



Case report: chronic inflammatory ulcer and osteoradionecrosis of the skull following radiotherapy in early childhood

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Abstract

Introduction Chronic inflammatory diseases of the skin are the most common differential diagnosis of tumorous lesions of the craniofacial region. Detailed information about a patient's medical history is important for the clinical diagnosis of such cases. Previous radiotherapy should be taken into account, especially in cases of chronic dermatitis, since complications include osteoradionecrosis of the adjacent bone structures with surrounding inflammation.

Case Report We present the case of a 77-year-old female patient who was admitted to our department with a slightly progressive ulcerating lesion of the frontotemporal skull. The patient had received radiotherapy in early childhood as primary therapy for hemangioma. Diagnostic imaging and biopsies revealed a diagnosis of chronic ulceration with underlying osteonecrosis and fibrotic osteomyelitis of the skull. A complex reconstruction of osseous structures and soft tissue was necessary to resolve her complaints.

Conclusion Chronic radiodermatitis and osteoradionecrosis in adults, occurring as late complications, are uncommon, but can be observed even nearly 80 years after radiation. Large defects of the skull require a complete reconstruction to avoid several complications.

Keywords Chronic radiodermatitis · Osteoradionecrosis · CAD/CAM · Cranioplasty · Patient-specific-implants

Introduction

Chronic radiodermatitis and osteoradionecrosis of the skull are common complications after radiotherapy. Depending on the radiation dose and patient risk factors, these complications can be seen even many years after radiation. The therapy, especially in cases of combined necrosis of soft and bony structures, includes complete resection and reconstruction. Three-dimensional (3-D) imaging is necessary for presurgical planning of a computer-aided designed/ computer aided manufactured (CAD/CAM) patient specific-implant (PSI). In combination with a soft tissue free flap, PSI might be a useful tool for reconstruction of larger defects especially of the skull.

Case report

A 77-year-old female was admitted to our clinical center with a chronic ulcer of the skin on the left side of the forehead.

Additionally, the patient showed a large scar in this area as a result of resecting a basal cell carcinoma several years prior (Fig. 1). Hence, the ulceration was suspected of being a recurrence of the previous known carcinoma. Biopsies of the tumorous lesion showed no malignancy but a chronic inflammatory ulcer of the skin and surrounding tissue. Medical history revealed an arterial hypertension and a stroke with an associated hemiplegia of the left side of the body few years ago. The patient did not suffer from any clinical residuals yet and was almost free of any other complaints.

As an interesting fact, the patient's medical history revealed radiotherapy centered on the forehead in early childhood as therapy for hemangioma in the 1940s (Fig. 2). Magnetic resonance imaging (MRI) showed an extended lesion of the underlying tissue and skull caused by osteoradionecrosis and chronic radiodermatitis as a late complication of the radiotherapy (Fig. 3).

The first step of surgical therapy comprised resecting the affected soft and hard tissue up to the brain tissue (Fig. 4), since the dura mater was not intact on this side. During the same session, the through-and-through defect was closed by using a radial forearm free flap (RFFF, Fig. 5). Anastomoses were performed between the radial and superficial temporal vessels. Histopathological findings revealed an extended necrosis of the skin and the

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Fig. 1 Presurgical photograph showing ulceration and scarring formation on the left side of the forehead

subepidermal layers according to a radiodermatitis. The resected, underlying bone showed a fibrotic osteomyelitis with an active inflammation of the surrounding soft tissue.

After primary surgery, a PSI was planned using the patient's postsurgical digital imaging and communications in medicine data obtained from computed tomography (CT) scans. The CAD/CAM-PSI itself consisted of a 3D-printed Titan mesh with a bioceramic, faceted surface, and retention arms for screw fixations, produced and delivered by our provider (Ossdsign®, Uppsala, Sweden, Figs. 6 and 7).

In the second step, the PSI was implanted for reconstruction of the bony skull. By inserting the PSI between the fascia covering the brain and the subcutaneous tissue—both of the former RFFF—the fascia became a new dura mater preventing direct PSI-to-brain tissue contact (Fig. 8). As of this writing, the patient has no symptoms and is very satisfied with the aesthetic result (Fig. 9).

Discussion

Today, radiotherapy for treatment of hemangioma in childhood—used from the 1940s to the 1970s—is almost obsolete [1, 2]. Chronic radiodermatitis and osteoradionecrosis in adults, occurring as late complications of radiotherapy administered in early childhood, are uncommon, but can be observed even nearly 80 years after radiation. The risk of developing such a complication after radiotherapy does not decrease over time but is persistent and even may accelerate, especially in cases of any risk factors. At least, radiotherapy-dependent risk factors must be considered such as total radiation dose and fractionation, and size of the irradiated area [3].

Besides common facial skin alterations such as basal cell carcinoma, squamous cell carcinoma, or actinic keratosis, chronic inflammatory ulcerations following extensive radiodermatitis—and even osteoradionecrosis—are a noteworthy differential diagnosis of alterations and lesions of the



Fig. 2 Patient in early childhood with hemangioma on the left side of her forehead

Fig. 3 Presurgical MRI scan (T2, sagittal view) showing lesions of the soft tissue and skull on the left side of the forehead

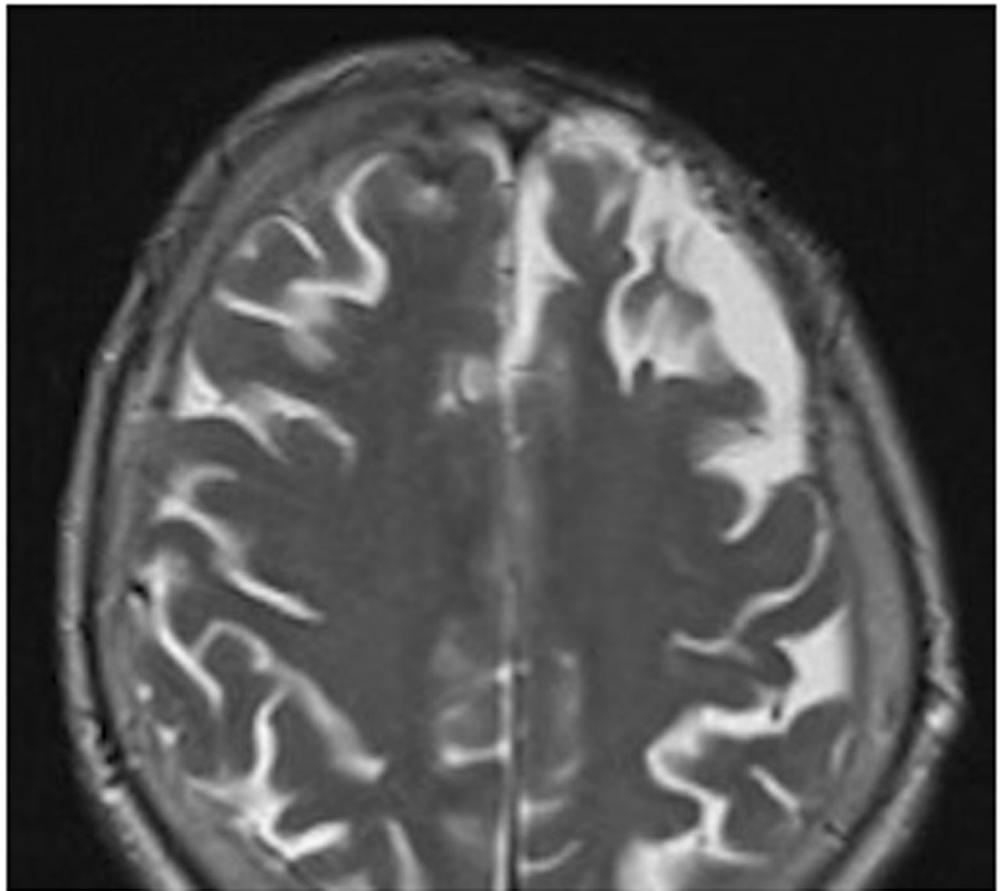


Fig. 4 Intraoperative view of the defect after resection of the osteoradionecrotic tissue

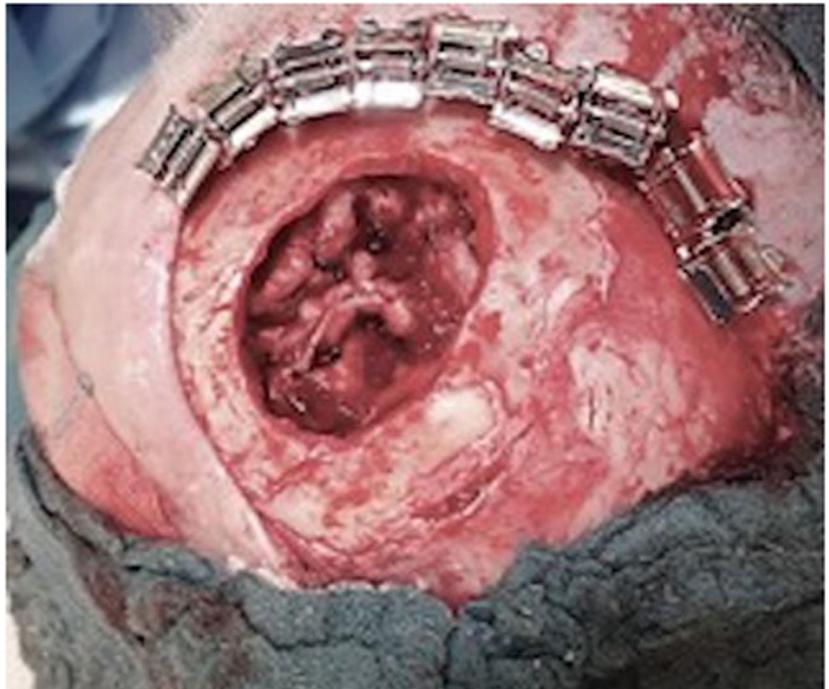


Fig. 5 RFFF for soft tissue covering of the skull defect



Fig. 6 Computer tomography after the first step of surgical therapy (3D reconstruction) with visualized pedicle of the RFFF

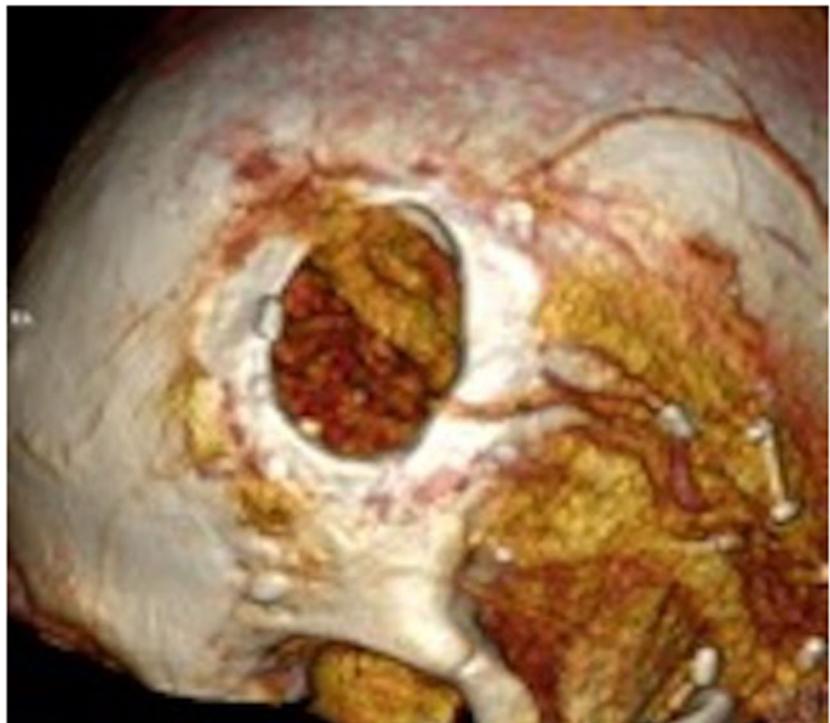


Fig. 7 CAD/CAM planning of the patient-specific implant

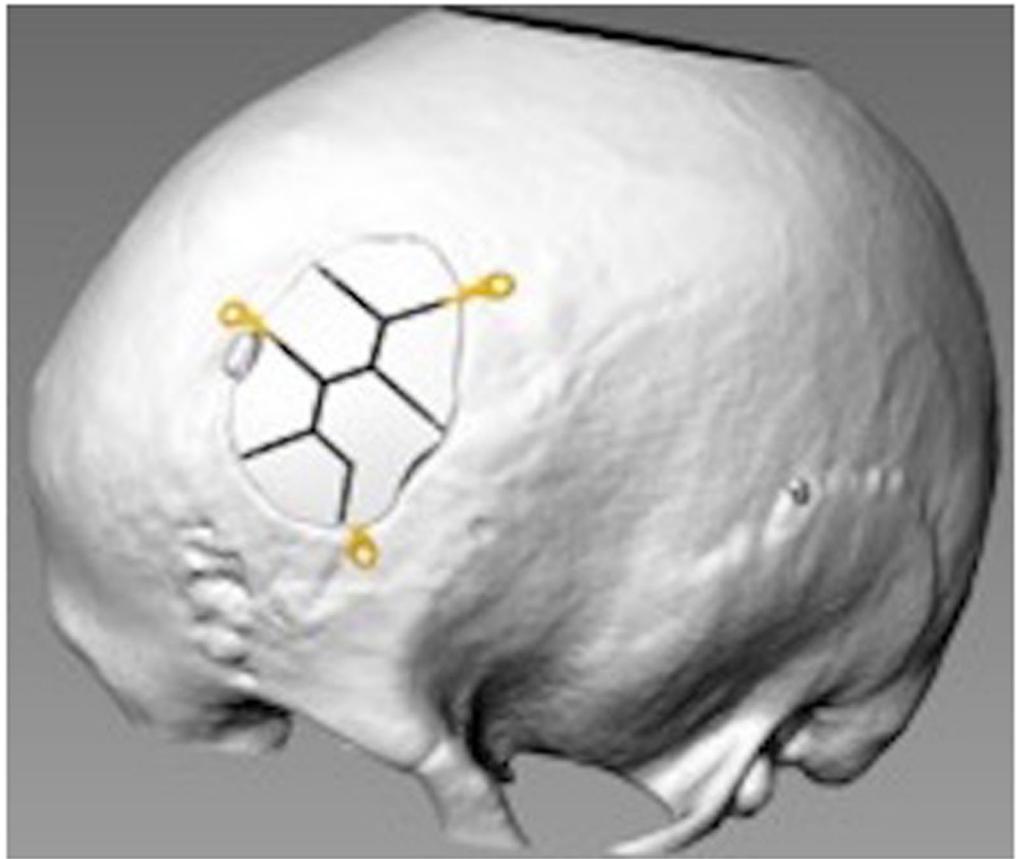


Fig. 8 Intraoperative view during the second step of therapy with reentry and reconstruction of the dura mater with the fascial part of the RFFF before implantation of PSI



Fig. 9 Postsurgical photo of the patient



facial skin that should not be underestimated in clinical practice [4].

Large defects of the skull require a complete reconstruction to avoid several complications. Such cranioplasties are mandatory to protect the underlying cerebral tissue and to improve the aesthetic appearance by mimicking the natural shape of the skull bone, especially in the forehead region [5, 6]. For these purposes, we decided to choose the aforementioned PSI, as we have achieved good results with it in the past. These PSIs are well proven with respect to stiffness and strength, and furthermore, even have the potency to induce bone healing of cranial defects [7, 8].

Beyond the indications for cranioplasty from the surgical point of view, another benefit is prevention of the so-called “sinking skin flap syndrome,” which contains neurological deficits like vertigo, headache, fatigue, and concentration problems as a result of intracranial pressure variations caused by the property of the skin to adhere directly to brain tissue [9].

Conclusion

Reconstructing through-and-through bony and dermal skull defects by an alloplastic PSI combined with a free flap is a

reasonable and relatively low-risk procedure with a high degree of efficiency and satisfactory outcome, even in elderly patients.

Compliance with ethical standards

Competing interests None of the authors have any conflict of interest to declare.

Ethical approval Not required.

Patient consent The subscribed patient consent declaration form is attached to the submission of this manuscript.

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