

dependently manner. In addition, PXD-101 inhibited about 40% the migration ability of Saos2 and 143B osteosarcoma cell lines [Wound closure (%). Saos2, Vehicle:  $99.9 \pm 0.4$ , PXD-101:  $63.2 \pm 2.1$ ,  $p < 0.0001$ . 143B, Vehicle:  $100.0 \pm 0.4$ , PXD-101:  $70.5 \pm 3.2$ ,  $p < 0.0001$ ]. These features were associated with a significant reduction of mRNA and protein expression of osteoblast markers (ALP, RUNX2, OSTERIX) in the human osteosarcoma Saos2 cell line.

**Conclusion:** Taken together, our study reveals the antitumor activity of PXD-101 in osteosarcoma cells suggesting a new potential therapeutic approach for osteosarcoma.

doi:[10.1016/j.bonr.2022.101488](https://doi.org/10.1016/j.bonr.2022.101488)

## P238

### Identification of differentially expressed immune receptor genes in osteosarcoma

Shasheen Payoe<sup>a</sup>, Angilla Safa<sup>a</sup>, Annette Payne<sup>b</sup>, Gudrun Stenbeck<sup>a</sup>

<sup>a</sup>Brunel University London, Life Sciences, London, United Kingdom

<sup>b</sup>Brunel University London, Computer Sciences, London, United Kingdom

Osteosarcoma (OS) is the most common primary malignant bone tumour, with a high incidence rate in children and adolescents. However, knowledge surrounding the underlying mechanism(s) and novel therapeutics, require better overall understanding. One important area in need of further exploration is the tumour microenvironment, which consists of a plethora of extracellular matrix components, tumour associated fibroblasts and immune cells. To identify new druggable targets, we searched an OS gene microarray dataset deposited within the Gene Expression Omnibus, which included 84 primary OS biopsies and 12 primary Mesenchymal stem cell control samples.

An alternative to R coding was utilised to identify differentially expressed genes (DEGs) in OS. The web tool (GEO2R) utilises the Bioconductor “limma” package to determine DEGs. In addition, R-coding with the ‘Weighted Gene Co-expression Network Analysis’ package, was used to construct a weighted gene co-expression network, thus, identifying gene modules associated with OS. Functional annotations were conducted via Gene Ontology (GO) and Kyoto Encyclopedia of Genes and Genomes overrepresentation analyses. Upregulated genes were additionally refined, based on the GO term “immune receptor activity”.

The results identified 267 DEGs in OS, consisting of 142 upregulated and 125 downregulated genes, with  $>2$  fold-change and an adjusted P-value  $<0.01$  cut-offs. Functional annotations showed the DEGs were involved in immune system processes, including defence and general immune responses. This suggests a dysregulation of the immune system is strongly linked to the OS microenvironment, with DEGs potentially contributing to OS development and metastasis. Via GO based refinement, immune receptors CXCR4 and CD74 were found upregulated in OS[GS(1)] (fold-change (3.38 and 4.35) and adjusted P-values (5.74E-26 and 7.8E-17), respectively). CXCR4 has been associated with other metastatic cancers, validating our approach. In summary, immune system dysregulation may contribute to OS development, with the upregulated immune receptors being potential druggable targets for novel drug delivery systems.

doi:[10.1016/j.bonr.2022.101489](https://doi.org/10.1016/j.bonr.2022.101489)

## P239

### Effectiveness of pamidronate therapy in chronic recurrent multifocal osteomyelitis (CRMO) with thoracic/lumbal spine localization – a case report

Aleksandra Opala<sup>a</sup>, Aleksandra Blachowska<sup>b</sup>, Pawel Matusik<sup>a</sup>

<sup>a</sup>Medical University of Silesia- Katowice- Poland, Department of Pediatrics- Pediatric Obesity and Metabolic Bone Diseases- Faculty of Medical Sciences in Katowice, Katowice, Poland

<sup>b</sup>Medical University of Silesia- Katowice- Poland, Department of Pediatrics- Pediatric Obesity and Metabolic Bone Diseases- Faculty of Medical Sciences in Katowice, Katowice, Poland

**Background:** Chronic recurrent multifocal osteomyelitis (CRMO), is a chronic noninfectious inflammatory disorder resulting the multifocal bone and bone marrow lesions with periodic relapses and remissions with the uncertain prognosis. Treatment options in CRMO is based on the expert opinion and relatively small case series, and the main options considered are: non-steroidal anti-inflammatory drugs (NSAIDs), corticosteroids, disease-modifying anti-rheumatic drugs (like methotrexate or sulfasalazine), anti-TNFagents, or bisphosphonates.

**Case presentation:** We describe an 11 years old girl with multifocal presentation of bone lesions in the thoracic and lumbal vertebral bodies with the presence of compression fractures. The patient was successfully treated with three days pulses of pamidronate (1 mg/kg/body weight/day) for every 3 months. In addition to clinical improvement, there was a significant remission of the inflammation and bone stucture healing assessed by MRI after 4 treatment cycles.

**Conclusions:** Intravenous bisphosphonates usage seems to be a good therapeutic option in CRMO pediatric patients with spinal localization of the lesions. We still need more data based on larger groups of patients to deliver detailed algoritm for the CRMO therapy in pediatric population.

doi:[10.1016/j.bonr.2022.101490](https://doi.org/10.1016/j.bonr.2022.101490)

## P240

### Synovitis, acne, pustulosis, hyperostosis, and osteitis syndrome with purely osteolytic, not osteosclerotic, lesions mimicking a malignant tumor

Hideyuki Kinoshita<sup>a</sup>

<sup>a</sup>Chiba Cancer Center, Orthopaedic Surgery, Chiba, Japan

Synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO) syndrome is a rare inflammatory disorder with multiple phenotypes. The syndrome has identifiable radiologic characteristics that are the most important when making a diagnosis. X-rays of cases diagnosed with SAPHO syndrome reveal sclerotic lesions or mixed lytic and sclerotic lesions. Pure osteolytic lesions in SAPHO syndrome are rare, and to the best of our knowledge, no study has reported the radiologic change of purely osteolytic lesions to osteosclerotic lesions over time. Herein, we report on the case of a woman experiencing severe left thigh acute pain and having a medical history of palmo-plantar pustulosis. Although SAPHO syndrome was suspected because of palmo-plantar pustulosis, based on radiologic findings, bone metastasis of a malignant tumor or chronic bacterial osteomyelitis owing to a purely osteolytic lesion was suspected. However, needle biopsy revealed no malignancy and bacterial culture was negative, thus suggesting SAPHO syndrome. Nonsteroidal anti-inflammatory drugs, bisphosphonates, and