

Publications acknowledging funding from the Bone Cancer Research Trust

- 111.** Balaraman K, Deniz E, Nelson E, Pilicer SL, Atasoy S, Molotkova A *et al.* Design, synthesis and biological evaluation of Nucleosidic CD99 inhibitors that selectively reduce Ewing sarcoma viability. *European Journal of Medicinal Chemistry*, **2023**; 251, 115244. DOI: [10.1016/j.ejmech.2023.115244](https://doi.org/10.1016/j.ejmech.2023.115244).
- 110.** Green D, Sing A, Tippett VL, Tattersall L, Shah KM, Siachisomo C, *et al.* YBX1-interacting small RNAs and RUNX2 can be blocked in primary bone cancer using CADD522. *Journal of Bone Oncology*, **2023**; 39, 100474. DOI: [10.1016/j.jbo.2023.100474](https://doi.org/10.1016/j.jbo.2023.100474).
- 109.** Cortes-Ciriano I, Steele CD, Piculell K, Al-Ibraheemi A, Eulo V, Bui MM *et al.* Genomic Patterns of Malignant Peripheral Nerve Sheath Tumor (MPNST) Evolution Correlate with Clinical Outcome and Are Detectable in Cell-Free DNA. *Cancer Discovery*, **2023**; 13 (3), 654-671. DOI: [10.1158/2159-8290.CD-22-0786](https://doi.org/10.1158/2159-8290.CD-22-0786).
- 108.** Ball N, Duncan S, Zhang YY, Payet R, Piec, I Whittle E, *et al.* 3' Untranslated Region Structural Elements in CYP24A1 Are Associated With Infantile Hypercalcemia Type 1. *Journal of Bone and Mineral Research*, **2023**; 38 (3), 414-426. DOI: [10.1002/jbmr.4769](https://doi.org/10.1002/jbmr.4769).
- 107.** Brookes MJ, Roundhill EA, Jeys L, Parry M, Burchill SA, Rankin KS. Comment on: Membrane-type 1 matrix metalloproteinase as predictor of survival and candidate therapeutic target in Ewing sarcoma. *Pediatric Blood & Cancer*, **2023**; 70 (5), e30206. <https://doi.org/10.1002/pbc.30206>.
- 106.** Passeri G, Vincent RA, Xiao ZY, Northcote-Smith J, Suntharalingam K. Encapsulation and Delivery of an Osteosarcoma Stem Cell Active Gallium(III)-Diflunisal Complex Using Polymeric Micelles. *ChemMedChem*, **2023**; 18 (4). DOI: [10.1002/cmdc.202200599](https://doi.org/10.1002/cmdc.202200599).
- 105.** Kurata K, Bott AJ, Tye MA, Yamamoto L, Samur MK, Tai YT, *et al.* Prolyl-tRNA synthetase as a novel therapeutic target in multiple myeloma. *Blood Cancer Journal*, **2023**; 13 (1), 12. DOI: [10.1038/s41408-023-00787-w](https://doi.org/10.1038/s41408-023-00787-w).
- 104.** De Noon S, Piggott R, Trotman J, Tadross JA, Fittall M, Hughes D, *et al.* Recurrent FOSL1 rearrangements in desmoplastic fibroblastoma. *The Journal of Pathology*, **2023**; 259 (2), 119-124. <https://doi.org/10.1002/path.6038>.
- 103.** Beird HC, Bielack SS, Flanagan AM, Gill J, Heymann D, Janeway KA, *et al.* Osteosarcoma. *Nature Reviews Disease Primers*. **2022**; 8 (1), 77. <https://doi.org/10.1038/s41572-022-00409-y>
- 102.** Vincent RA, Passeri G, Northcote-Smith J, Singh K, Suntharalingam K. The Osteosarcoma Stem Cell Activity of a Gallium(III)-Phenanthroline Complex Appended to Salicylate. *ChemBioChem*, **2022**; 23 (24). DOI: [10.1002/cbic.202200532](https://doi.org/10.1002/cbic.202200532).
- 101.** Brookes MJ, Roundhill EA, Jeys L, Parry M, Burchill SA, Rankin KS. Membrane-type 1 matrix metalloproteinase as predictor of survival and candidate therapeutic target in Ewing sarcoma. *Pediatric Blood & Cancer*, **2022**; 69 (12) e29959. DOI: [10.1002/pbc.24272](https://doi.org/10.1002/pbc.24272).
- 100.** Cottone L, Ligammarri L, Lee HM, Knowles HJ, Henderson S, Bianco S, *et al.* Aberrant paracrine signalling for bone remodelling underlies the mutant histone-driven giant cell tumour of bone. *Cell Death and Differentiation*, **2022**; 29 (12), 2459-2471. DOI: [10.1038/s41418-022-01031-x](https://doi.org/10.1038/s41418-022-01031-x).

- 99.** Usher I, Ligammar L, Ahrabi S, Hepburn E, Connolly C, Bond GL, *et al.* Optimizing CRISPR/Cas9 Editing of Repetitive Single Nucleotide Variants. *Frontiers in Genome Editing*, **2022**; 4, 932434. DOI: [10.3389/fgeed.2022.932434](https://doi.org/10.3389/fgeed.2022.932434).
- 98.** Park J, Jia SY, Salter D, Bagnaninchi P, Hansen CG. The Hippo pathway drives the cellular response to hydrostatic pressure. *EMBO Journal*, **2022**; 41 (13), e108719. DOI: [10.15252/embj.2021108719](https://doi.org/10.15252/embj.2021108719).
- 97.** Lyskjaer I, Kara N, De Noon S, Davies C, Rocha AM, Strobl AC, *et al.* Osteosarcoma: Novel prognostic biomarkers using circulating and cell-free tumour DNA. *European Journal of Cancer*, **2022**; 168, 1-11. DOI: [10.1016/j.ejca.2022.03.002](https://doi.org/10.1016/j.ejca.2022.03.002).
- 96.** Cunningham R, Hansen CG. The Hippo pathway in cancer: YAP/TAZ and TEAD as therapeutic targets in cancer. *Clinical Science*, **2022**; 136 (3), 197-222. DOI: [10.1042/CS20201474](https://doi.org/10.1042/CS20201474).
- 95.** Riley SE, Feng Y, Hansen CG. Hippo-Yap/Taz signalling in zebrafish regeneration. *NPJ Regenerative Medicine*, **2022**; 7 (1), 9. DOI: [10.1038/s41536-022-00209-8](https://doi.org/10.1038/s41536-022-00209-8).
- 94.** Baldwin MJ, Mimpin JY, Cribbs AP, Stace E, Philpott M, Dakin SG, *et al.* Electrospun Scaffold Micro-Architecture Induces an Activated Transcriptional Phenotype within Tendon Fibroblasts. *Frontiers in Bioengineering and biotechnology*, **2022**; 9, 795748. DOI: [10.3389/fbioe.2021.795748](https://doi.org/10.3389/fbioe.2021.795748).
- 93.** Spencer HLM, Shnyder SD, Loadman PM, Falconer RA. The role of MT1-MMP in the progression and metastasis of osteosarcoma. *Journal of Cancer Metastasis and Treatment*, **2022**; 8, 2. <http://dx.doi.org/10.20517/2394-4722.2021.174>.
- 92.** Tattersall L, Shah KM, Lath DL, Singh A, Down JM, De Marchi E, *et al.* The P2RX7B splice variant modulates osteosarcoma cell behaviour and metastatic properties. *Journal of Bone Oncology*, **2021**; 31 100398. DOI: [10.1016/j.jbo.2021.100398](https://doi.org/10.1016/j.jbo.2021.100398).
- 91.** Lyskjaer I, Davies C, Strobl AC, Hindley J, James S, Lalam RK, *et al.* Circulating tumour DNA is a promising biomarker for risk stratification of central chondrosarcoma with IDH1/2 and GNAS mutations. *Molecular Oncology*, **2021**; 15 (12), 3679-3690. DOI: [10.1002/1878-0261.13102](https://doi.org/10.1002/1878-0261.13102).
- 90.** Roundhill EA, Chicon-Bosch M, Jeys L, Parry M, Rankin KS, Droop A, Burchill SA. RNA sequencing and functional studies of patient-derived cells reveal that neurexin-1 and regulators of this pathway are associated with poor outcomes in Ewing sarcoma. *Cellular Oncology*, **2021**; 44 (5) 1065-1085. DOI: [10.1007/s13402-021-00619-8](https://doi.org/10.1007/s13402-021-00619-8).
- 89.** Park J, Hansen CG. Cellular feedback dynamics and multilevel regulation driven by the hippo pathway. *Biochemical Society Transactions*, **2021**; 49 (4) 1515-1527. DOI: [10.1042/BST20200253](https://doi.org/10.1042/BST20200253).
- 88.** Tarabichi M, Demeulemeester J, Verfaillie A, Flanagan AM, Van Loo P, Konopka T. A pan-cancer landscape of somatic mutations in non-unique regions of the human genome. *Nature Biotechnology*, **2021**; 39 (12), 1589-1596. DOI: [10.1038/s41587-021-00971-y](https://doi.org/10.1038/s41587-021-00971-y).
- 87.** Philpott M, Watson J, Thakurta A, Brown T, Brown T, Oppermann U, Cribbs AP. Nanopore sequencing of single-cell transcriptomes with scCOLOR-seq. *Nature Biotechnology*, **2021**; 39 (12), 1517-1520. DOI: [10.1038/s41587-021-00965-w](https://doi.org/10.1038/s41587-021-00965-w).
- 86.** De Noon S, Ijaz J, Coorens THH, Amary F, Ye H, Strobl A, *et al.* MYC amplifications are common events in childhood osteosarcoma. *The Journal of Pathology Clinical Research*, **2021**; 7:425-431 <https://doi.org/10.1002/cjp2.219>.

- 85.** Lyskjær I, De Noon S, Tirabosco R, Ana Maia Rocha AM, Lindsay D, Amary F, *et al.* DNA methylation-based profiling of bone and soft tissue tumours: a validation study of the 'DKFZ Sarcoma Classifier'. *The Journal of Pathology Clinical Research*, **2021**; 7 (4), 350-360. <https://doi.org/10.1002/cjp2.215>.
- 84.** Cribbs AP, Filippakopoulos P, Philpott M, Wells G, Penn H, Oerum H *et al.* Dissecting the Role of BET Bromodomain Proteins BRD2 and BRD4 in Human NK Cell Function. *Frontiers in Immunology*, **2021**; 12, 626255. DOI: [10.3389/fimmu.2021.626255](https://doi.org/10.3389/fimmu.2021.626255).
- 83.** Gerrand C, Bate J, Seddon B, Dirksen U, Randall RL, van de Sande M, *et al.* Seeking international consensus on approaches to primary tumour treatment in Ewing sarcoma. *Clinical Sarcoma Research*, **2020**; 10 (1), 21. DOI: [10.1186/s13569-020-00144-6](https://doi.org/10.1186/s13569-020-00144-6).
- 82.** Muller L, Berkeley R, Barr T, Ilett E, Errington-Mais F. Past, Present and Future of Oncolytic Reovirus. *Cancers*, **2020**; 12 (11), 3219. DOI: [10.3390/cancers12113219](https://doi.org/10.3390/cancers12113219).
- 81.** Cottone L, Cribbs AP, Khandelwal G, Wells G, Ligammar L, *et al.* Inhibition of Histone H3K27 Demethylases Inactivates Brachyury (TBXT) and Promotes Chordoma Cell Death. *Cancer Research*, **2020**; 80 (20), 4540-4551. DOI: [10.1158/0008-5472.CAN-20-1387](https://doi.org/10.1158/0008-5472.CAN-20-1387).
- 80.** Fittall MW, Lyskjaer I, Ellery P, Lombard P, Ijaz J, Strobl AC, *et al.* Drivers underpinning the malignant transformation of giant cell tumour of bone. *Journal of Pathology*, **2020**; 252 (4), 433-440. DOI: [10.1002/path.5537](https://doi.org/10.1002/path.5537).
- 79.** Rogers MJ, Monkkonen J, Munoz MA. Molecular mechanisms of action of bisphosphonates and new insights into their effects outside the skeleton. *Bone*, **2020**; 139, Article Number: 115493. DOI: [10.1016/j.bone.2020.115493](https://doi.org/10.1016/j.bone.2020.115493).
- 78.** Curtin NJ, Szabo C. Poly (ADP-ribose) polymerase inhibition: past, present and future. *Nature Reviews Drug Discovery*, **2020**; 19, (10), 711-736. DOI: [10.1038/s41573-020-0076-6](https://doi.org/10.1038/s41573-020-0076-6).
- 77.** Philpott M, Cribbs AP, Brown T Jr, Brown T Sr; Oppermann U. Advances and challenges in epigenomic single-cell sequencing applications. *Current Opinion in Chemical Biology*, **2020**; 57 (SI), 17-26. DOI: [10.1016/j.cbpa.2020.01.013](https://doi.org/10.1016/j.cbpa.2020.01.013).
- 76.** De Noon S, Flanagan AM, Tirabosco R, O'Donnell P, Amary F. EWSR1-SMAD3 fibroblastic tumour of bone: expanding the clinical spectrum. *Skeletal Radiology*, **2020**; 50 (2), 445-450. DOI: [10.1007/s00256-020-03563-0](https://doi.org/10.1007/s00256-020-03563-0).
- 75.** Prendergast SC, Strobl AC, Cross W, Pillay N, Strauss SJ, Ye HT, *et al.* Sarcoma and the 100,000 Genomes Project: our experience and changes to practice. *Journal of Pathology Clinical Research*, **2020**; 6 (4), 297-307. DOI: [10.1002/cjp2.174](https://doi.org/10.1002/cjp2.174).
- 74.** Mirabello L, Zhu B, Koster R, Karlins E, Dean M, Yeager M, *et al.* Frequency of Pathogenic Germline Variants in Cancer-Susceptibility Genes in Patients with Osteosarcoma. *JAMA Oncology*, **2020**; 6 (5), 724-734. DOI: [10.1001/jamaoncol.2020.0197](https://doi.org/10.1001/jamaoncol.2020.0197).
- 73.** Gama A, Vargas-Franco JW, Mesa DCS, Bedoya ER, Amiaud J, Babajko, S, *et al.* Origins of Alterations to Rankl Null Mutant Mouse Dental Root Development. *International Journal of Molecular Sciences*, **2020**; 21, (6), 2201. <https://doi.org/10.3390/ijms21062201>.
- 72.** Kosir U, Bowes L, Taylor RM, Gerrand C, Windsor R, Onasanya M, Martins A. Psychological adaptation and recovery in youth with sarcoma: a qualitative study with practical implications for clinical care and research. *BMJ Open*, **2020**; Article Number: e038799. DOI: [10.1136/bmjopen-2020-038799](https://doi.org/10.1136/bmjopen-2020-038799).
- 71.** Amary F, Perez-Casanova L, Ye HT, Cottone L, Strobl AC, Cool P, *et al.* Synovial chondromatosis and soft tissue chondroma: extraosseous cartilaginous tumor defined by FN1 gene rearrangement. *Modern Pathology*, **2019**; 32, (12), 1762-1771. DOI: [10.1038/s41379-019-0315-8](https://doi.org/10.1038/s41379-019-0315-8).

- 70.** Vargas-Franco JW, Castaneda B, Gama A, Mueller CG, Heymann D, Redini, F, *et al.* Genetically-achieved disturbances to the expression levels of TNFSF11 receptors modulate the effects of zoledronic acid on growing mouse skeletons. *Biochemical Pharmacology* **2019**; 168, 133-148. DOI: [10.1016/j.bcp.2019.06.027](https://doi.org/10.1016/j.bcp.2019.06.027).
- 69.** Martins A, Storey L, Wells M, Fern LA, Gerrard C, Bennister L, *et al.* Qualitative study of patients' experiences of living with and beyond a soft tissue sarcoma diagnosis: The impact of sarcoma specialist services. *Annals of Oncology*, **2019**; Meeting Abstract:1681P, 30 (S5), 690-690. <https://doi.org/10.1093/annonc/mdz283.014>.
- 68.** Phillips KL, Wright N, McDermott E, Cross, N. TRAIL responses are enhanced by nuclear export inhibition in osteosarcoma. *Biochemical and Biophysical Research Communications*, **2019**; 517 (2), 383-389. DOI: [10.1016/j.bbrc.2019.07.047](https://doi.org/10.1016/j.bbrc.2019.07.047).
- 67.** Pavlou M, Shah M, Gikas P, Briggs T, Roberts SJ, Cheema U. Osteomimetic matrix components alter cell migration and drug response in a 3D tumour-engineered osteosarcoma model. *Acta Biomaterialia*, **2019**; 96, 247-257. DOI: [10.1016/j.actbio.2019.07.011](https://doi.org/10.1016/j.actbio.2019.07.011).
- 66.** Martins A, Whelan JS, Bennister L, Fern LA, Gerrard C, Onasanya M, *et al.* Qualitative study exploring patients' experiences of being diagnosed and living with primary bone cancer in the UK. *BMJ Open*, **2019**; 9 (9), Article Number: e028693. DOI: [10.1136/bmjopen-2018-028693](https://doi.org/10.1136/bmjopen-2018-028693).
- 65.** Heymann MF, Lezot F, Heyman D. The contribution of immune infiltrates and the local microenvironment in the pathogenesis of osteosarcoma. *Cellular Immunology*, **2019**; 343, Special Issue, Article Number:103711. DOI: [10.1016/j.cellimm.2017.10.011](https://doi.org/10.1016/j.cellimm.2017.10.011).
- 64.** Barrell WB, Griffin JN, Harvey JL, Danovi D, Beales P, Grigoriadis AE, Liu, KJ. Induction of Neural Crest Stem Cells from Bardet-Biedl Syndrome Patient Derived hiPSCs. *Frontiers in Molecular Neuroscience*, **2019**; 12, Article Number:139. DOI: [10.3389/fnmol.2019.00139](https://doi.org/10.3389/fnmol.2019.00139).
- 63.** Brown HK, Tellez-Gabriel M, Cartron PF, Vallette FM, Heymann MF, Heymann, D. Characterization of circulating tumor cells as a reflection of the tumor heterogeneity: myth or reality? *Drug Discovery Today*, **2019**; 24 (3), 763-772. <https://doi.org/10.1016/j.drudis.2018.11.017>.
- 62.** Lavender V, Gibson F, Brownsdon A, Fern, L, Whelan J, Pearce S. Health professional perceptions of communicating with adolescents and young adults about bone cancer clinical trial participation. *Supportive Care in Cancer*, **2019**; 27 (2), 467-475. DOI: [10.1007/s00520-018-4337-4](https://doi.org/10.1007/s00520-018-4337-4).
- 61.** Tellez-Gabriel M, Cochonneau D, Cade M, Jubelin C, Heymann MF, Heymann, D. Circulating Tumor Cell-Derived Pre-Clinical Models for Personalized Medicine. *Cancers*, **2019**; 11 (1), Article Number:19. DOI: [10.3390/cancers11010019](https://doi.org/10.3390/cancers11010019).
- 60.** Navet B, Ando K, Vargas-Franco JW, Brion R, Amiaud J, Mori, K *et al.* The Intrinsic and Extrinsic Implications of RANKL/RANK Signaling in Osteosarcoma: From Tumor Initiation to Lung Metastases. *Cancers*, **2018**; 10 (11), Article Number:398. DOI: [10.3390/cancers10110398](https://doi.org/10.3390/cancers10110398).
- 59.** Pearce S, Brownsdon A, Fern L, Gibson F, Whelan J, Lavender V. The perceptions of teenagers, young adults and professionals in the participation of bone cancer clinical trials. *European Journal of Cancer Care*, **2018**; 27 (6), Special Issue. <https://doi.org/10.1111/ecc.12476>.
- 58.** Chalopin A, Tellez-Gabriel M, Brown HK, Vallette F, Heymann MF, Gouin F, Heymann D. Isolation of circulating tumor cells in a preclinical model of osteosarcoma: Effect of chemotherapy. *Journal of Bone Oncology*, **2018**; 12, 83-90. DOI: [10.1016/j.jbo.2018.07.002](https://doi.org/10.1016/j.jbo.2018.07.002).

- 57.** Jacques C, Renema N, Lezot F, Ory B, Walkley CR, Grigoriadis AE, Heymann, D. Small animal models for the study of bone sarcoma pathogenesis: characteristics, therapeutic interests and limitations. *Journal of Bone Oncology*, **2018**; 12, 7-13. DOI: [10.1016/j.jbo.2018.02.004](https://doi.org/10.1016/j.jbo.2018.02.004).
- 56.** Strauss SJ, Anninga J, Baglio R, Baumhoer D, Behjati S, Bielack S, et al. Report from the 4th European Bone Sarcoma Networking meeting: focus on osteosarcoma. *Clinical Sarcoma Research*, **2018**; 8, Article Number:17. <https://doi.org/10.1186/s13569-018-0103-0>.
- 55.** Holme H, Gulati A, Brough R, Fleuren EDG, Bajrami I, Campbell J, et al. Chemosensitivity profiling of osteosarcoma tumour cell lines identifies a model of BRCAness. *Scientific Reports*, **2018**; 8, Article Number:10614. <https://doi.org/10.1038/s41598-018-29043-z>.
- 54.** Bosch MC, Roundhill EA, Droop AP, Parry M, Jeys L, Burchill SA. RNAseq of patient-derived cancer stem-like cells and exosomes provides new insights into Ewing's sarcoma. *Cancer Research*, **2018**; Meeting Abstract:3696, 78 (13). <https://doi.org/10.1158/1538-7445.AM2018-3696>.
- 53.** Zhang T, Kastrenopoulou A, Larrouture Q, Athanasou NA, Knowles HJ. Angiopoietin-like 4 promotes osteosarcoma cell proliferation and migration and stimulates osteoclastogenesis. *BMC Cancer*, **2018**; 18, Article Number:536. DOI: [10.1186/s12885-018-4468-5](https://doi.org/10.1186/s12885-018-4468-5).
- 52.** de Ridder D, Marino S, Bishop RT, Renema N, Chenu C, Heymann D, Idris AI. Bidirectional regulation of bone formation by exogenous and osteosarcoma-derived Sema3A. *Scientific Reports*, **2018**; 8, Article Number:6877. DOI: [10.1038/s41598-018-25290-2](https://doi.org/10.1038/s41598-018-25290-2).
- 51.** Koster R, Panagiotou OA, Wheeler WA, Karlins E, Gastier-Foster JM, de Toledo SRC, et al. Genome-wide association study identifies the GLDC/IL33 locus associated with survival of osteosarcoma patients. *International Journal of Cancer*, **2018**; 142 (8), 1594-1601. DOI: [10.1002/ijc.31195](https://doi.org/10.1002/ijc.31195).
- 50.** Blakey K, Feltbower RG, James PW, Libby G, Stiller C, Norman P, et al. Socio-economic patterning in early mortality of patients aged 0-49 years diagnosed with primary bone cancer in Great Britain, 1985-2008. *Cancer Epidemiology*, **2018**; 53, 49-55. DOI: [10.1016/j.canep.2018.01.012](https://doi.org/10.1016/j.canep.2018.01.012).
- 49.** Brown HK, Schiavone K, Gouin F, Heymann MF, Heymann D. Biology of Bone Sarcomas and New Therapeutic Developments. *Calcified Tissue International*, **2018**; 102 (2), Special Issue, 174-195. DOI: [10.1007/s00223-017-0372-2](https://doi.org/10.1007/s00223-017-0372-2).
- 48.** Gonzalez-Fernandez Y, Brown HK, Patino-Garcia A, Heymann D, Blanco-Prieto MJ. Oral administration of edelfosine encapsulated lipid nanoparticles causes regression of lung metastases in pre-clinical models of osteosarcoma. *Cancer Letters*, **2018**; 430, 193-200. DOI: [10.1016/j.canlet.2018.05.030](https://doi.org/10.1016/j.canlet.2018.05.030).
- 47.** Vormoor B, Schlosser YT, Blair H, Sharma A, Wilkinson S, Newell DR, Curtin, N. Sensitizing Ewing sarcoma to chemo-and radiotherapy by inhibition of the DNA-repair enzymes DNA protein kinase (DNA-PK) and poly-ADP-ribosepolymerase (PARP) 1/2. *Oncotarget* **2017**; 8 (69), 113418-113430. <https://doi.org/10.1863/oncotarget.21300>.
- 46.** Redondo PA, Pavlou M, Loizidou M, Cheema U. Elements of the niche for adult stem cell expansion. *Journal of Tissue Engineering*, **2017**; 8, Article Number:2041731417725464. DOI: [10.1177/2041731417725464](https://doi.org/10.1177/2041731417725464).
- 45.** Behjati S, Tarpey PS, Haase K, Ye HT, Young MD, Alexandrov LB, et al. Recurrent mutation of IGF signalling genes and distinct patterns of genomic rearrangement in osteosarcoma. *Nature Communications*, **2017**; 8, Article Number:15936. DOI: [10.1038/ncomms15936](https://doi.org/10.1038/ncomms15936).

- 44.** Knowles HJ. Multiple Roles of Angiopoietin-Like 4 in Osteolytic Disease. *Frontiers in Endocrinology*, **2017**; 8, Article Number:80. DOI: [10.3389/fendo.2017.00080](https://doi.org/10.3389/fendo.2017.00080).
- 43.** Tellez-Gabriel M, Charrier C, Brounais-Le Royer B, Mullard, M, Brown HK, Verrecchia F, Heymann D. Analysis of gap junctional intercellular communications using a dielectrophoresis-based microchip. *European Journal of Cell Biology*, **2017**; 96 (2), 110-118. DOI: [10.1016/j.ejcb.2017.01.003](https://doi.org/10.1016/j.ejcb.2017.01.003).
- 42.** Brown HK, Tellez-Gabriel M, Heymann D. Cancer stem cells in osteosarcoma. *Cancer Letters*, **2017**; 386, 189-195. DOI: [10.1016/j.canlet.2016.11.019](https://doi.org/10.1016/j.canlet.2016.11.019).
- 41.** Brown HK, Schiavone K, Tazzyman S, Heymann D, ChicoTJA. Zebrafish xenograft models of cancer and metastasis for drug discovery. *Expert Opinion on Drug Discovery*, **2017**; 12 (4), 379-389. DOI: [10.1080/17460441.2017.1297416](https://doi.org/10.1080/17460441.2017.1297416).
- 40.** Tellez-Gabriel M, Ory B, Lamoureux F, Heymann MF, Heymann D. Tumour Heterogeneity: The Key Advantages of Single-Cell Analysis. *International Journal of Molecular Sciences*, **2016**; 17 (12), Article Number:2142. DOI: [10.3390/ijms17122142](https://doi.org/10.3390/ijms17122142).
- 39.** Dumars C, Ngyuen JM, Gaultier A, Lanel R, Corradini N, Gouin F, *et al.* Dysregulation of macrophage polarization is associated with the metastatic process in osteosarcoma. *Oncotarget*, **2016**; 7 (48), 78343-78354. DOI: [10.18632/oncotarget.13055](https://doi.org/10.18632/oncotarget.13055).
- 38.** Heymann MF, Brown HK, Heymann D. Drugs in early clinical development for the treatment of osteosarcoma. *Expert Opinion on Investigational Drugs*, **2016**; 25 (11), 1265-1280. DOI: [10.1080/13543784.2016.1237503](https://doi.org/10.1080/13543784.2016.1237503).
- 37.** Tellez-Gabriel M, Brown HK, Young R, Heymann MF, Heymann D. The Challenges of Detecting Circulating Tumor Cells in Sarcoma. *Frontiers in Oncology*, **2016**; 6, Article Number:202. DOI: [10.3389/fonc.2016.00202](https://doi.org/10.3389/fonc.2016.00202).
- 36.** Inagaki Y, Hookway ES, Kashima TG, Munemoto M, Tanaka Y, Hassan AB, *et al.* Sclerostin expression in bone tumours and tumour-like lesions. *Histopathology*, **2016**; 69 (3), 470-478. DOI: [10.1111/his.12953](https://doi.org/10.1111/his.12953).
- 35.** Behjati S, Gundem G, Wedge DC, Roberts ND, Tarpey PS, Cooke SL, *et al.* Mutational signatures of ionizing radiation in second malignancies. *Nature Communications*, **2016**; 7, Article Number:12605. DOI: [10.1038/ncomms12605](https://doi.org/10.1038/ncomms12605).
- 34.** Inagaki Y, Hookwa E, Williams KA, Hassan AB, Oppermann U, Tanak Y, *et al.* Dendritic and mast cell involvement in the inflammatory response to primary malignant bone tumours. *Clinical Sarcoma Research*, **2016**; 6, Article Number:13. DOI: [10.1186/s13569-016-0053-3](https://doi.org/10.1186/s13569-016-0053-3).
- 33.** Sabokbar A, Mahoney DJ, Hemingway F, Athanasou NA. Non-Canonical (RANKL-Independent) Pathways of Osteoclast Differentiation and Their Role in Musculoskeletal Diseases. *Clinical Reviews in Allergy and Immunology*, **2016**; 51 (1), 16-26. DOI: [10.1007/s12016-015-8523-6](https://doi.org/10.1007/s12016-015-8523-6).
- 32.** Johansson C, Velupillai S, Tumber A, Szykowska A, Hookway ES, Nowak RP, *et al.* Structural analysis of human KDM5B guides histone demethylase inhibitor development. *Nature Chemical Biology*, **2016**; 12 (7), 539-545. DOI: [10.1038/nchembio.2087](https://doi.org/10.1038/nchembio.2087).
- 31.** Weekes D, Kashima TG, Zandueta C, Perurena N, Thomas DP, Sunters, A, *et al.* Regulation of osteosarcoma cell lung metastasis by the c-Fos/AP-1 target FGFR1. *Oncogene*, **2016**; 35 (22), 2852-2861. DOI: [10.1038/onc.2015.344](https://doi.org/10.1038/onc.2015.344).
- 30.** McCarthy C, Anderson WJ, Vlychou M, Inagaki Y, Whitwell D, Gibbons CL, Athanasou NA. Primary synovial chondromatosis: a reassessment of malignant potential in 155 cases. *Skeletal Radiology*, **2016**; 45 (6), 755-762. DOI: [10.1007/s00256-016-2353-3](https://doi.org/10.1007/s00256-016-2353-3).

- 29.** Mirabello L, Koster R, Moriarity BS, Spector LG, Meltzer PS, Gary, *et al.* Genome-Wide Scan Identifies Variants in NF1B Associated with Metastasis in Patients with Osteosarcoma. *Cancer Discovery*, **2015**; 5 (9), 920-931. DOI: [10.1158/2159-8290.CD-15-0125](https://doi.org/10.1158/2159-8290.CD-15-0125).
- 28.** Bhatia C, Oerum S, Bray J, Kavanagh KL, Shafqat N, Yue W, Oppermann U. Towards a systematic analysis of human short-chain dehydrogenases/reductases (SDR): Ligand identification and structure-activity relationships. *Chemico-Biological Interactions*, **2015**; 234, Special Issue, 114-125. DOI: [10.1016/j.cbi.2014.12.013](https://doi.org/10.1016/j.cbi.2014.12.013).
- 27.** Inagaki Y, Kashima TG, Hookway ES, Tanaka Y, Hassan AB, Oppermann U, Athanasou NA. Dentine matrix protein 1 (DMP-1) is a marker of bone formation and mineralisation in soft tissue tumours. *Virchows Archiv*, **2015**; 466 (4), 445-452. DOI: [10.1007/s00428-014-1706-3](https://doi.org/10.1007/s00428-014-1706-3).
- 26.** Alholle A, Brini AT, Bauer J, Gharanei S, Niad S, Slater A, *et al.* Genome-wide DNA methylation profiling of recurrent and non-recurrent chordomas. *Epigenetics*, **2015**; 10 (3), 213-220. DOI: [10.1080/15592294.2015.1006497](https://doi.org/10.1080/15592294.2015.1006497).
- 25.** Cheng X, Hookway ES, Kashima T, Oppermann U, Galione A, Athanasou NA. The Role of Calcium and Nicotinic Acid Adenine Dinucleotide Phosphate (NAADP) in Human Osteoclast Formation and Resorption. *Calcified Tissue International*, **2015**; 96 (1), 73-79. DOI: [10.1007/s00223-014-9939-3](https://doi.org/10.1007/s00223-014-9939-3).
- 24.** Guilhamon P, Butcher LM, Presneau N, Wilson GA, Feber A, Paul DS, *et al.* Assessment of patient-derived tumour xenografts (PDXs) as a discovery tool for cancer epigenomics. *Genome Medicine*, **2014**; 6 (12), Article Number:116. DOI: [10.1186/s13073-014-0116-0](https://doi.org/10.1186/s13073-014-0116-0).
- 23.** Brownhill S, Cohen D, Burchill S. Proliferation Index: A Continuous Model to Predict Prognosis in Patients with Tumours of the Ewing's Sarcoma Family. *PLOS ONE*, **2014**; 9 (8), Article Number: e104106. <https://doi.org/10.1371/journal.pone.0104106>.
- 22.** Amary MF, Ye HT, Berisha F, Khatri B, Forbes G, Lehouky K, *et al.* Fibroblastic growth factor receptor 1 amplification in osteosarcoma is associated with poor response to neo-adjuvant chemotherapy. *Cancer Medicine*, **2014**; 3 (4), 980-987. <https://doi.org/10.1002/cam4.268>.
- 21.** Vormoor B, Curtin NJ. Poly (ADP-ribose) polymerase inhibitors in Ewing sarcoma. *Current Opinion in Oncology*, **2014**; 26 (4), 428-433. DOI: [10.1097/CCO.0000000000000091](https://doi.org/10.1097/CCO.0000000000000091).
- 20.** Blakey K, Feltbower RG, Parslow RC, James PW, Pozo BG, Stiller C, *et al.* Is fluoride a risk factor for bone cancer? Small area analysis of osteosarcoma and Ewing sarcoma diagnosed among 0-49-year-olds in Great Britain, 1980-2005. *International Journal of Epidemiology*, **2014**; 43 (1), 224-234. DOI: [10.1093/ije/dyt259](https://doi.org/10.1093/ije/dyt259).
- 19.** Vormoor B, Knizia HK, Batey MA, Almeida GS, Wilson I, Dilley P, *et al.* Development of a Preclinical Orthotopic Xenograft Model of Ewing Sarcoma and Other Human Malignant Bone Disease Using Advanced In Vivo Imaging. *PLOS ONE*, **2014**; 9 (1), Article Number: e85128. DOI: [10.1371/journal.pone.0085128](https://doi.org/10.1371/journal.pone.0085128).
- 18.** Alholle A, Brini AT, Gharanei S, Vaiyapuri S, Arrigoni E, Dallol A, *et al.* Functional epigenetic approach identifies frequently methylated genes in Ewing sarcoma *Epigenetics*, **2013**; 8 (11), 1198-1204. <https://doi.org/10.4161/epi.26266>.
- 17.** Jeys L, Matharu GS, Nandra RS, Grimer, RJ. Can computer navigation-assisted surgery reduce the risk of an intralesional margin and reduce the rate of local recurrence in patients with a tumour of the pelvis or sacrum? *Bone & Joint Journal*, **2013**; 95B (10), 1417-1424. DOI: [10.1302/0301-620X.95B10.31734](https://doi.org/10.1302/0301-620X.95B10.31734).
- 16.** Gharanei S, Brini AT, Vaiyapuri S, Alholle A, Dallol A, Arrigoni E, *et al.* RASSF2 methylation is a strong prognostic marker in younger age patients with Ewing sarcoma. *Epigenetics*, **2013**; 8 (9), 893-898. DOI: [10.4161/epi.25617](https://doi.org/10.4161/epi.25617).

- 15.** Janowitz T, Welsh SJ, Zaki K, Mulders P, Eisen T. Adjuvant Therapy in Renal Cell Carcinoma-Past, Present, and Future. *Seminars in Oncology*, **2013**; 40 (4), 482-491. DOI: [10.1053/j.seminoncol.2013.05.004](https://doi.org/10.1053/j.seminoncol.2013.05.004).
- 14.** Savage SA, Mirabello L, Wang ZM, Gastier-Foster JM, Gorlick R, Khanna C, *et al.* Genome-wide association study identifies two susceptibility loci for osteosarcoma. *Nature Genetics*, **2013**; 45 (7), 799-803. <https://doi.org/10.1038/ng.2645>.
- 13.** Lombardo CM, Welsh SJ, Strauss SJ, Dale AG, Todd AK, Nanjunda R, *et al.* A novel series of G-quadruplex ligands with selectivity for HIF-expressing osteosarcoma and renal cancer cell lines. *Bioorganic & Medicinal Chemistry Letters*, **2012**; 22 (18), 5984-5988. DOI: [10.1016/j.bmcl.2012.07.009](https://doi.org/10.1016/j.bmcl.2012.07.009).
- 12.** McNally RJQ, Blakey K, Parslow RC, James PW, Pozo BG, Stiller C, *et al.* Small-area analyses of bone cancer diagnosed in Great Britain provide clues to aetiology. *BMC Cancer*, **2012**; 12, Article Number: 270. <https://doi.org/10.1186/1471-2407-12-270>.
- 11.** Windsor RE, Strauss SJ, Kallis C, Wood NE, Whelan JS. Germline Genetic Polymorphisms May Influence Chemotherapy Response and Disease Outcome in Osteosarcoma. *Cancer*, **2012**; 118 (7), 1856-1867. DOI: [10.1002/cncr.26472](https://doi.org/10.1002/cncr.26472).
- 10.** Duhamel LAE, Ye HT, Halai D, Idowu BD, Presneau N, Tirabosco R, Flanagan AM. Frequency of Mouse Double Minute 2 (MDM2) and Mouse Double Minute 4 (MDM4) amplification in parosteal and conventional osteosarcoma subtypes. *Histopathology*, **2012**; 60 (2), 357-359. DOI: [10.1111/j.1365-2559.2011.04023.x](https://doi.org/10.1111/j.1365-2559.2011.04023.x).
- 9.** Amary MF, Damato S, Halai D, Eskandarpour M, Berisha F, Bonar F, *et al.* Ollier disease and Maffucci syndrome are caused by somatic mosaic mutations of IDH1 and IDH2. *Nature Genetics*, **2011**; 43 (12), 1262-U129. DOI: [10.1038/ng.994](https://doi.org/10.1038/ng.994).
- 8.** Kunita A, Kashima TG, Ohazama A, Grigoriadis AE, Fukayama M. Podoplanin Is Regulated by AP-1 and Promotes Platelet Aggregation and Cell Migration in Osteosarcoma. *American Journal of Pathology*, **2011**; 179 (2), 1041-1049. DOI: [10.1016/j.ajpath.2011.04.027](https://doi.org/10.1016/j.ajpath.2011.04.027).
- 7.** Fujiwara M, Kashima TG, Kunita A, Kii, I, Komura D, Grigoriadis AE, *et al.* Stable knockdown of S100A4 suppresses cell migration and metastasis of osteosarcoma. *Tumor Biology*, **2011**; 32 (3), 611-622. DOI: [10.1007/s13277-011-0160-y](https://doi.org/10.1007/s13277-011-0160-y).
- 6.** Knowles HJ, Schaefer KL, Dirksen U, Athanasou NA. Hypoxia and hypoglycaemia in Ewing's sarcoma and osteosarcoma: regulation and phenotypic effects of Hypoxia-Inducible Factor. *BMC Cancer*, **2010**; 10, Article Number:372. <http://www.biomedcentral.com/1471-2407/10/372>.
- 5.** Eyre R, Feltbower RG, James PW, Blakey K, Mbwandrikwa E, Forman D, *et al.* The epidemiology of bone cancer in 0-39 year olds in northern England, 1981-2002. *BMC Cancer*, **2010**; 10, Article Number:357. DOI: [10.1186/1471-2407-10-357](https://doi.org/10.1186/1471-2407-10-357).
- 4.** Facey K, Boivin A, Gracia J, Hansen HP, Lo Scalzo A, Mossman J, Single A. Patients' perspectives in health technology assessment: A route to robust evidence and fair deliberation. *International Journal of Technology Assessment in Health Care*, **2010**; 26 (3), 334-340. DOI: [10.1017/S0266462310000395](https://doi.org/10.1017/S0266462310000395).
- 3.** Roelofs AJ, Thompson K, Ebetino FH, Rogers MJ, Coxon FP. Bisphosphonates: Molecular Mechanisms of Action and Effects on Bone Cells, Monocytes and Macrophages. *Current Pharmaceutical Design*, **2010**; 16, (27), 2950-2960. DOI: [10.2174/138161210793563635](https://doi.org/10.2174/138161210793563635).
- 2.** Eyre R, Feltbower RG, Mbwandrikwa E, EdenTOB, McNally RJQ. Epidemiology of Bone Tumours in Children and Young Adults. *Pediatric Blood & Cancer*, **2009**; 53 (6), 941-952. DOI: [10.1002/pbc.22194](https://doi.org/10.1002/pbc.22194).
- 1.** Eyre R, Feltbower RG, Mbwandrikwa E, Jenkinson HC, Parkes S, Birch JM, *et al.* Incidence and survival of childhood bone cancer in northern England and the West

Midlands, 1981-2002. *British Journal of Cancer*, **2009**; 100 (1), 188-193.
DOI: [10.1038/sj.bjc.6604837](https://doi.org/10.1038/sj.bjc.6604837).

Book chapters

Luke Tattersall, Zoe Davison, Alison Gartland

Osteosarcoma. Encyclopedia of Bone Biology, Elsevier, Reference Module in Biomedical Sciences, **2020**; 362-378. DOI:[10.1016/B978-0-12-801238-3.62259-6](https://doi.org/10.1016/B978-0-12-801238-3.62259-6).

Camille Jacques, Nathalie Renema, Benjamin Ory, Carl R. Walkley, Agamemnon E. Grigoriadis, Dominique Heymann, Idris A.

Murine Models of Bone Sarcomas. (eds) Bone Research Protocols. Methods in Molecular Biology, vol 1914. Humana Press, New York, NY. **2019**, p. 331-342. DOI: [10.1007/978-1-4939-8997-3_18](https://doi.org/10.1007/978-1-4939-8997-3_18).