Abstract

Introduction: Osteogenesis imperfecta (OI) is a congenital skeletal disorder characterized by bone fragility. Bisphosphonates (BISs) have become the mainstream treatment in children with OI. However, an optimal treatment protocol has not yet been established, while BIS treatment tends to be administered to normalize bone mineral density (BMD). Bone quality is an important component of bone strength. The trabecular bone score (TBS) is a quantitative measure of the microstructure that affects bone quality. This study investigated the TBS during BIS treatment in children with OI.

Materials and Methods: Twenty-nine children with OI were enrolled and classified into two groups: mild (type 1) and moderate to severe (types 3 and 4). Dual-energy X-ray absorptiometry images were retrospectively analyzed for TBS calculation. The relationship between the areal BMD (aBMD), its Z-score, height-adjusted BMD (BMD_{HAZ}) Z-score, TBS, and TBS Z-score with the treatment duration was assessed for each group.

Results: In the mild group, the aBMD, its Z-score, and BMD_{HAZ} Z-score showed a significant positive correlation with treatment duration (r=0.68, 0.68, 0.72, respectively, $p<0.01$). The TBS Z-score tended to increase with treatment duration, albeit without reaching significance. In the moderate to severe group, the TBS Z-score showed a significant positive correlation with treatment duration (r=0.48, $p<0.01$), in contrast to the aBMD Z-score, which did not increase. Finally, the BMD_{HAZ} Z-score only showed a weak positive correlation with treatment duration (r=0.37, $p<0.01$).

Conclusion: Because BIS affect the BMD and TBS differently based on the severity of OI, treatment goals may need to be stratified by disease severity.