

Post-implantation erythema in 3 patients and a review of reticular telangiectatic erythema

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Summary

Background. The appearance of erythematous, blanchable patches or plaques overlying an implant suggests possible reticular telangiectatic erythema (RTE). RTE is a benign reactive cutaneous manifestation that can present following the implantation of a cardiac pacemaker, defibrillator or intrathecal infusion pump in an otherwise asymptomatic, non-infectious patient.

Objective. To demonstrate the variety in clinical presentation of patients presenting with RTE or similar patch test-negative post-implantation erythema.

Patients/Materials/Methods. After institutional board approval had been obtained, patient information was obtained from electronic medical record files, which included surgical reports, pathology reports, and notes from outpatient encounters.

Results. We report post-implantation erythema following insertion of an elbow prosthesis, a knee prosthesis, and a spinal cord stimulator, which have not previously been cited as aetiologies of RTE.

Conclusion. Owing to the delayed onset and variable recovery, RTE remains a diagnostic challenge. RTE should be included in the differential diagnosis of any patient presenting with erythema over the site of a previously implanted device.

Key words: implantable device; pacemaker; patch test; post-implantation erythema; prosthesis; reticular telangiectatic erythema.

In 2005, international surveys underscored the increasing popularity of implantable cardiac devices worldwide and in the United States, where there are over 200 000 cardiac pacemakers in use (1). Orthopaedic implants are similarly ubiquitous, evident as knee replacements alone, which accounted for over 500 000 of the surgical procedures performed in the United States in 2006 (2, 3). Commonly cited cutaneous pathologies following orthopaedic and cardiac device implantation range from eczematous

dermatitis to bullous eruptions. Less frequently, the appearance of erythematous, blanchable reticular telangiectatic lesions overlying an implant prompts concern for reticular telangiectatic erythema (RTE). RTE was first described by Gensch and Schmitt (4), and has since been chronicled in multiple case reports in the English and European literature (Table 2). In each case, a reaction was preceded by the implantation of a cardiac pacemaker, defibrillator or intrathecal infusion pump in an otherwise asymptomatic, non-infectious patient (5–7). The clinical course and outcomes of RTE reports are variable; some patients benefited from removal, replacement or repositioning of the device, whereas others reported that RTE lesions resolved spontaneously. Previously, RTE was exclusively associated with cardiac devices, but, in the last 5 years, reports of RTE following the insertion of other implants have been documented (6–8). Here, we

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report three patients presenting with post-implantation erythema following the implantation of devices not previously associated with RTE (Table 1). We also review 22 published cases of RTE (Table 2).

Materials and Methods

After institutional review board approval had been obtained, patient information was deidentified and collected from electronic medical record files, which included surgical reports, pathology reports, and notes from outpatient encounters. Patch testing was carried out in accordance with the guidelines established by the North American Contact Dermatitis Group (NACDG) for an NACDG complete or abbreviated standard series and Cleveland Clinic prosthesis series (20–22). Patch-testing products were purchased from Chemotechnique Diagnostics (Vellinge, Sweden) and from AllergEAZE, SmartPractice Canada (Calgary, Alberta, Canada). Patch tests were applied to unaffected skin sites on the back with the use of aluminium disks (Finn Chambers®; Epitest Ltd, Hyryla, Finland) on pressure-sensitive, acrylic-based adhesive medical tape (Scanpor®; Norgesplaster AIS, Aksjeselskap, Norway). Patch tests were removed after 48 hr and read on day 2 or 3, and again on day 6 or 7. Interpretation of the patch test results was performed according to NACDG guidelines.

Case Reports (Table 1)

Case 1

Six months after undergoing a total left elbow arthroplasty, a 76-year-old woman presented with red patches on the dorsal aspect of her left arm and forearm that had appeared days after the implantation of her titanium elbow prosthesis (Fig. 1). The patient presented with no pain and a full range of motion, and the radiograph studies showed favourable alignment of the replaced joint. Prior to the total elbow arthroplasty, the patient had had plates and screws in her left arm for the treatment of elbow arthritis for 6 months, without incident. Twenty-five years earlier, the patient had metal plates and screws placed in her right arm, and this was followed by an uneventful recovery. She had seasonal allergies and asthma, and reported having skin irritation when using certain laundry detergents; however, it was not known whether the latter was an irritant reaction or contact allergy to specific ingredients.

A skin biopsy of the affected region showed mild fibrosis, chronic inflammation, and telangiectasia. Patch testing with the NACDG standard screening and the prosthesis series yielded negative results, which, with the clinical presentation and biopsy results, led to a diagnosis of RTE. After a bacterial culture grew methicillin-resistant *Staphylococcus epidermidis*, the

Table 1. Post-implantation erythema following insertion of orthopaedic devices

Case number	Age (years)/sex	Type of implant	Site	Associated clinicopathological features	Time of onset after device implantation	Patch test results and history of metal sensitivity	Outcome
1	76/F	Titanium elbow prosthesis	Left elbow	Telangiectatic blood vessels, chronic inflammation, mild fibrosis	Within days	Negative patch test results, including titanium and nickel. Patient reported possible jewellery allergy that resolved spontaneously 50 years earlier	Waxing and waning course
2	66/M	Spinal cord stimulator and pulse generator	Right flank	Vascular dilatation, perivascular interstitial lymphocytic infiltrate	8 months	Negative patch test results. No history of metal sensitivity	Unresolved with minimal improvement
3	64/F	30% cobalt chromium and 7% molybdenum knee prosthesis	Right knee	Perivascular interstitial lymphocytic infiltrate, angioplasia	7 months	Positive patch test for <i>Myroxylon pereirae</i> ; remainder of patch tests were negative. No history of metal sensitivity	Resolved without treatment

Table 2. Published reports of reticulated telangiectatic erythema

Source	Age (years)/sex	Type of implant	Site	Associated clinicopathological features	Time of onset after procedure	Duration	Patch test results and history of metal sensitivity	Outcome	Comments
Gensch and Schmitt (4)	NA/M	Cardiac pacemaker	Right side of thorax	Telangiectatic blood vessels, lymphohistiocytic infiltrate	3 weeks	Not specified	Not specified	Skin remained unchanged during long-term follow-up over several years	Spontaneously resolved after 3 months but reoccurred following electrode replacement
Kint and Vermander (9)	60/M	Cardiac pacemaker	Right side of chest	Vascular dilatation, moderate lymphohistiocytic infiltrate, burning sensation, redness was more pronounced upon exertion	'A few weeks'	1 year	Not specified	Resolved after replacement of pacemaker battery	—
Kopera et al. (8)	NA	Cardiac pacemaker and defibrillator	NA	Vascular dilatation, lymphohistiocytic infiltrate	NA	NA	Negative	Skin remained unchanged during long-term follow-up over several years	Three patients were documented
Maushagen et al. (10)	35/NA	Cardiac pacemaker	Chest	Circumscribed erythema, painful and fluctuating swelling	NA	10 years	Negative	NA	—
Cierpka et al. (11)	NA	Cardioverter defibrillator	Chest	Circumscribed erythema, telangiectatic blood vessels	NA	NA	Negative	Unchanged to follow-up after 28 months	—
	NA							Unchanged to follow-up after 10 months	

Table 2. Continued

Source	Age (years)/sex	Type of implant	Site	Associated clinicopathological features	Time of onset after procedure	Duration	Patch test results and history of metal sensitivity	Outcome	Comments
	NA								
Krasagakis et al. (12)	69/M	Cardioverter defibrillator	Left abdomen	Telangiectatic blood vessels, lymphohistiocytic infiltrate	8 months	Not specified	Negative	Improved without treatment at 9-month follow-up	1 month after first activation
Dinulos et al. (13)	63/F	Cardioverter defibrillator	Right lower abdominal quadrant	Tingling sensation, telangiectatic blood vessels, lymphohistiocytic infiltrate	4 years	7 months	Positive reaction to neomycin sulfate, ethylenediamine dihydrochloride, diethylene triamine, and nickel sulfate	Resolved with removal of device	Patient's second device: the first device was removed because of infection
Ferringer and Mowad (5)	76/M	Cardioverter defibrillator	Mid-abdomen to left flank	Vascular dilatation, occasional burning, lymphohistiocytic infiltrate	3 weeks	3 years	Negative	Increasingly intense erythema developed	The patient's third device
Herbst and Weiss (14)	69/M 26/M 68/M 70/M	Cardioverter defibrillator	Chest	Telangiectatic vessels	Not specified	Not specified	Negative	Not specified	—
Lin et al. (15)		Cardiac pacemaker	Chest	Telangiectatic vessels, minimal inflammatory response	16 months	Not specified	Negative	'Telangiectasia somewhat involuted after the procedure of skin biopsy'	—

Table 2. Continued

Source	Age (years)/sex	Type of implant	Site	Associated clinicopathological features	Time of onset after procedure	Duration	Patch test results and history of metal sensitivity	Outcome	Comments
Mercader et al. (6)	50/M	Intrathecal drug delivery system	Left flank	Telangiectatic vessels in the recticular and papillary dermis, lymphohistiocytic infiltrate, itchy, poorly defined margins	15 days	28 months	Negative	2 months following the removal of the device, the lesion disappeared	2 years following the outbreak, skin erosion of the site forced the device to be removed
Morgan et al. (16)	59/M	Sternal wire used in coronary bypass grafting	Chest	Telangiectasia, perivascular lymphocytic infiltrate, epidermal atrophy	12 months	NA	Negative	NA	Author identified as sternal erythema
Pitarch et al. (17)	68/M	Cardioverter defibrillator	Chest	Superficial telangiectasia was observed, but no histological study was conducted	18 months	6 months	Negative	Spontaneously disappeared, leaving residual hyperpigmentation	—
	73/M				24 months		Not performed		
	69/M				14 months		Negative		
Garcia et al. (18)	38/M	Cardioverter defibrillator	Lower abdominal area	Lymphohistiocytic infiltrate, slight tumefaction in the vessel walls	Not specified	Not specified	Negative	Not specified	—
Milpied-Homs et al. (7)	56/M	Morphine pump	Abdomen side	Pigmented telangiectatic lesion	NA	NA	NA	NA	—
Martin et al. (19)	60/M	Cardiac pacemaker	Left chest	Dermal telangiectasia, subacute spongiosis	3 months	Not specified	Not specified	Ulceration of the lesion led to device removal; mild improvement upon removal	This was the patient's second pacemaker

NA; not available



Fig. 1. Erythema over the left elbow following implantation of a titanium elbow prosthesis in case 1.

patient was treated with antibiotics for more than a year, without improvement. The affected area spontaneously but temporarily improved following a joint aspiration of sero-sanguinous fluid, but since then has had a waxing and waning course.

Case 2

A 66-year-old man presented with a 3-week history of a $16 \times 6 \text{ cm}^2$ warm reticular erythematous patch over the right side of his abdomen (Fig. 2). He described the affected area as intermittently itchy and swollen, but did not complain of any pain. Eight months earlier, he had had undergone surgery for the implantation of a replacement T8–9 spinal cord stimulator electrode via thoracic



Fig. 2. Reticular erythematous patch over the right flank following implantation of a spinal cord stimulator and pulse generator in case 2.

laminectomy, and a pulse generator on the right flank. Previously, he had two metal rods, a spinal cord stimulator and a pulse generator implanted without any cutaneous complications. The patient was treated with clindamycin 300 mg, and noticed a transient improvement in redness. A skin biopsy revealed superficial perivascular and interstitial lymphocytic infiltrate associated with smudging of the dermoepidermal junction and the presence of a few dyskeratotic keratinocytes. Patch tests with an abbreviated screening and prosthesis series were all negative. The absence of mucin excluded a diagnosis of reticular erythematous mucinosis. There was no history of heat application to the site, which argued against erythema ab igne and narrowed the differential diagnosis to RTE. In subsequent follow-up appointments, the affected skin area did not significantly improve, and it appeared that the efficacy of the spinal cord stimulator was declining, possibly indicating an internal device leak. The orthopaedic surgeon was prompted to replace the implanted devices for a third time. Three days after this, an erythematous patch again developed over the right flank area, suggesting that a faulty device could not account for the skin eruption. No follow-up information was available.

Case 3

A 64-year-old woman presented with a 7-month history of an occasionally pruritic rash above her knee. The eruption began on the lateral aspect of her right leg, and moved medially around the area of her knee prosthesis, but did not involve the incision site. The rash was unresponsive to treatment with clobetasol and doxycycline. The patient denied any history of contact allergies to topical antibiotics, fragrances, or metals. Examination revealed a non-tender 10-cm area of telangiectasia, blanchable erythema, and mild oedema (Fig. 3). Patch testing with an abbreviated screening and prosthesis series was positive for *Myroxylon pereirae* (Balsam of Peru), but the relevance of this result was unknown. Tissue culture and stains were all negative. A biopsy revealed a mild perivascular lymphocyte inflammatory infiltrate in association with a reactive vascular proliferation. Some of the vessels showed intravascular reactive angioendotheliomatosis, which was confirmed with an immunohistochemical stain for CD31. There was also some dermal fibroplasia.

Discussion

The reported clinicopathological features of RTE have been protean and relatively non-specific. Nevertheless,



Fig. 3. Blanchable erythema and mild oedema over the right leg following implantation of a knee prosthesis in case 3.

the clinical and histological findings evident in case 1 are clearly consistent with previous descriptions of RTE. The post-implantation erythema in cases 2 and 3 also resembles prior reports of RTE. In case 3, telangiectasia was noted clinically but not histologically, which made the diagnosis more challenging. Two of the three (cases 1 and 2) patients previously tolerated implanted metal devices. All three cases had negative patch test reactions to the prosthesis series, which included titanium. The positive patch test reaction to *M. pereirae* in case 3 was not relevant to the skin reaction observed at the implantation site.

We reviewed the 22 published cases of RTE, which are summarized in Table 2. The most frequently associated implants were cardiac pacemakers, cardioverter defibrillators, and morphine pumps. The onset of erythema ranged from days up to 4 years following implantation; the mean onset was 30 months. Clinical outcomes were inconsistent among the reports. Some authors, such as Martin and Wendschuh (19), reported mild improvement of erythema following removal of the implanted pacemaker device. A report by Kint and Vermander (9) indicated that erythema improved following replacement of a pacemaker battery. Conversely, some skin findings remained unchanged in long-term follow-up, years after the onset of erythema (5, 12, 13).

In evaluating a patient presenting with a cutaneous reaction near or at the site of an implanted device, we recommend that a detailed contact dermatitis history be obtained, as well as a complete account of the perioperative and post-procedural periods (21, 23). As described in Table 2, patch testing results in patients with RTE were negative for the materials commonly found in implantable devices, such as polyurethane,

silicone rubber, silicone medical adhesive, titanium, and parylene-coated titanium (13). In 2003, Herbst and Weiss (14) contended that patch testing and histological evaluation are unnecessary if a patient presents with all of the clinical features of RTE, including the absence of infection and dermatitis (14, 17). However, we believe that patch testing may be useful, because negative patch test results support a diagnosis of RTE, and aid in excluding a diagnosis of erythema stemming from allergic contact dermatitis. Furthermore, we recommend histological examination of skin biopsy specimens in patients with post-implantation erythema, as many pathologies, including intravascular reactive angioendotheliomatosis and reticular erythematous mucinosis, may resemble RTE clinically, but could be distinguished from RTE by histological findings.

The most consistent finding on biopsy has been telangiectasia and a perivascular to interstitial lymphohistiocytic infiltrate. Spongiosis and increased dermal mucin has occasionally been reported. One of our cases demonstrated intravascular reactive angioendotheliomatosis, which, to our knowledge, has not been previously described in RTE. Intravascular reactive angioendotheliomatosis has most commonly been associated with infection, but has also been associated with a variety of other conditions, including cholesterol emboli, vascular shunts, antiphospholipid antibody syndrome, haematological malignancies, hepatitis, and renal disease, none of which were co-existing conditions in our patients (24, 25). Given that our case 3 was clinically characteristic of RTE, the biopsy finding of intravascular reactive angioendotheliomatosis expands the potential histological spectrum of RTE.

RTE is most often a diagnosis of exclusion, supported primarily by a temporal relationship with device implantation, negative patch test results, and the absence of signs of infection (tenderness, fever, pain, and response to antibiotics) (5, 14). However, it should be noted that RTE can appear concurrently with infection, as Dinulos et al. (13) reported a case of RTE following the implantation of a cardioverter defibrillator that was associated with a positive *S. epidermidis* culture. Similarly, positive cultures for methicillin-resistant *S. epidermidis* were seen in our case 1, who presented with RTE following an elbow prosthesis. As in the report by Dinulos et al. (13), our patient did not respond significantly to a rigorous antibiotic treatment. The role of *S. epidermidis* in RTE warrants further investigation.

Because of the delayed onset and variable recovery, RTE remains a diagnostic challenge. The differential diagnosis

Table 3. Differential diagnosis of post-implantation erythema

Annular erythema
Erythema ab igne
Intralymphatic histiocytosis
Intravascular reactive angioendotheliomatosis
Localized livedo reticularis
Metal sensitivity
Pacemaker dermatitis
Pressure dermatitis
Reticular telangiectatic erythema
Reticulated erythematous mucinosis
Sternal erythema
Telangiectasia
Wells syndrome

of RTE includes generalized telangiectasia (26), localized livedo reticularis, isolated telangiectasia (16), reticulated erythematous mucinosis, erythema ab igne (27), intravascular reactive angioendotheliomatosis (25, 28), pressure dermatitis (29), pacemaker dermatitis (30, 31), intralymphatic histiocytosis, metal sensitivity (23, 32), and Wells syndrome (33) (Table 3). Our cases 1 and 2 did not fit the criteria of these diagnoses, only the latter five of which have been associated with implanted devices. Gerdson et al. (34) reported annular erythema caused by impending pacemaker extrusion, which closely resembles RTE, as both are characterized by negative patch test results, vascular dilatation, lymphohistiocytic infiltrate, and erythema overlaying the implantation site. However, the two conditions differ in the presence of pain over the affected area, which is only seen in the case of impending pacemaker extrusion. Similarities between RTE and pathologies such as sternal erythema prompted Bowers et al. to suggest that many of these skin findings represent 'a larger and incompletely understood phenomena' (35, 36). The clinicopathological features of our case 1 were consistent with a diagnosis of RTE. Case 2 was diagnosed as having post-implantation erythema with evidence of vascular dilatation. Case 3 was histologically consistent with intravascular reactive angioendotheliomatosis, although there was no evidence of systemic infection, as might be found in intravascular reactive angioendotheliomatosis.

On the basis of our experiences with these cases and a review of previous RTE reports, we suggest that the diagnostic criteria for RTE include: (i) clinical observation of an erythematous, blanchable reticular telangiectatic area over an implanted device; (ii) biopsy demonstrating increased dermal telangiectasia and lymphohistiocytic infiltrate; (iii) absence of pain over the affected

area; (iv) negative patch test results with metals; and (v) absence of infection at the site or a failure to respond to appropriate antibiotic therapy. Previously, it was thought that implantable devices generated small electromagnetic fields and heat that resulted in the pathological findings of RTE (5). However, in 2005, Mercader-Garcia et al. (6) noted the appearance of RTE with an intrathecal infusion pump, which was unable to produce electromagnetic waves or heat in amounts sufficient to cause telangiectasia in the dermis. The lack of clinical signs of infection have led to hypotheses regarding microcirculatory changes that occur at the site of implantation as the underlying pathology (19, 37). This theory is most consistent with the findings of RTE in the cases that we present here, as 2 of the 3 patients were noted to develop RTE following the implantation of prosthetic devices that are not known to generate appreciable electromagnetic fields or heat.

It is very difficult to reconcile the rarity of RTE with the widespread use of implantable devices. However, it is likely that many cases go unreported, owing to the mild severity and relative lack of pain, which may not warrant medical attention. Furthermore, because the signs of RTE may appear months to years after surgical procedures, the temporal relationship between implantation and an erythematous reaction may be overlooked. Spontaneous resolution may be part of the reason why it is underdiagnosed.

Conclusion

The underlying pathogenesis and risk factors for RTE have yet to be elucidated, and await further study. With the increasing use of implantable devices, it is likely that the diagnosis of RTE will become more prevalent. Dermatologists, cardiothoracic surgeons and orthopaedic surgeons should be aware of the potential for RTE following the implantation of a device, as it can simplify diagnosis, treatment, and long-term management. In the past, RTE was exclusively found following implantation of cardiac devices; however, in recent reports it has been associated with other devices, such as morphine pumps and sternal wiring. Here, we present 3 cases of post-implantation erythema following insertion of an elbow prosthesis, a knee prosthesis, and a spinal cord stimulator, which have not previously been cited as aetiologies of RTE. We hypothesize that the scope of RTE goes beyond the spectrum of cardiac devices, and that it should be included in the differential diagnosis of any patient presenting with erythema at the site of the implanted device.

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