

Osteonecrosis in children and young adults treated for acute lymphoblastic leukaemia: a scoping review

Michael Callum Chapman, Maisie Tustian, Joseph D Wilson, Mark A Williams and Robyn J Stiger
Swansea University Medical School

Introduction: Osteonecrosis (ON) is a common disabling complication of treatment for patients with acute lymphoblastic leukaemia (ALL). Reported incidence rates range from 1-61% and multiple possible risk factors have been identified. This review explored existing evidence to provide new perspectives and recommendations for future interdisciplinary research.

Methodology: PEDro, CINAHL, AMED, EMBSAE, OVID, EMCARE databases were systematically searched from their inception to March 2022. Published original research reporting the incidence rates of osteonecrosis in patients aged 10-25 with ALL were included. Study reporting quality was assessed against appropriate reporting guidelines (STROBE, CONSORT and CROSS). All relevant data reporting incidence rates and risk factors were extracted for narrative synthesis.

Results: 3146 report titles were screened, with 34 studies included (n=12,056) (30 observational cohort studies, three randomised trials, and one questionnaire study). The median study quality reporting score was 68% (IQR 64-82%). Median overall incidence rate of ON was 51.8% (IQR 41.4-58.9%) and 15.65% (IQR 9.2-24.2%) for asymptomatic and symptomatic patient screening respectively. Five possible risk factor categories were identified: sex assigned at birth, age, ethnicity, steroid regimen, and genotype. The female sex and white ethnicity were consistently reported as risk factors independently associated with an increased risk of osteonecrosis in all studies.

Conclusion: A heterogeneous body of literature with moderate reporting quality identified a high incidence rate of osteonecrosis in patients with ALL. Future research investigating the efficacy of stratified treatments that focus on reducing the risk of osteonecrosis through modification of steroid regimen particularly in females of white ethnicity is needed. Obtaining multidisciplinary consensus with regards to screening methodologies and intervention outcomes may also help to improve evidence synthesis in this area. This may in turn facilitate early diagnosis and improve long term patient outcomes through treatment regimen modification and possible prevention of ON progression.

