Basal Cell Carcinoma Arising 57 Years after Interstitial Radiotherapy of a Nasal Hemangioma

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Cutaneous hemangiomas are benign vascular tumors that represent the most common benign tumor of infancy.[1] These tumors characteristically have an early phase of rapid proliferation followed by a slower regression phase; over 90% of cutaneous hemangiomas will regress completely by age 10.[2–4] Irradiation of infantile hemangiomas was commonly performed in the 1950s and 1960s, but this treatment was largely abandoned after the long-term risks of radiotherapy and the benign nature of most hemangiomas were appreciated.

Basal cell carcinoma (BCC) is the most common type of skin cancer in fair-skinned individuals, and BCC arising within prior radiation fields has been widely reported.[5,6] BCC occurring specifically at the site of a previously irradiated hemangioma is uncommon but has been reported. We present a rare case of an infiltrating BCC occurring adjacent to a spent radon seed that had been inserted into the nose of a female patient 57 years earlier for interstitial radiotherapy of a nasal hemangioma.

Case Report

A 57-year-old female presented to clinic for Mohs micrographic surgery (MMS) of an infiltrative BCC at the right nasal tip. At 6 months of age, the patient received interstitial radiotherapy for a hemangioma on her nose via implantation of two radon-222 seeds into her right nasal ala. The nasal hemangioma subsequently regressed, and for decades she reported only cold sensitivity at the right nostril that was attributed to the metal casings of the seeds. At age 30, while employed by a hospital radiology department, Geiger counter measurements failed to detect any remaining radiation at her nasal tip. Four years later, she developed some temporary scabbing at the implant site that healed with a slight indentation. Around age 50, a radon seed began to extrude from the surface, and the patient subsequently removed the seed herself. Removal of the extruding seed left a small "canal" shaped defect at the right nasal rim. Two years prior to her presentation for MMS, she began experiencing intermittent bleeding and scabbing in the right nostril, and the "canal" at her right nasal rim began to enlarge and change in appearance. One year later during a consultation with plastic surgery for nasal reconstruction, a biopsy of the right nasal tip revealed a BCC. Because of the tumor's large size, ill-defined borders, and location at a site of previous radiation, MMS followed by reconstructive plastic surgery was planned.

As seen in the preoperative photo (Figure 1), a full-thickness defect was present at the right nasal rim and right nasal tip; ulceration of the adjacent nasal

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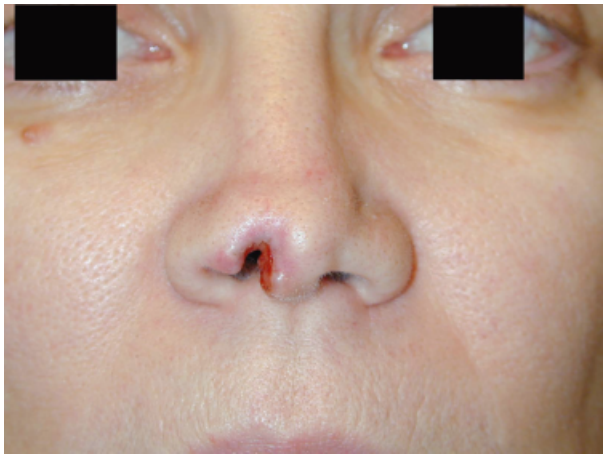

Figure 1. Preoperative photo demonstrating clinical extent of tumor.

and septal mucosa was apparent. A five-stage MMS excision was required to obtain tumor-free deep and lateral margins, and the remaining radon seed was excised with tumor (Figure 2). The additional stages were necessary due to peripheral and deep extension of the tumor through the nasal ala and into the nasal septum. A full-thickness, triangular surgical defect involving the right dorsal nose, nasal tip, and nasal ala/rim measured 1.8×2.0 cm (Figure 3A). The five-stage surgery also involved partial excision of the right septal mucosa.

Plastic surgical reconstruction of the large cutaneous defect utilized a paramedian forehead flap combined with an intranasal mucosal flap to provide a functional nasal lining for the surgical defect (Figure 3B). The patient recovered without complications; 1-year postoperative photos demonstrated a good cosmetic and functional outcome (Figure 3C).

Figure 2. Spent radon-222 seed excised with tumor.

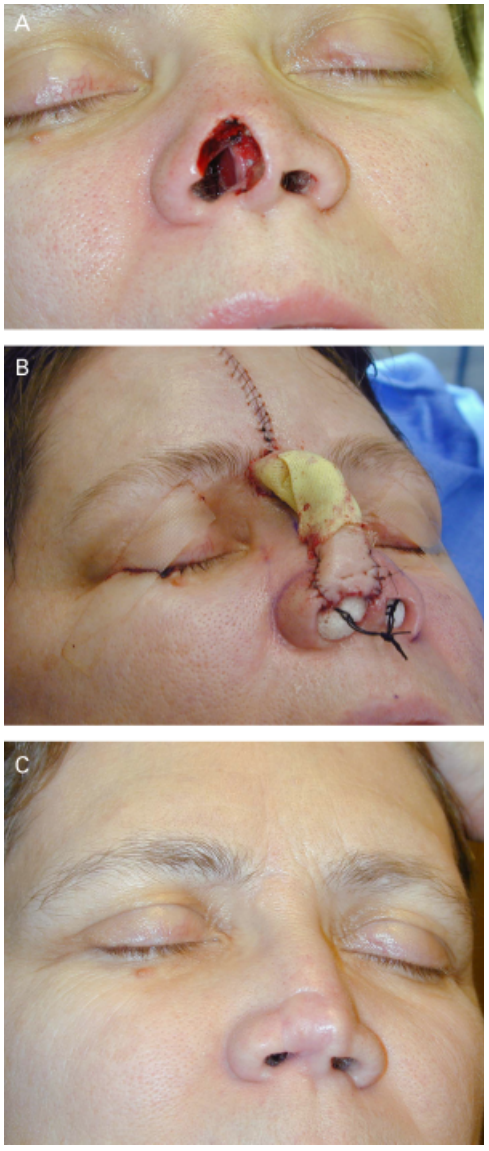

Figure 3. (A) Postoperative Mohs defect involving right dorsal nose, nasal tip, nasal ala, and septal mucosa. (B) Postoperative paramedian forehead flap and intranasal mucosal flap repair of Mohs defect. (C) One-year postoperative photo demonstrating good cosmetic and functional outcome.

Discussion

Radon and its decay products are known human carcinogens; numerous epidemiologic studies have

demonstrated increased incidences primarily of lung, tracheal, and nasal cancer among chronically exposed humans.[7] Radon occurs naturally by radioactive decay of radium and uranium. The greatest human exposure to radon is through environmental inhalation or ingestion of contaminated groundwater.[7] Ionizing radiation from radon containing gold seeds and radium-containing tubes, sheets, and needles was routinely used to treat a variety of benign and malignant tumors and skin conditions until the early 1970s.[8]

Throughout the 1950s and 1960s, ionizing radiation was commonly used to successfully treat cutaneous hemangiomas in children.[8,9] One popular treatment method entailed inserting radon seeds into the skin and leaving them in place indefinitely. These gold seeds contained radon-222 (alpha particle emitter, $t_{1/2} = 3.8$ days), and most useful radiation from these radon seeds lasted only 1 to 8 weeks.[10] As physicians began appreciating both the potential long-term risks of radiotherapy and the fact that most hemangiomas regressed spontaneously, routine radiotherapy of hemangiomas became much less common. However, radiation therapy is still considered an indicated treatment for life- or function-threatening hemangiomas not responsive to corticosteroids or other treatments.[1] A review of 153 reported cases of Kasabach-Merritt syndrome concluded that radiation plus steroid therapy was superior to surgical, steroid, or radiation treatment alone.[11]

Radiation-induced malignancies are a well-documented potential adverse effect of radiotherapy. A number of long-term studies have revealed a significantly increased rate of various malignancies, including BCCs, in patients who underwent radiotherapy for hemangiomas.[8,9,12,13] As well, numerous case reports have documented BCCs arising at sites of previously irradiated tinea capitis,[14] port-wine stains,[15,16] and hemangiomas.[17,18] In these reports, BCCs typically arose 10 to 50 years after low-dose irradiation of the skin. In addition, delayed skin ulceration and extrusion of spent radon seeds have been reported to occur as late as 20 to 40 years after insertion.[10,19,20] Our patient experienced delayed extrusion of a spent radon seed 51 years after initial implantation.

MMS is indicated for BCC arising within an area of prior radiotherapy. Since tumor borders are often clinically ill-defined due to fibrosis and other skin changes resulting from radiotherapy, the MMS technique of complete margin assessment offers the highest cure rate for these high-risk tumors.[21–23] In our patient, the need for a five-stage excision to obtain clear lateral and deep margins illustrates the fundamental importance of full margin visualization in such cases.

We report an interesting case of an infiltrating BCC occurring at a site of radon seed implantation 57 years earlier. A five-stage MMS excision provided tumor-free margins, and a paramedian forehead flap and intranasal mucosal flap reconstruction produced a good functional and cosmetic outcome for our patient. The colocalization of the radon seeds with the tumor suggests that this skin cancer represented a late adverse effect of radon seed treatment of a nasal hemangioma. Patients with a history of radiotherapy require lifelong monitoring for development of radiation-induced neoplasms. Physicians caring for these patients should have a low threshold to biopsy any irregular or unexplained skin change or any nonhealing erosion at sites of previous radiation therapy.

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