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SURGERY

Gangrenous Keloid of the Ear Pinna Following Intralesional Triamcinolone Injection: Proper Consent Is Essential

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Abstract

Keloid formation involving ears is commonly encountered in medical practice. There are variable treatment options with different outcomes particularly on recurrence. With treatment, there might be complications with varying severity. One of the commonest preferred treatment is intralesional steroid injection using triamcinolone acetonide (TCA). We encountered a complication of intralesional TCA injection when a patient who had an ear keloid developed gangrenous keloid involving the pinna after receiving a TCA injection that required surgical debridement. We believe such a complication has been underreported. We reviewed the literatures to highlight the complications following TCA injection of ear keloid.

Keywords Pinna keloid · Triamcinolone injection complication · Keloid gangrene · Keloid treatment complication

Introduction

Keloid formation is one of the two types of aberrant wound healing, the other being hypertrophic scars. Aberrant wound healing can arise with derangements in any of the healing phases including the inflammatory, proliferative, and remodelling phases [1]. Normal wound healing requires a balanced deposition of extracellular matrix protein and its subsequent degradation. This is described as the anabolic and catabolic state. An abnormal balance in this state will result in abnormal scarring [2]. These abnormal scars contain large amounts of inflammatory cells. During keloid formation, aggressive fibroblast activity causes deposition of more extracellular matrix resulting in the pathognomonic appearance of hypertrophic spread beyond the margins of the pre-existing wound [1]. Morphological and immunohistochemical difference between keloids and hypertrophic scars has been described. Keloids are known to have more type III collagen, chondroitin 4-sulfate, and glycosaminoglycan. Collagen fibrils are also more irregular and abnormally thick. Epidemiologically, keloids are known to occur commonly among people of darker pigment especially Africans, Hispanics, and Asians [1]. Familial predisposition is evident as well. Numerous theories postulating the aetiology of keloid formation have been discussed including genetics, metabolic, circulatory, immunologic, and even nutrition-related. Nonetheless, the exact aetiology is still unknown [3].

The objective of this article was to highlight possible complications following intralesional steroid treatment of keloid using triamcinolone acetonide (TCA) and to underscore the need for a proper consent taking in the management of keloid in general.

Case Presentation

A 17-year-old Asian female presented with a large pinna keloid which developed following ear piercing over a year ago. The keloid has been gradually growing and mainly affected the lobule of her left pinna. This was her first presentation with such a swelling. She reported no other similar swelling elsewhere on her body. The patient has no known comorbidity and no significant family history. On examination, there was a 4 × 3 cm keloid formation at her left pinna lobule primarily on the posterior surface. The other ear examination was normal. No facial dysmorphism was noted. Clinical diagnosis of keloid

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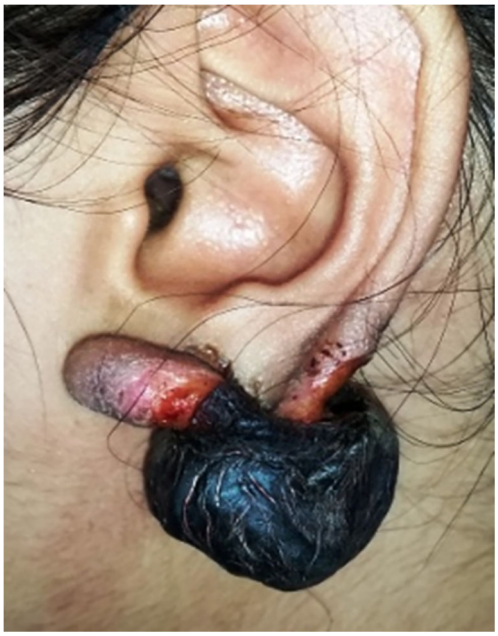

Fig. 1 Left pinna keloid 3 weeks post intralesional triamcinolone acetonide injection

was made, and the option of serial steroid injections offered for which she consented and underwent the first dose of 2 ml of 10 mg/ml intralesional TCA injection in the clinic. The procedure was uneventful. Subsequently, she developed blebs and swelling over the keloid after a week which later progressed into a gangrenous mass (Fig. 1). The gangrene was already dry and painless during her review in the clinic 20 days after the first and only TCA injection. An excision of the gangrenous keloid was done under general anaesthesia. Preoperatively, full blood count assay was normal with haemoglobin and platelet levels within normal limits. However, no other specific haematological investigation for blood disorder was done as she has no prior history of bleeding disorder. Intraoperatively, the base of keloid at the lobule was sloughy with no signs of perichondritis seen. The wound was dressed daily with saline and it eventually healed (Fig. 2).

Discussion

Keloid formation in the pinna is a well-known complication of ear piercings as encountered in this case. Despite the abundant knowledge and advancement on treatment options for keloid, there are neither standardized guidelines on the management nor existence of a focused consent form specific to keloids. Nonetheless, protocols such as the combination of steroid injections and surgical excision as proposed by Rosen et al. have produced encouraging results [4].

Non-surgical treatment modalities include intralesional injection with corticosteroid, bleomycin, 5-fluorouracil, pulse dye laser, carbon dioxide laser, pressure therapy, and cryosurgery. Surgery often consists of excision alone or excision followed by either grafting or adjuvant steroid injections. The choice of surgical excision with preoperative TCA appears to be well-established [5]. TCA injection was first used in 1961 [6]. The success of this treatment was later corroborated by several studies [7]. It acts by suppressing vascular endothelial growth factor as well as inhibitory action on fibroblast proliferation. This potentiates scar regression which remains as the objective of keloid management. Biochemically, TCA contains properties that inhibit transforming growth factor (TGF)–β1 expression which causes fibroblast apoptosis [2]. The reported complications following TCA injection of keloid are listed in Table 1.

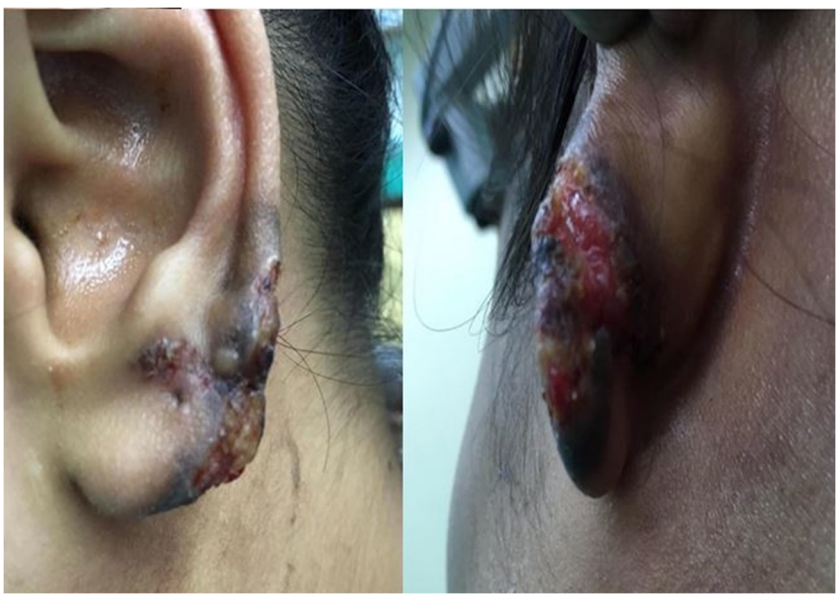

Fig. 2 Left pinna keloid 2 weeks post surgical debridement

Table 1 Keloid treatment with triamcinolone acetonide (TCA) alone or in combination and its complications

Author	Type of study	Treatment	Regime	Complication
Laisuan W et al.,2017 [8]	Case report (a 24-year-old woman with keloid over her back developed IgE-mediated hypersensitivity after TCA injection)	TCA injection	TCA dose unspecified	Anaphylaxis reaction (urticaria, facial numbness, and hypotension 15 min post injection).
Sukhumthammarat W et al., 2017 [9]	Case report (a 34-year-old woman with keloid over cesarean section scar and left shoulder)	TCA injection	100 mg of TAC monthly for 6 months	Cushing's syndrome
Finken MJ, Mui D, 2010 [10]	Case report (a 6-year-old girl with keloid over lower abdomen following burn)	TCA injection	160 mg of TCA followed by another 40 mg 3 months later	Cushing's syndrome (facial puffiness, weight gain 3 months after TCA).
Liu MF, Yencha M, 2006 [11]	Case report (a 25-year-old woman with recalcitrant keloid over the ear, back, hip, and chest received TCA injection following surgery)	TCA injection	Total of 1200 mg of TCA over 4 weeks at 2-week interval	Cushing's syndrome (presented 6 months after treatment with amenorrhea, moon face, hirsutism, and multiple reddish striae over the trunk).
Margaret Shanthi FX et al., 2008 [12]	Randomized study (54 keloid from the face, ears, trunks, and extremities randomized equally to receive verapamil and TCA)	Verapamil vs TCA injection	40 mg of TCA against 2.5 mg of verapamil injections every 3 weeks for 6 months.	Hypopigmentation, irregular menstrual cycles and profuse sweating with TCA. None specified with verapamil.
P.Kumar, S.Adolph, 1998 [13]	Case report (14 years old girl with recalcitrant keloid post burn scar over the left shoulder given TCA injections)	TCA injection	40 mg of TCA injection given twice in 4 weeks; following recurrence after 6 months after 4 cycles of 40 mg of TCA	Ulcer over keloid scar, linear hypopigmentation running along the subcutaneous vein (regression seen after 9 months of treatment discontinuance).
Abdul Fattah AM, 1976 [14]	Case series (a 17-year-old girl with presternal keloid post excision and a 20-year-old girl with presternal keloid following scalding)	TCA injection	10 mg/ml of TCA (dose unspecified) 4 weekly doses of 10-mg TCA	Burning sensation at the time of injection, breakdown of skin with ulcer followed by necrosis 1 week after. Cushingoid manifestation of a puffy face, hirsutism, painful pink stria over breast and flanks,and scanty menstruation.
Francisco Miguel Camacho-Martinez et al., 2013 [15]	Prospective study (37 keloids on trunk and extremities)	Bleomycin and TCA injection	4 mg of TCA injection every 3 months for 1–2 years In combination with 0.374 IU of bleomycin	Pain following bleomycin. Skin atrophy, necrosis at central quadrants in large keloids, telangiectasia postulated due to the combined effect.
Peter C. Amene, 1983 [16]	Case report (a 19-year-old man with large keloid over neck secondary to laceration wound)	TCA injection	Total of 7 injections of 40–80 mg of TCA over 12 months	Activation of pulmonary tuberculosis (patient was asymptomatic with clear chest x-ray prior to TCA injection; however, coincidental occurrence was conjectured by the author).

Complications encountered post steroid injection are generally mild such as skin hypopigmentation, hyperpigmentation, pain, pruritus, skin atrophy, and telangiectasia [17]. Nonetheless, severe reactions such as anaphylaxis following TCA injection over keloid scar have been reported [8]. Another unusual yet evidently significant complication is Cushing's syndrome secondary to TCA injection as few cases have been reported. These cases involved both paediatric and adult patients who received multiple injections to keloid on trunks and limbs [9–11]. Plausible reason in children could be the high body surface area facilitating higher systemic absorption causing adrenal insufficiency. Despite the different anatomical site association with systemic absorption of steroids and Cushing's, it is still imperative to be aware of the similar possibility in ear keloid particularly in children. There was no reported case of auricular keloid steroid treatment causing adrenal suppression leading to cushingoid manifestation. This may be due to the preponderance to larger keloid formation more at the trunk and extremities in children due to scalding, bumps, and falls [18]. Moreover, auricular keloid is relatively smaller and may be managed expectantly due to high rates of recurrence or reserved for surgery at a much older age.

Studies on intralesional injection treatments have also compared TCA with various other agents such as verapamil, bleomycin, and etanercept [19]. TCA was noted to cause more complications, particularly skin pigmentary changes and pruritus. In addition, adverse reactions such as profuse sweating and irregular menstrual cycle were also observed in patients injected with TCA in a study by Margaret Shanthi et al. [12]. An occurrence of necrosis followed by gangrene due to TCA is thus far only reported twice [13, 14]. However, partial necrosis followed by an ulcer was documented in patients in two prospective studies using a combination of bleomycin and TCA injection and another with radiation [15, 20]. One salient point that can be derived from this review is the occurrence of morbid complications such as those involving keloid formations over the trunk and extremities. There was no serious adverse event encountered from steroid injection to auricular keloid up to now [21]. Perhaps the absence of such reactions may be attributed to correct dosage, regime, and technique. The standard practice observed from this review was an intralesional administration of 1 to 2 cc of 10 to 20 mg/ml of TCA given monthly until resolution.

An analysis of the response of keloids to triamcinolone in various regions of the face showed a glaring failure only among lesions of the pinna [22]. These were noticed to be predominantly pedunculated. Complete absorption of the contents of a pedunculated lesion may be made difficult by the relatively narrow stalk which serves as a bottleneck. This may explain the 100% persistence observed despite shrinkage in size among all other treated lesions [23]. Other contributing factors might be the sudden increase in pressure after TCA injection, disrupted the blood supply by compressing vessels resulting in ischemia. This is supported by a study which elucidated on the severe ischemic and avascular central portion of keloids [24].

This unusual complication albeit logical given the possible mechanism raises the need for awareness to the caregiver, thus ensuring thorough and precise explanation of treatment options and known complications. On that note, possibilities of morbid complications such as gangrene, anaphylaxis reactions, and Cushing's syndrome especially in the paediatric population as reported in literature shed the need for a more stringent look into the management of keloid especially from the medico-legal aspect to ensure awareness on both parties.

Conclusions

Pinna keloid can be managed by a variety of surgical and non-surgical modalities. Intralesional TCA injection is one of the most common modalities of treatment either alone or in combination with others. Significant complications of TCA injection such as gangrene, anaphylaxis, and Cushing's syndrome, albeit rare, are extremely important to watch out for, as the associated morbidity can be troublesome. In counselling and obtaining consent from patients with pinna keloid, the option of TCA intralesional injection should be provided along with a thorough explanation of its known associated complications.

Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no conflict of interest.

Ethical Approval Not required for literature review and case report.

Informed Consent Written informed consent obtained from the patient.

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