

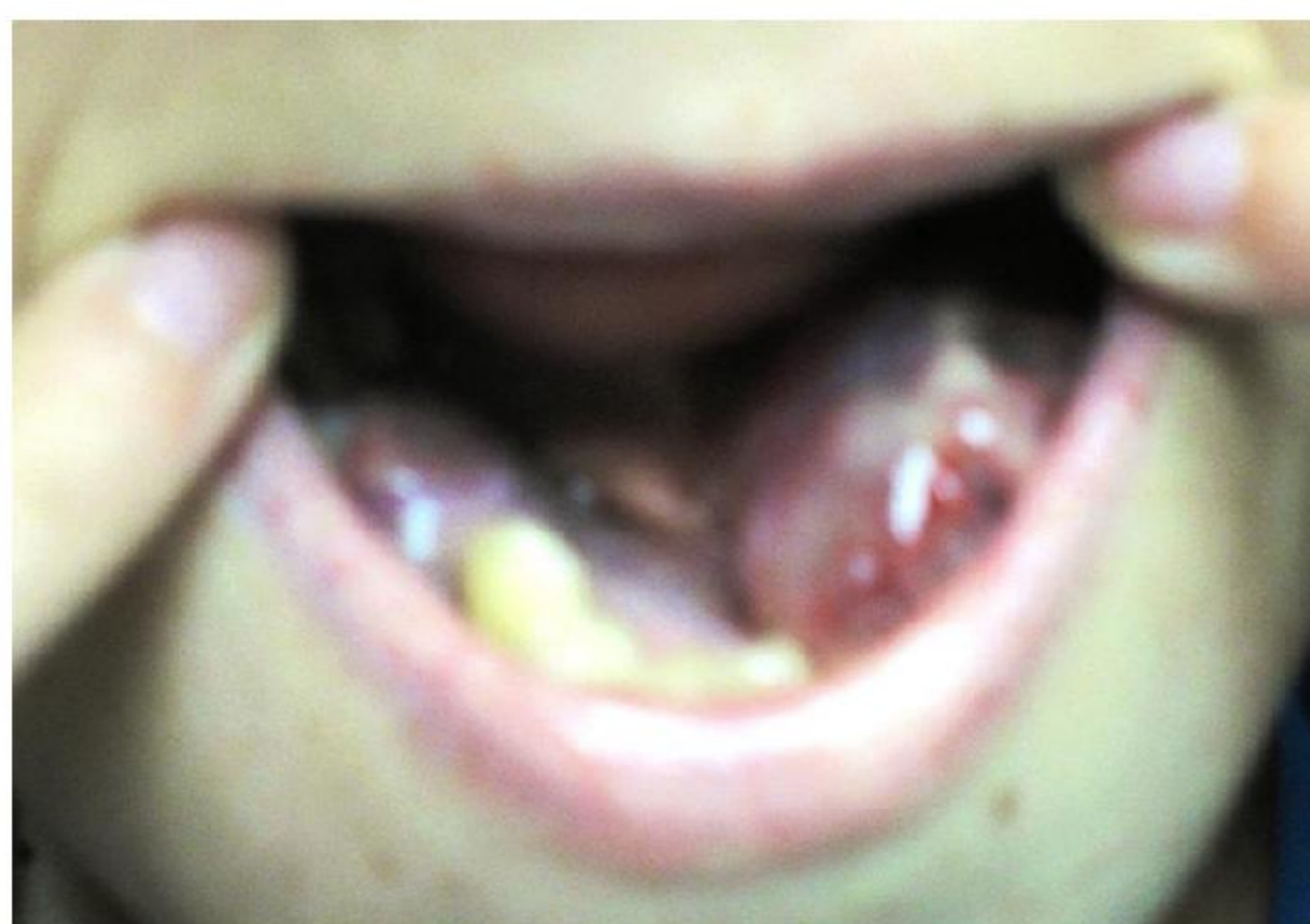
Rapid and profound decrease in resorption markers preceded early and profound increase in BMD after successful surgery in osteitis fibrosa cystica

Daniel Grigorie^{1,2}, Andra Caragheorgheopol¹, Bogdan Stanescu¹, Dumitru Ioachim¹, Maria Iacob¹, Alina Sucaliuc^{1,2}

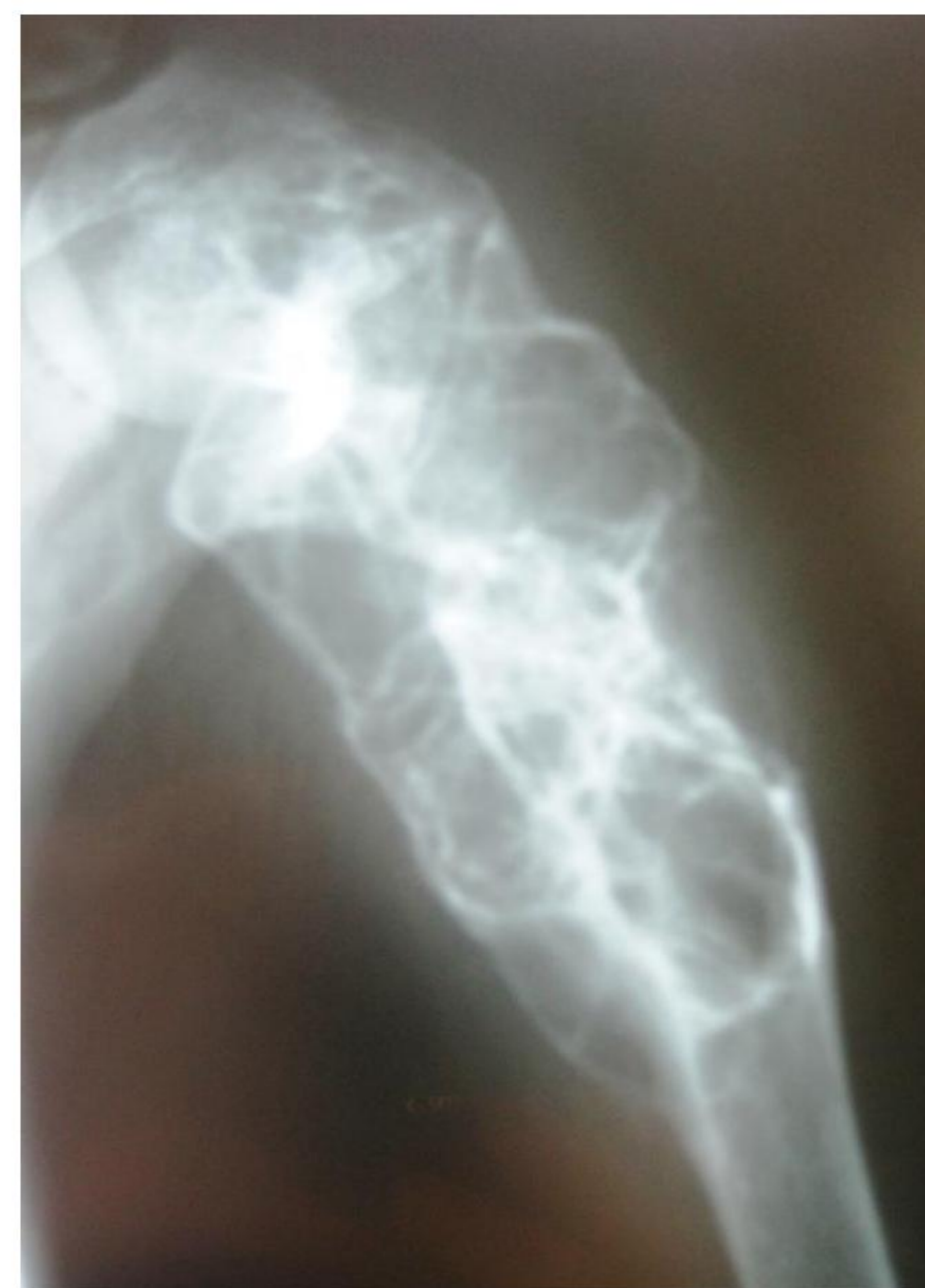
¹National Institute of Endocrinology; ²“Carol Davila” University of Medicine and Pharmacy, Bucharest, Romania

Introduction. Successful parathyroid surgery in patients with OFC leads to early and marked improvements in BMD but data on very early changes in bone markers are missing.

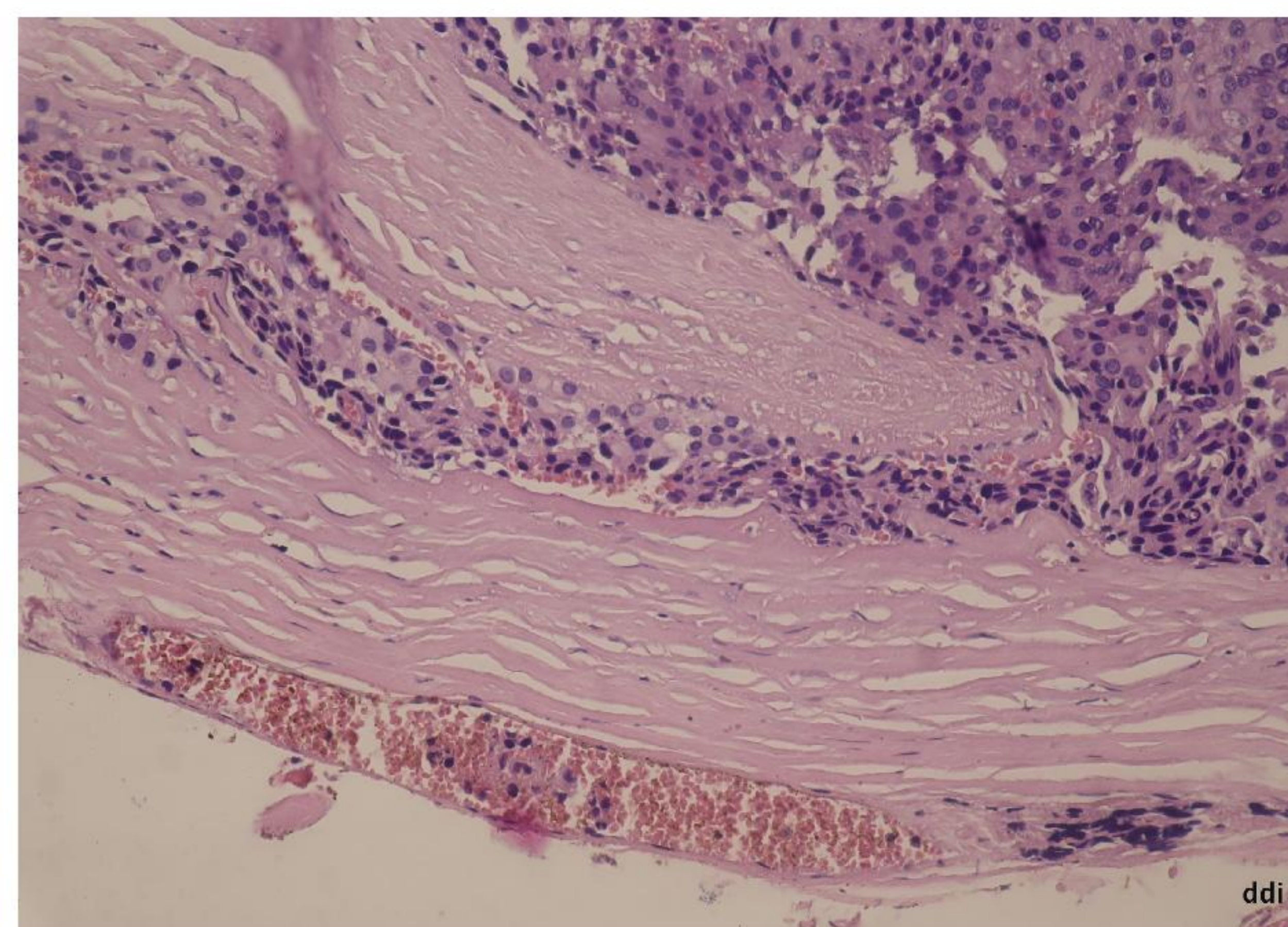
Case report. A 20-yr-old girl was hospitalized because of severe bone pains and multiple pathological fractures (left humerus and left proximal femur) after minimal trauma. She had lost weight, was amenorrheic for the last 12 months and was bleeding from brown tumors in her mouth. She was unable to walk and was complaining for general fatigue, anorexia, polyuria and polydipsia.



Skeletal X-rays revealed multiple osteolytic lesions through the entire skeleton, brown tumors in the mandible and OFC change of the left shoulder.

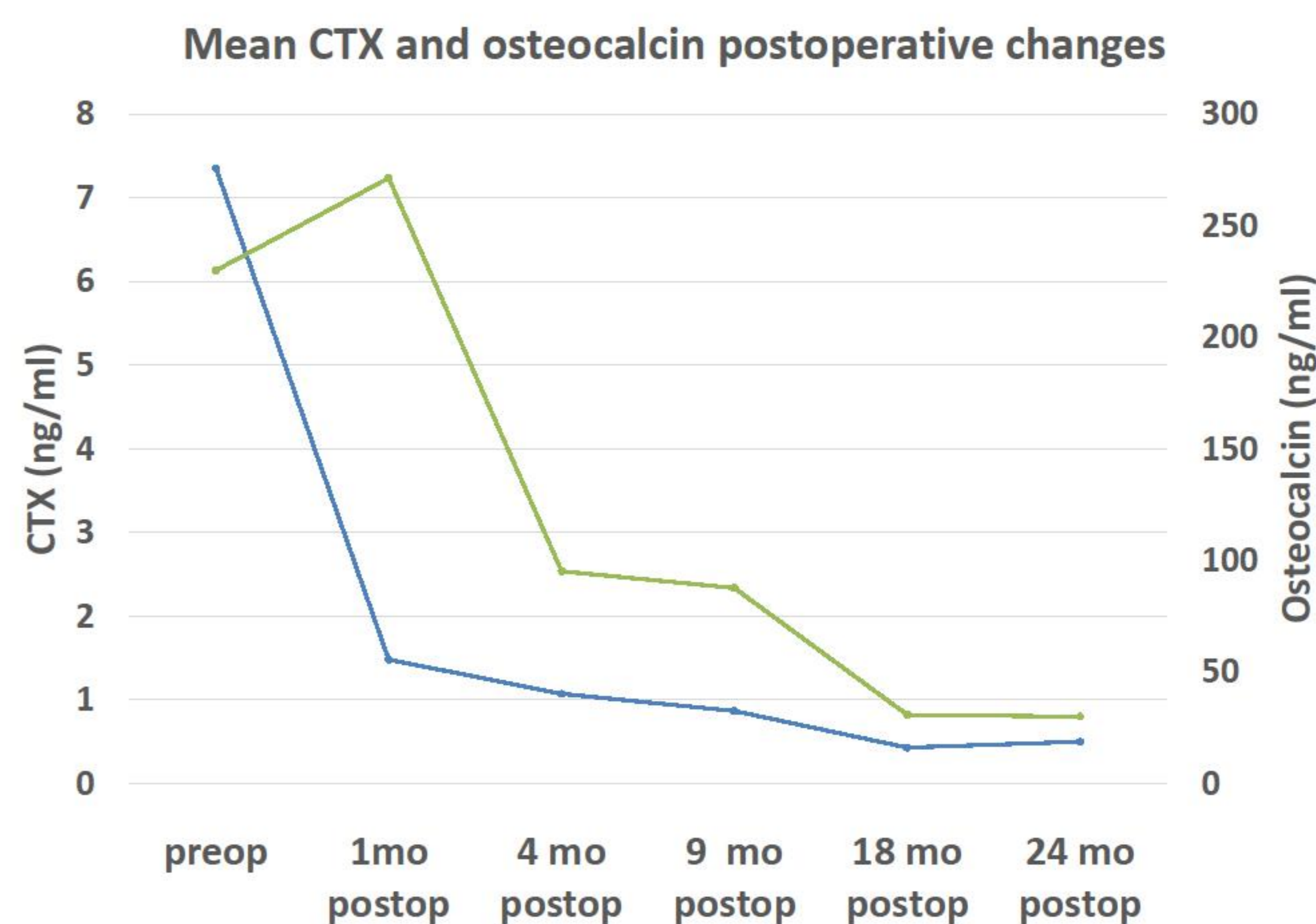


Serum calcium and PTH were of 14.7 mg/dL and 1243 pg/mL, respectively; alkaline phosphatase activity was 1372 U/L. After hydration with iv sodium chloride, the patient was given iv pamidronate (60 mg) and serum Ca fell to 12.4 after 2 days. The patient underwent parathyroid surgery and a right superior parathyroid tumor with capsule and blood vessel invasion (2.2 g) was removed (see figure, HE, x200). She was treated with oral calcium carbonate (2.4g daily) and cholecalciferol 2000 U/daily.



A marked reduction in BMD at LS (0.594 g/cm), FN (0.530 g/cm) and Total Body (0.680 g/cm) was seen by DXA, with Z-scores in the markedly osteoporotic range (-4.9 SD and -3.9 SD, respectively). After 9 mo there were increases of 23.7%, 39.6% and 31.2%, respectively; after 2 yr of 44.1%(LS) and 64%(FN).

In the patient and 3 other similar cases (OFC on X-ray) serum CTX decreased after 1 mo by 3-6 times, while OC increased in one patient by 1.2 times and decreased in the other two by 90% and 53%, respectively. After 12-18 mo CTX and OC were normal in the three patients with available data. (ref. range for CTX 0.142-0.522 ng/ml, for osteocalcin 8.8-37.6 ng/ml)



Conclusion: The cases show an unexpected rapid and profound decrease in resorption after just one mo after surgery while formation decreased at a much slower pace, changes that preceded the marked increase in BMD, especially at the cortical sites.

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Calcium, vitamin D and bone

