

## Here, There and Nowhere: Following Adult Survivors of Childhood Cancer A Case Report of Recurrent Osteosarcoma in a Young Adult

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### ABSTRACT

Approximately 1 in 285 children in the United States (US) will be diagnosed with cancer before the age of 20.<sup>1</sup> More than 80% of children diagnosed with cancer will become long-term survivors.<sup>2</sup> As of January, 2010, there are more than 380,000 adult survivors of childhood cancer in the US.<sup>3</sup> More than two-thirds of survivors will develop chronic conditions.<sup>4</sup> Professional organizations have advocated for specialized risk-based care of survivors.<sup>5</sup> Locally and nationally, lack of transition services and insurance coverage are barriers to care of these adult survivors.<sup>6</sup> We describe one such case to illustrate these challenges and their impact.

**KEYWORDS:** cancer survivorship, healthcare transition, adult survivors of childhood cancer, emerging adults

### CASE REPORT

A 15-year-old boy was diagnosed in November, 2005, with osteosarcoma of the right distal femur. He underwent two cycles of chemotherapy in his home country, the Dominican Republic, and travelled to Rhode Island (RI) for surgery and further treatment. He continued treatment in RI according to Pediatric Oncology Group (POG) protocol 9754, undergoing 2 cycles of chemotherapy, an amputation to mid-femur with rotationplasty of the lower extremity and 4 additional cycles of adjuvant chemotherapy completed in October, 2006. His treatment was complicated by typhlitis, febrile neutropenia and hemorrhagic cystitis. He was discharged from his final treatment with follow-up with the brace shop, orthopedics, physical therapy and hematology/oncology, and his port was removed 5/2007. He reported having no primary care physician. He was not seen in oncology follow-up from 11/2008 at the time of a negative CT scan for metastatic disease, until re-presenting in 8/2010 with report of skin breakdown at his prosthesis site, fever, pain, swelling and drainage from his right great toe. He received podiatric care and screening x-rays and echocardiogram for late-effects, but no additional follow-up. He re-presented 3/2011 to an oncologist, reporting a 3-day history of facial pain and swelling, with concern for tooth abscess. He next presented to the Emergency Department (ED) in 11/2011, for pain and swelling of his right leg stump. An ultrasound showed deep

venous thrombosis of the femoral vein, and he underwent drainage of a paronychia of his right great toe. He received cephalexin, warfarin and low dose molecular heparin in the ED but reported being able only to purchase the warfarin due to cost. He presented 5 days later to the pediatric oncology clinic. Social work was involved to facilitate insurance coverage, medications, follow-up with orthopedics and podiatry, audiology for previously recommended hearing aids and primary care visits. He was scheduled for follow-up with the late effects clinic, and seen in Transition Consultation Clinic in 11/2011. Routine screening indicated learning disabilities, poor nutrition, limited physical activity, clinical depression and overweight. He lacked insurance coverage, a functioning prosthesis and a vocational plan. His pain and swelling were attributed to thrombosis and infection, but failed to improve over the following 4 weeks. He denied fever or night sweats, but endorsed more focal right thigh pain, fatigue and daily vomiting with an unintended 30 pound weight loss in the two weeks preceding the visit. He underwent a bone scan at a survivorship clinic visit 1/2012 for possible osteomyelitis, given persistent erythema of the right great toe and an elevated white blood cell count. The scan indicated "a large, round, markedly hypervascular mass in the right thigh, predominantly extraosseous in appearance with more focal intense areas of uptake within the soft tissues of the right side with possible involvement of the right proximal femur in the area of internal fixation hardware, highly suspicious for recurrence of osteosarcoma."

The patient was admitted for a CT-guided biopsy of the right thigh mass, which confirmed recurrent osteosarcoma. He underwent a right hip disarticulation. With no evidence for metastatic disease at that time, no adjuvant chemotherapy was pursued. However, subsequent 5/2012 surveillance chest CT demonstrated pulmonary nodules consistent with metastatic disease. The patient was treated with neoadjuvant chemotherapy consisting of methotrexate, doxorubicin and cisplatin, with subsequent decreased size of nodules and then underwent a right thoracotomy with wedge resection of metastatic nodules in the right middle and lower lobes in 7/2012. This was followed with an individualized adjuvant chemotherapy protocol, including methotrexate, doxorubicin, carboplatin, cyclophosphamide, etoposide and zoledronic acid completed in 12/2012. He next presented to the survivorship clinic in 8/2014, reporting cough and intermittent chest pain, after a period of lost-to-follow-up. A chest

CT and bone scan did not show new disease and his echocardiogram was normal. He was again advised to establish primary care. He presented to primary care in 9/2014, and returned for a health maintenance visits 12/2014 and 1/2016. At his most recent well adult visit it was noted that he had not had interim care with the survivorship program, and was encouraged to follow-up. He presented for survivorship follow-up 1/2016, and had negative surveillance imaging and a normal echocardiogram.

## DISCUSSION

Our patient experienced multiple secondary effects of chemotherapy, including hearing loss, learning disabilities and infertility. In addition, he experienced recurrence of his osteosarcoma. Adherence to routine monitoring and care coordination might have mitigated some of these conditions and allowed for earlier detection. Importantly, adherence to recommended off-therapy tumor surveillance imaging may have detected this patient's tumor recurrence earlier. Early detection followed by early treatment of relapsed disease may have also decreased this patient's risk for pulmonary metastatic disease, for which he required a substantial amount of additional chemotherapy. In the past 35 years, the 5-year survival rate for localized osteosarcoma improved from 40-76% for children <15 years, and from 56-66% in adolescents ages 15–19 years.<sup>7</sup>

Our patient sought care primarily to evaluate acute conditions, and engaged in primary preventive and survivor-related follow-up only when prompted by other providers. Among the more than 10,000 adult survivors in the Childhood Cancer Survivor Study (CCSS), more than 62% reported one or more chronic conditions.<sup>8</sup> However, although 87% reported general medical contact and 71% a general physical exam within the prior 2 years, only 19% had a visit at a cancer center.<sup>9</sup> Thus, the majority of adult survivors of childhood cancer are getting care from providers who are not oncologists. Encouraging routine care and developing and maintaining an awareness of secondary effects and screening recommendations are important steps for primary care providers. Yet, in a survey of internists, only 12% stated that they felt at least "somewhat familiar" with available guidelines.<sup>10</sup> The Children's Oncology Group's Long-Term Follow-Up Guidelines for Survivors of Childhood, Adolescent, and Young Adult Cancers summarizes secondary effects of treatments, surveillance recommendations, and provides sample treatment summaries. <http://www.survivorshipguidelines.org/>.<sup>11</sup>

Our patient identified cost of care and lack of insurance as significant barriers. Uninsured survivors in the CCSS cohort had lower utilization of both survivor-focused and general preventive health care than privately or publicly insured survivors.<sup>12</sup> Fewer than 14% and 10% of survivors, respectively, have ever been enrolled in the supplemental security income (SSI) or social security disability insurance (SSDI) federal disability programs providing income and insurance

support for disabled adults.<sup>13</sup> The 2010 Patient Protection and Affordable Care Act (ACA) established provisions intended to increase access to affordable health insurance, including requiring employer-sponsored health insurance plans to cover enrolled members' children up to age 26 and limiting exclusions for pre-existing conditions. During the first full two years following the passage of the ACA, young adults (YA) ages 19–25 per capita health care spending grew at a rate nearly double that for the non-YA adult population.<sup>14</sup> However, in a 2011–12 survey of survivors, only 27% reported familiarity with the ACA.<sup>15</sup> And most recently, a comparison of adult childhood cancer survivors to adults without cancer found fewer CCS were insured (76% vs 81%), and were more likely to report delaying medical care (25% vs 13%) and to report needing but not getting medical care in the previous 12 months (20% vs 10%).<sup>16</sup> A regional review of all of the academic pediatric oncology programs in New England found that funding for resource intense programs and transitioning care to adult clinical services were common challenges.<sup>17</sup>

Hasbro Children's Hospital's Comprehensive Health Assessment and Management for Pediatric Cancer Survivors (CHAMPS) Program is a local resource that includes monitoring for late-effects, as well as social work support that can assist patients seeking coverage and resources: <http://www.hasbrochildrenshospital.org/Programs.html>.

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