z-score
L1	0.763	72	-2.5	72	-2.4
L2	0.676	60	-3.7	60	-3.7
L3	0.675	61	-3.7	61	-3.6
L4	0.724	66	-3.2	66	-3.2
L1-L4	0.707	64	-3.3	64	-3.2
L2-L4	0.693	62	-3.5	62	-3.5

Figure 1: Spine BMD in 2018.



Region	BMD	YA	YA	AM	AM
	(g/cm²)	(%)	T-score	(%)	Z-score
Neck	0.651	63	-2.8	71	-2.2
Upper Neck	0.506	62	-2.6	-	-
Wards	0.488	54	-3.2	59	-2.6
Shaft	0.785	-	-	-	-
Total	0.659	65	-2.8	71	-2.3

Figure 2: Left femur BMD in 2018.

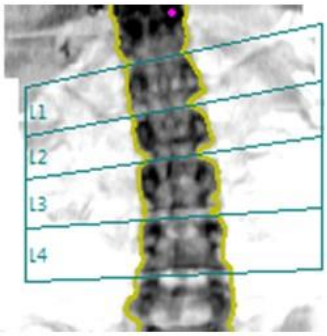


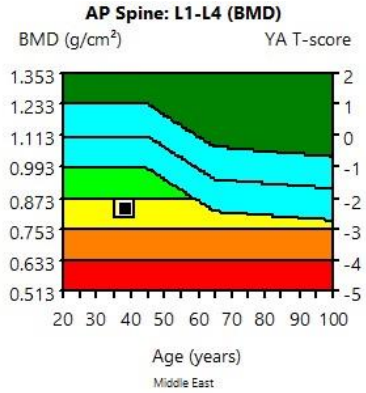
Region	BMD	YA	YA	AM	AM
	(g/cm²)	(%)	T-score	(%)	Z-score
Radius UD	0.409	95	-0.4	95	-0.4
Ulna UD	0.284	-	-	-	-
Radius 33%	0.64	79	-1.9	79	-1.9
Ulna 33%	0.546	-	-	-	-
Both UD	0.365	-	-	-	-
Both 33%	0.595	-	-	-	-
Radius Total	0.509	82	-1.9	82	-1.9
Ulna Total	0.422	-	-	-	-
Both Total	0.474	-	-	-	-

Figure 3: Left forearm BMD in 2018.

Then after 1 year of therapy with teriparatide and mineral supplementation (vitamin D, calcium…):

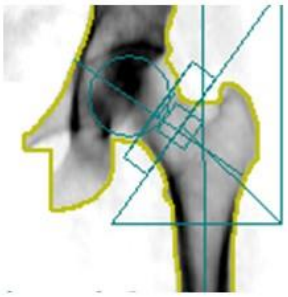
BMD in 2019:

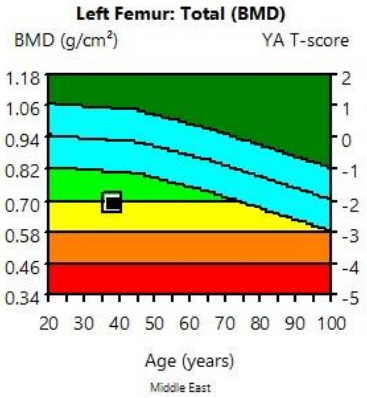




Region	BMD	YA	YA	AM	AM
	(g/cm²)	(%)	T-score	(%)	Z-score
L1	0.78	74	-2.3	74	-2.3
L2	0.841	74	-2.4	75	-2.4
L3	0.859	76	-2.2	77	-2.2
L4	0.83	75	-2.4	75	-2.3
L1-L4	0.829	75	-2.3	75	-2.3
L2-L4	0.843	75	-2.3	75	-2.3

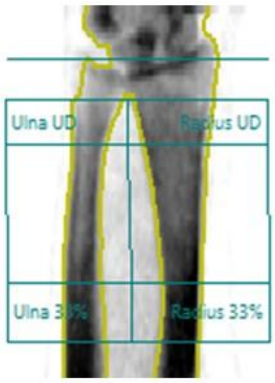
Figure 4: Spine BMD in 2019.

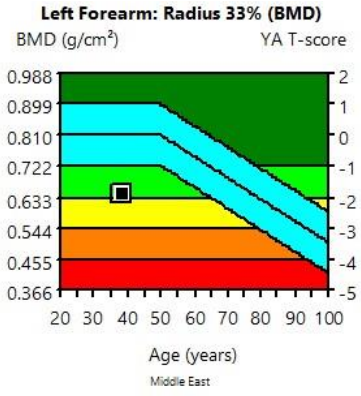




Region	BMD	YA	YA	AM	AM
	(g/cm²)	(%)	T-score	(%)	Z-score
Neck	0.696	75	-1.9	76	-1.8
Upper Neck	0.548	-	-	-	-
Wards	0.518	60	-2.6	63	-2.4
Shaft	0.817	-	-	-	-
Total	0.689	73	-2.1	74	-2

Figure 5: Left femur BMD in 2019.

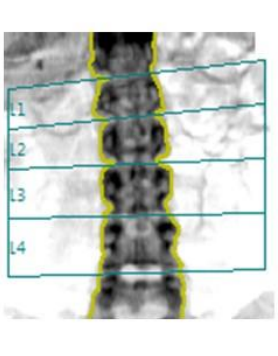


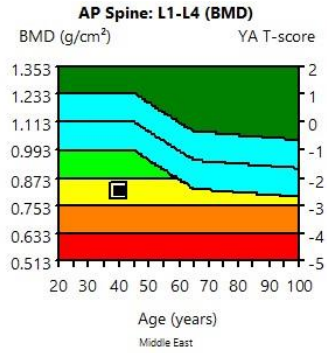


Region	BMD	YA	YA	AM	AM
	(g/cm²)	(%)	T-score	(%)	Z-score
Radius UD	0.411	96	-0.4	96	-0.4
Ulna UD	0.283	-	-	-	-
Radius 33%	0.642	79	-1.9	79	-1.9
Ulna 33%	0.516	-	-	-	-
Both UD	0.364	-	-	-	-
Both 33%	0.578	-	-	-	-
Radius Total	0.497	80	-2.1	80	-2.1
Ulna Total	0.412	-	-	-	-
Both Total	0.462	-	-	-	-

Figure 6: Left forearm BMD in 2019.

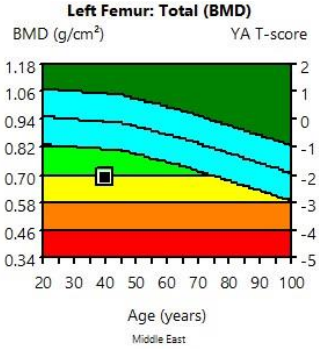
Continuing the same therapy for one more year, BMD became as follows in 2021-2022:





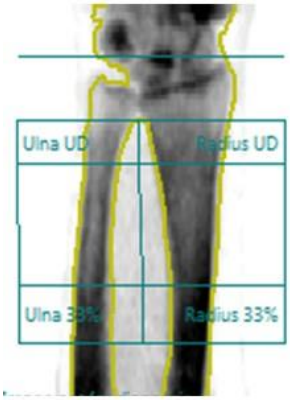
Region	BMD	YA	YA	AM	AM
	(g/cm²)	(%)	T-score	(%)	Z-score
L1	0.763	72	-2.5	72	-2.5
L2	0.796	70	-2.8	70	-2.8
L3	0.869	77	-2.1	77	-2.1
L4	0.803	72	-2.6	72	-2.6
L1-L4	0.811	73	-2.4	73	-2.4
L2-L4	0.822	73	-2.5	73	-2.5

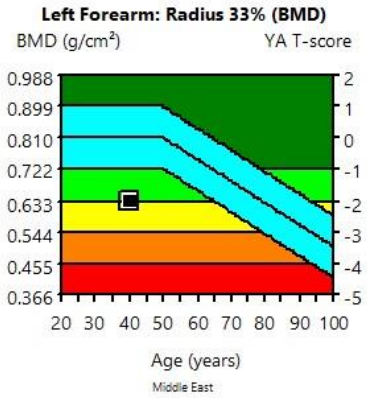
Figure 7: Spine BMD in 2021-2022.



Region	BMD	YA	YA	AM	AM
	(g/cm²)	(%)	T-score	(%)	Z-score
Neck	0.69	74	-2	76	-1.8
Upper Neck	0.545	-	-	-	-
Wards	0.512	60	-2.7	63	-2.4
Shaft	0.819	-	-	-	-
Total	0.692	74	-2.1	74	-2

Figure 8: Left femur BMD in 2021-2022.





Region	BMD	YA	YA	AM	AM
	(g/cm²)	(%)	T-score	(%)	Z-score
Radius UD	0.407	95	-0.5	95	-0.5
Ulna UD	0.293	-	-	-	-
Radius 33%	0.632	78	-2	78	-2
Ulna 33%	0.494	-	-	-	-
Both UD	0.368	-	-	-	-
Both 33%	0.565	-	-	-	-
Radius Total	0.501	80	-2	80	-2
Ulna Total	0.409	-	-	-	-
Both Total	0.465	-	-	-	-

Figure 9: Left forearm BMD in 2021-2022.

Table 4: Summary of BMD results done in each year.

BMD site	BMD (g/cm²) in 2018	BMD (g/cm²) in 2019	BMD (g/cm²) in 2021-2022
L1-L4	0.707	0.829	0.811
RADIUS 33%	0.64	0.642	0.639
LEFT HIP NECK	0.651	0.696	0.69

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Discussion

Pregnancy and lactation are associated with mineral and hormonal changes that may affect bone metabolism. During pregnancy, there is an increase in calcitriol production in the maternal kidneys, possibly in response to an increase in prolactin and placental lactogen. Increased calcitriol leads to enhanced maternal absorption of phosphorus and calcium by the intestines in order to match the increased fetal calcium demand (Sanz-Salvador *et al*., 2014). Lactation is more associated with net bone loss than pregnancy due to the effect of low estradiol, increased parathormone related protein (PTHrP), elevated prolactin. Weight-bearing, lordotic pregnancy posture and immobilization could be added causes. In addition, genetic predisposition and decreased physical activity in the peripubertal period may have a role (kovacs, 2016).

Risk factors for PLO include maternal age above 30 years, smoking, family history of osteoporosis, low BMI, glucocorticoid use in pregnancy, vitamin D deficiency, some anti-psychotic medication usage, gestational diabetes, thrombophilia, anemia, invitro fertilization, twin pregnancy, postpartum thyroiditis, systemic lupus, and others (Carsote *et al*., 2023).

To evaluate, analyze, and describe women with PLO and vertebral fractures, 338 cases from 65 articles were included in a recent systematic review by Ying Qian *et al*. The mean age was 35.7 years, the mean BMI of 46 studies was 22.2 kg/m2 (ranged from 16.0 kg/m2 to 39.0 kg/m2), fracture sites were identified in 155 cases showing 684 vertebral fractures, with an average of 4.4 vertebrae fractured per patient. Most cases had several vertebral fractures, with only 14 single-segment vertebral fractures. As for exact fracture sites, the three most commonly involved vertebral fractures were L1, L2, and T12 (32.6% of all the fractures; 149 out of 173 cases were in primiparity; 19 cases were in the second pregnancy; four cases were in the third pregnancy; and one case was in the fourth pregnancy). 94.4% were breastfeeding (Qian *et al*., 2021).

Two therapeutic approaches were mentioned: conservative and pharmacological. In the conservative approach, lactation cessation was the first and most important step; early mobilization, avoiding heavy lifting, using supporting vertebral corsets, elemental calcium and vitamin D supplementation, and vertebroplasty were considered (Hadji *et al*., 2017; Kovacs and Rlaston, 2015). For the pharmacologic approach, teriparatide is better than bisphosphonate in patients seeking future pregnancies as it has no teratogenic effect and does not stay in the bone matrix (Laroche *et al*., 2017).

Back to our case, the decision to give teriparatide treatment was made due to the severity of the presentation and the pain associated with the severe height loss. Since no randomized controlled trials

comparing the efficacy of treatment head-to-head are present, a terparatide trial on top of the conservative approach was started.

Conclusion

For pregnant or breastfeeding patients with back pain, PLO should be suspected. PLO is usually managed by stopping breastfeeding and taking calcium and vitamin D supplements. Specific pharmacological treatments (bisphosphonates or teriparatide) can be used in selected cases. Further studies are needed to find potential risk factors for PLO and encourage early identification and screening. To our knowledge, this is the first case of PLO published in Lebanon.

Conflict of Interest: none of the authors have any conflict of interest.

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