Giant keloid of left buttock treated with post-excisional radiotherapy

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ABSTRACT

Keloids are defined as excessive scar tissue formation extending beyond the area of the original skin injury and occurring in predisposed individuals. While no single treatment has proven widely effective, several series report excellent outcomes for keloids with post-surgery radiation therapy as described in the literature. We present a patient with recurrent giant keloid of left buttock after several surgical removals, that at physical examination shows the size of 40x22x10 cm in the largest dimension. Patient underwent a surgical excision of gluteal lesion and postoperative radiotherapy using photons at 8 MV of linear accelerator: the total dose delivered was 22 Gy in 11 days, with a daily fraction of 2 Gy. No relapse was showed at 36 months post-therapy. Several methods seem unsatisfactory for preventing keloid recurrence. The combination of surgery and adjuvant radiotherapy seems an excellent strategy to prevent recurrent disease.

CASE REPORT

CASE REPORT

A 22 years old man presents with a giant keloid of left buttock and an umbilical scar hypertrophy. He shows a small stature, minimal cognitive deficit, and clinical history of seizures treated with drugs.

The patient was treated with several surgical removals of keloids after excision of lipoma of the left buttock at the age of 11 years. Hypertrophied umbilical scar was the stable result of cutting the umbilical cord at the childbirth and was never treated. The lesion has a rosy color, roughly ovoid and mammillated, the size of about 40x22x10 cm, sometimes ulcerated, not sore, occupying the left buttock, medially to the root of the thigh and surrounding the anal orifice, histologically corresponding to scared giant and protuberant keloid (Fig 1).

MRI of the pelvis shows a giant gluteal lesion covering the adjacent local irregular fat profile. The lesion has a signal iso-hypointense to skeletal muscle in T1-weighted sequences, hypo-intense to subcutaneous fat in T2 images and shows hyperintense signals on fat suppression sequences with moderate and irregular pattern of contrast enhancement (Figures 2-6).

PET-CT reveals an increased and heterogeneous uptake of 18F-FDG at the left buttock (Figures 7-8).

The patient has considerable discomfort and negative impact on walking. Patient underwent a surgical excision of gluteal lesion and a rotation flap was performed to cover the wide wound. Histological examination describes a giant keloid of 3.4 kg in weight.

Two months following surgical excision, the patient was treated with radiotherapy using photons at 8 MV of linear accelerator. The total dose delivered was 22 Gy in 11 days, with a daily fraction of 2 Gy. The dose fraction was split between four fields with 20°-270°-50°-230° portal arrangement. Bolus of 12 mm was administered. The 95% isodose target area included the entire postoperative scar and any suture with a margin of 3-5 mm around the lesion. Non-target areas were shielded with multileaf collimator (Figure 9).

At 36 months post-therapy, the lesion in question had not recurred; no skin complications have been noted, and the patient is currently being followed up (Figure 10).

DISCUSSION

A keloid may be defined as a benign growth of dense fibrous tissue developing from an abnormal healing response to a cutaneous injury such as surgery, extending beyond the original borders of the wound or inflammatory response. Keloids, in distinction to normal scars, generally increase in dimension over time, and in addition to creating deformity can cause numbness, tingling and itching (1,2).

From an epidemiological viewpoint, certain races such as Blacks, Hispanic, and Asians are more susceptible to keloid formation than Caucasians (3,4).

The pathogenesis of keloids is complex and involves both genetic and environmental factors (5). While most keloids are sporadic, familiar keloids seem to represent an incomplete penetrance autosomal dominant disease, with varying degrees of clinical severity within a pedigree (6). Environmental factors are surgery, burns, trauma, inflammation, foreign-body reactions, endocrine dysfunction. However, they occasionally occur without apparent external cause. They are characterized by excessive collagen and glycosaminoglycan deposition within the dermis, an increase in collagen turnover, and micro-vasculature regeneration (7,8,9).

They can range in size from small papules limited to only a few millimetres in diameter to football size and larger. Their texture can vary between a soft and dough-like to a hard and rubbery consistency.

Even if histological analysis (performed as first step in the diagnostic worklflow) clarifies the nature of the lesion, the non-specific morphological features and the necessity to assess the involvement of neighboring structures lead to the necessity of a differential diagnosis with dermatofibrosarcoma protuberans (DFSP), foreign body granulomas, lobomycosis (also known as keloidal blastomycosis) and scars with sarcoidosis.

Dermatofibrosarcoma protuberans (DFSP) is a low-intermediate grade sarcoma originating from the dermal layer of the skin with aggressive tendency to infiltration (possible bone erosion) and fungating growing.

Even if X-Ray, Ultrasound and CT are useful to detect the heterogeneous lesion and, above all, signs of extra-compartmental invasion, MRI has a primary role in differential diagnosis showing irregular hyper-intensity signal in T2 sequences and subcutaneous fat infiltration in fat sat images.

Scintigraphy and PET-CT underline growing activity of the tumor with intense-moderate uptake of radiotracer (10,11,12).

Radiological findings of foreign body granuloma depend on its chemical nature. Even when not directly visible at X-Ray or Ultrasound, the identification of a surrounding wall is a useful indirect sign of the presence of foreign body. CT can detect a well circumscribed lesion of variable intensity of attenuation with enhancing wall. MRI in T1 sequences usually shows a hypo- or iso-intense lesion with possible peripheral high-signal-intensity rim (13,14,15). In both the techniques contrast enhancement has a variable, irregular pattern with evidence of enhancement rim. Scintigraphy and PET-CT show variable, irregular increased uptake of radiotracer with a ring-shaped pattern (16).

Lobomycosis is a cutaneous fungal infection characterized by skin nodules and plaques resembling keloid. As the disease is limited to the cutis, the diagnostic workflow ends with the isolation of fungus at hysto-cytological analysis and radiological exams are not performed (17,18,19).

Infiltration of old scars with sarcoid granuloma is usually a clinical diagnosis of exclusion confirmed by biopsy.

In keloid lesions, the therapy chosen is predicated upon several factors, including: size of lesion, location, depth of lesion, age of patient, and past response to treatment (20,21). The low local control rate achieved by surgery alone has led to the use of many other treatment methods (22); but variable success has followed either single use or combination (e.g. intralesional steroids, 5-fluorouracil, interferon, pulsed dye laser, compression with silicon sheeting, surgery, pressure devices, radiation, cryosurgery and radiotherapy): the optimal treatment for keloids is actually still undefined (23).

This multitude of treatment modalities underlines how little known is disease process and indicates the complex and multi-variable pathogenesis of this disease (24). The meta-analysis of 70 treatment series for various clinical measures showed an approximately 70% chance to obtain 60% improvement in keloids; however, there was no statistically significant difference between treatments (25).

It has been shown treatment of keloid with excision and postoperative radiation therapy to be an effective therapy (26). With appropriate doses the recurrence rate can be reduced to a < 10-20% (27,28,29). Radiation therapy is applied with superficial kilovolt X-rays, electron beams, telecobalt, high X-rays (4-6-8 MV) and interstitial radiation therapy either at low or high dose rate.

The only prospective randomized trial of any kind for keloids demonstrated greater control rates for surgical excision and radiotherapy compared to surgery and corticosteroid

injection, with recurrence rates of 12.5 % after surgery and radiotherapy, versus 33% after surgery and steroid injections, though with a statistically non- significant mean differential (30).

The favourable outcomes with this approach are attributable to destruction of keloid fibroblasts by ionizing radiation, which has been shown to enhance apoptosis when delivered in small to moderate dose (31).

TEACHING POINT

Keloids are lesions with still undefined pathogenesis and non-specific morphological features that require adequate differential diagnosis and treatment approach. In giant keloids an extended radiological study is recommended in order to evaluate the involvement of neighboring structures and to provide useful information for interdisciplinary therapy.

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FIGURES

Figure 1. 22 years old man with giant left buttock keloid. In the pre-treatment photograph the lesions shows rosy color, roughly ovoid and mammillated shape, some ulcerations and a size of about 40x22x10 cm.

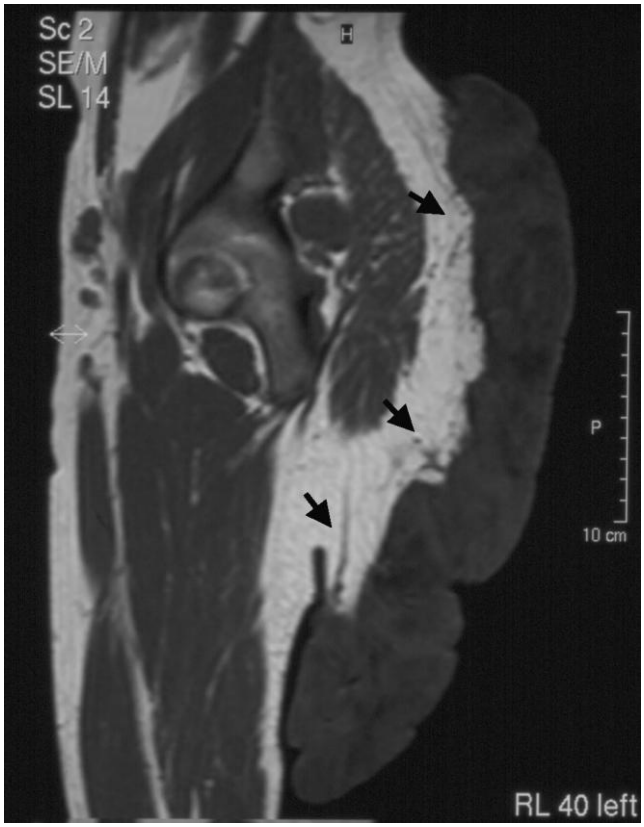


Figure 2. 22 years old man with giant left buttock keloid.
MRI of the left buttock showing a large iso-hypointense mammilliform lesion extending in cranio-caudal direction and covering the adjacent local irregular fat profile (arrows). Protocol: Philips Gyroscan 1T: Sagittal T1-SE, Thickness: 6.0/0.6, FOV: 450/1.2, NSA2: 205/256.

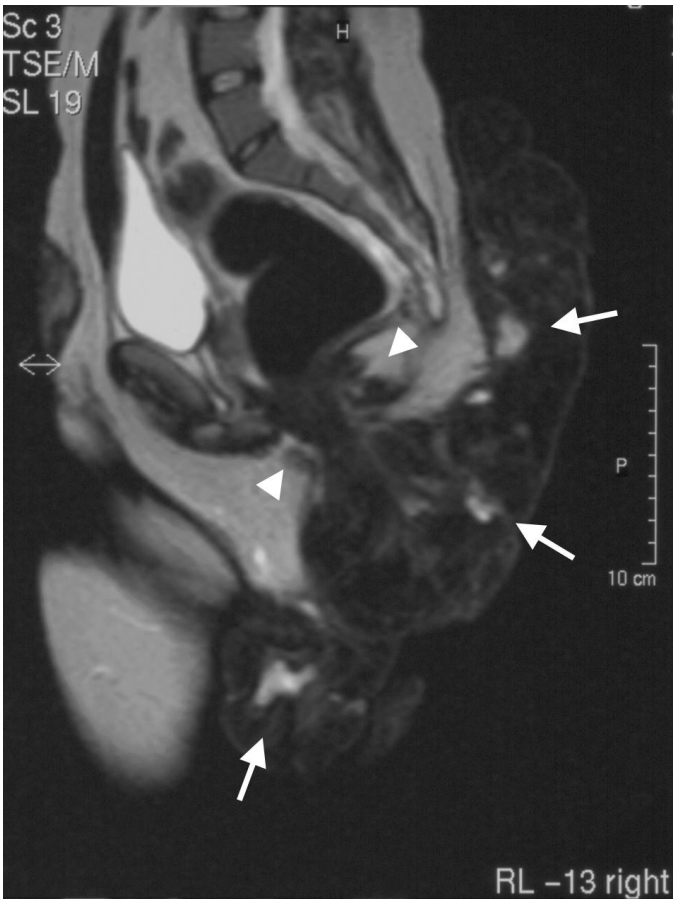


Figure 3. 22 years old man with giant left buttock keloid. MRI of the left buttock showing a large hypointense mammilliform lesion with hyperintense cystic/colliquative necrotic areas inside (arrows), covering the adjacent local irregular fat profile and surrounding the anal orifice (arrowheads). Protocol: Philips Gyroscan 1T: sagittal T2-TSE, Thickness: 7.0/0.6, FOV: 470/1.2, NSA 6: 195/256.

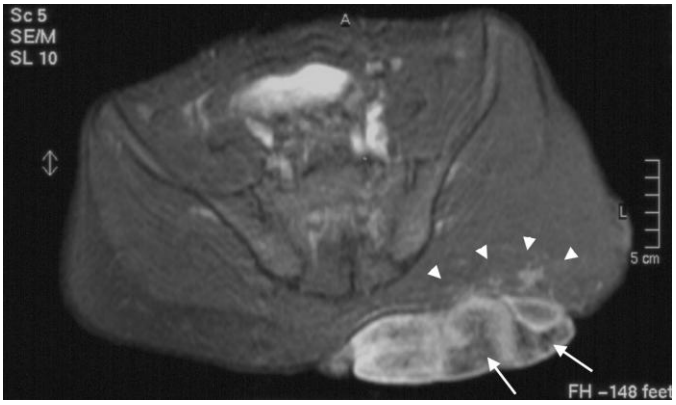


Figure 4. 22 years old man with giant left buttock keloid. MRI shows an intense and heterogeneous signal with irregular fat profile (arrowheads) and colliquative necrotic areas (arrows) inside the mammilliform lesion. Protocol: Philips Gyroscan 1T: axial T1-weighted MR fat sat (SPIR), Thickness: 10/1.0, FOV: 380/1.2, NSA 2: 179/256.

Figure 5. 22 years old man with giant left buttock keloid. MRI shows an intense and heterogeneous contrast enhancement of the mammilliform lesion, irregular enhancement of adjacent fat (arrowheads) and colliquative necrotic areas inside the lesion (arrows). Protocol: Philips Gyroscan 1T: axial T1-weighted MR fat sat (SPIR) after intravenous gadolinium administration (20 ml Omniscan 0,5 mmol/ml), Thickness: 15/1.0, FOV: 400/1.1, NSA 2: 179/256.

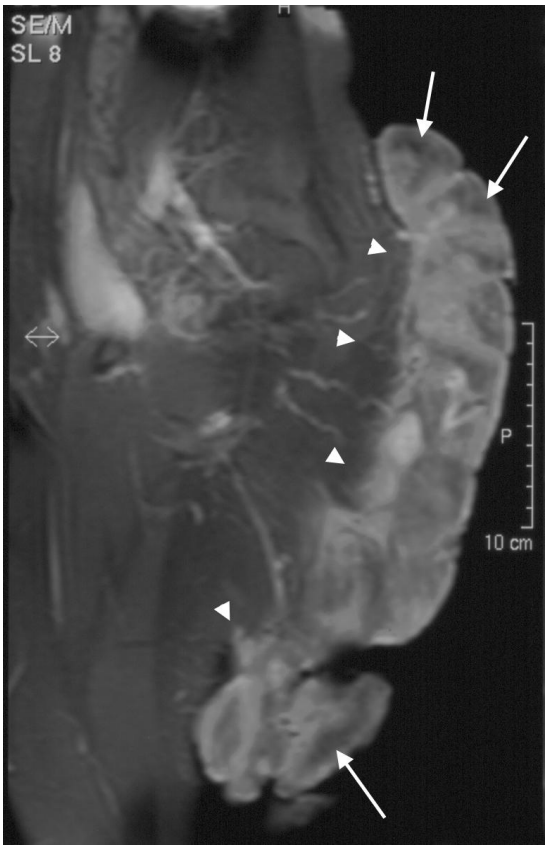


Figure 6. 22 years old man with giant left buttock keloid. MRI shows an intense and heterogeneous contrast enhancement with hypointense colliquative necrotic areas inside the lesion (arrows) and irregularity of fat profile (arrowheads). Protocol: Philips Gyroscan 1T: sagittal T1-weighted MR fat sat image after intravenous gadolinium administration (20 ml Omniscan 0,5 mmol/ml), Thickness: 12/1.0, FOV: 470/1.1, NSA 2: 179/256.

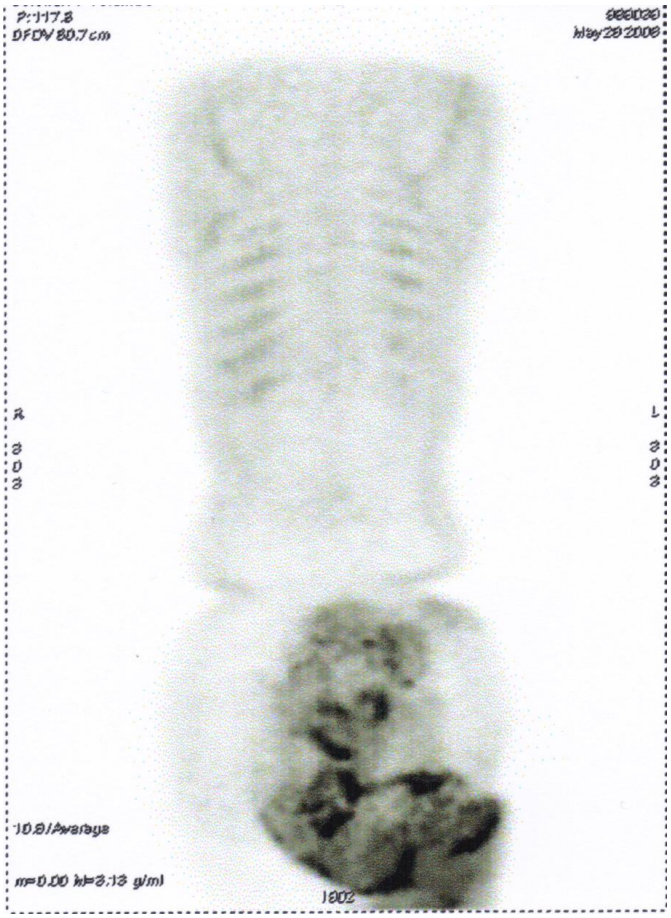
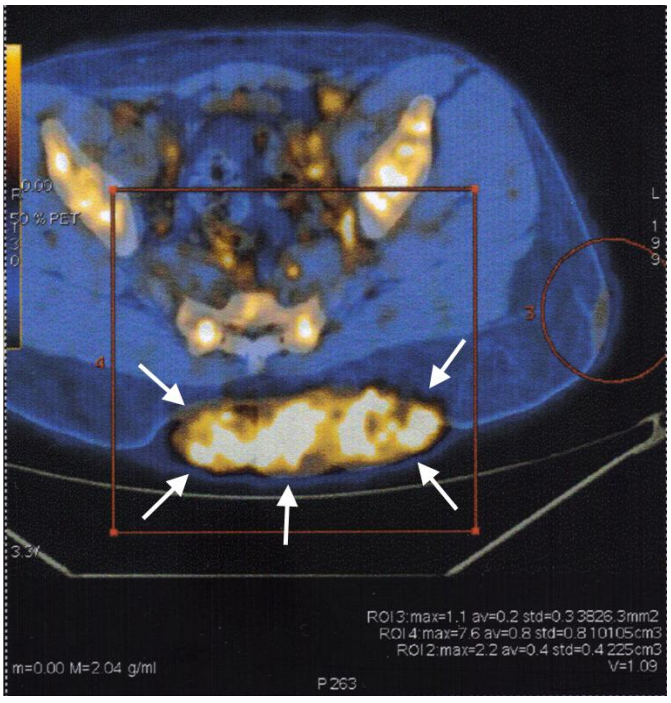


Figure 7. 22 years old man with giant left buttock keloid. Coronal PET-CT image; injected activity of 307 MBq; 3D acquisition starting at 50 min after intravenous administration of 18F-FDG. Delayed acquisitions (120 min after acquisition) show intense and heterogeneous radiotracer uptake (maxSUV 4.00 - 8.30).



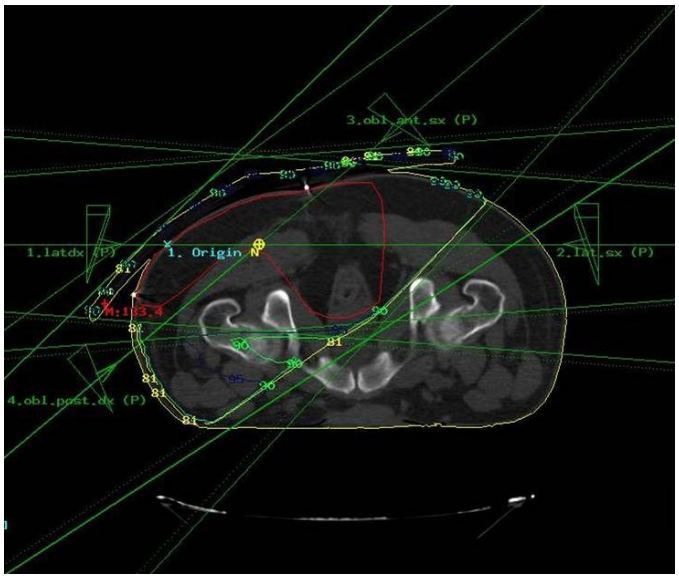


Figure 9. 22 years old man with giant left buttock keloid. Axial CT image with radiation treatment maps. The 95% isodose target area included the entire postoperative scar and any suture with a margin of 3-5 mm around the lesion.

Figure 10. 22 years old man with giant left buttock keloid. Post-therapy photograph at 36 months: the lesion had not recurred, and the patient reported no skin toxicity.

Figure 8 (left). 22 years old man with giant left buttock keloid. Axial PET-CT image; injected activity of 307 MBq; 3D acquisition started at 50 min after intravenous administration of 18F-FDG. Delayed acquisitions (120 min after acquisition) show intense and heterogeneous radiotracer uptake (maxSUV 4.00 - 8.30) (arrows).

Etiology	Unknown	
Incidence	Blacks, Hispanic and Asians are more susceptible than Caucasians	
Gender ratio	1:1	
Age predilection	No age predilection	
Risk factors	Genetic Factors Environmental factors • surgery • burns • trauma • inflammation • foreign–body reactions • endocrine dysfunction	
Treatment	Surgery excision, usually followed by post-operative treatments: • radiotherapy • radiation • cryosurgery • intralesional steroids • 5-fluorouracil • interferon • pulsed dye laser • compression with silicon sheeting • pressure devices	
Prognosis	Recurrence rates depend on treatment methods	
Findings on imaging	**X- Ray**	soft-tissue mass or soft-tissue swelling
	US	heterogeneous echogenic soft-tissue mass
	CT	Cutaneous and sub-cutaneous involvement with heterogeneous architecture (multi-lobulated) and tissue attenuation approximately equal to (or less than) skeletal muscle
	MRI – T1	isointense to muscle
	MRI – T2	hypointense
	Pattern of contrast enhancement	avid and heterogeneous
	Scintigraphy	Delayed increased uptake
	PET	Increased uptake

Table 1: Summary table of giant keloid

	Keloid	Dermatofibrosarcoma protuberans (DFSP)	Foreign body granuloma (1)
X-Ray	Soft-tissue mass or soft-tissue swelling without evidence of bone involvement or calcification	Soft-tissue mass or soft-tissue swelling with possible bone involvement or calcification	Foreign body or soft tissue mass or soft-tissue swelling
US	Heterogeneous echogenic soft-tissue mass	Heterogeneous echogenic soft-tissue mass	Heterogeneous echogenic soft-tissue mass
CT	Cutaneous and sub-cutaneous involvement with heterogeneous architecture (multi-lobulated) Tissue attenuation approximately equal to (or less than) skeletal muscle	Distinct lobular or nodular architecture with signs of extra-compartmental invasion and tissue attenuation approximately equal to (or greater than) skeletal muscle	Foreign body, homogeneous or heterogeneous architecture with varying intensity attenuation (well circumscribed hypodense lesion with enhancing wall)
MRI – T1	Hypo-Iso-intensity to skeletal muscle	Hypo-iso intensity to skeletal muscle Hyper-intensity in case of hemorrhage	Hypo- or Iso-intensity with possible peripheral high-signal-intensity rim
MRI – T2	Hypo-intensity to subcutaneous fat Hyper-intensity to subcutaneous fat of cystic/colliquative necrotic areas.	Irregular intensity, greater to subcutaneous fat Hyper-intensity in case of hemorrhage or colliquative necrotic areas	Homogeneous hyper-intensity or hypo-intense bands inside the lesion
Pattern of contrast enhancement	Low or moderate, Irregular	Intense or Moderate, Irregular	Variable, irregular, possible enhancement rim
Scintigraphy	Delayed increased uptake	Delayed increased uptake	Variable, irregular increased uptake with possible enhancement rim
PET	Increased uptake	Increased uptake	Variable, irregular increased uptake with possible enhancement rim

Table 2: Differential diagnoses of giant keloid
[1] Radiological findings of foreign body granuloma depend on the chemical nature of the foreign body.

ABBREVIATIONS

CT: Computed Tomography
MRI: Magnetic Resonance Imaging
PET-CT: Positron Emission Tomography - Computed Tomography
Gy: Gray
MV: MegaVolt
18F-FDG: 18F - Fluoro-DeoxyGlucose
DFSP: DermatoFibroSarcoma Protuberans
SUV: Standardized Uptake Value

KEYWORDS

giant keloid; post-excisional radiotherapy; local control