

Pigmentary Complications after Non-Medical Male Circumcision

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Abstract

Objectives: A wide spectrum of complications are reported after male circumcision (MC), the non-aesthetic complications are well known, but the pigmentary complications scale are not reported precisely.

Methods: This is a prospective cohort study of 550 circumcised boys, who were examined and appropriately investigated for the incidence of pigmentary complications after circumcision. Most diagnoses were clinically, but dermoscopy was done for 17 case and a skin biopsy for 14 cases. Patients with personal or family history of vitiligo, or congenital nevi were excluded. Available hospital records details and parents' statements were revised. The main outcome measures are the incidence of different pigmentary complications and circumcision details; data were analyzed by Fisher's exact probability test, two tailed, and non-parametric tests including the Mann-Whitney U test.

Results: 69 cases had 72 confirmed pigmentary complications discovered at 2 to 36 months after commencement of circumcision (mean 18). 48 cases had pigmentary complications directly related to MC, 11 cases were probably related and 10 unrelated to MC. The most common lesion is the circular hyperpigmented scar (29 cases); liner hyperpigmented scar in 13, spotted exogenous melanosis in 18 cases, melanocytic nevi (7), hypopigmentation diagnosed in 3 cases, but kissing nevus is the rarest finding (2). Topical corticosteroid was tried in 15 cases, surgical excision of pigmented scar were done for 19 cases, local laser used for 4 resistant cases and reassurance with follow up for the rest.

Conclusion: Pigmentary complications after male circumcision are not rare and its management is challenging.

Introduction

Male circumcision (MC) is usually practiced for many reasons as social, religious, or cultural, but rarely for medical reasons. This procedure is considered one of the oldest and most common surgical procedures practiced globally, recently the MC rate across several countries is declining. This declining rate may reflects the changes in demographic patterns and parental believes that raised by studies in psychology and ontogeny ¹. There is a wide spectrum of post-MC complications. Abnormal penile pigmentations are frequently affecting men's physical and psychological wellbeing, they develop slowly and can be very extensive with late effects on sexual function and self esteeming. The incidence and prevalence of post MC pigmentary complications are unknown, and there is a clear evidence gap in the literature with only a few case reports describing these pigmentary complications after MC ². Mostly, genital hyperpigmentation is constitutive and racially variable. However, facultative pigmentation induced by the sun rays is rarely relevant in the genital area, but post-inflammatory hyperpigmentation is common. Linear hyper-pigmentation along the median raphe, of the ventral penile shaft, is usually seen as normal. Melanocytic lesions are considered the cause of genital pigmentation, but also post-inflammatory hyperpigmentation that occurs after an inflammatory dermatosis as lichen planus is much more commonly encountered ³. Genital melanosis is characterized by increased pigmentation of basal keratinocytes and melanocytes, but there is no increase in melanocyte number. If, however, melanocytes are present in increased numbers, the term 'genital lentiginosis' is more appropriate, genital freckles are usually seen as one or more brown punctate macules on the skin of the genitalia ⁴. Minor cases of abnormal genital hyperpigmentation may only need patient or his parent's assurance and follow up. Local corticosteroids may improve some cases, others may be ablated by laser therapy, but a disfigured pigmented scar deserves excision, especially if it is associated with bending or skin bridging over the glans ⁵.

Patients And Methods

A systematic genital examination was performed for all children and adolescents attending the outpatient clinic for genitourinary diseases or MC follow up; from June 2016 to March 2020. The examination commenced sequentially for the circumcision scar, the coronal sulcus, the frenular remnants, the penile shaft, the glans, and the urinary meatus.

A systematic general examination was done for detection of similar other pigmentary or vascular lesions, especially in the buccal mucosa. Examination of the scrotum and scrotal contents and palpation of inguinal lymph nodes also conducted. Onset and progression of the pigmentary lesions, previous use of drugs, or other remedies were reported. A detailed history of the circumcision was obtained concerning the timing of MC, method, type of anesthesia, bandage (type and for how long applied), previous use of drugs, and progression of post MC complications; type and management were reported. Dermoscopy done for 17 cases and a punch or excisional biopsy with the histological examination was done for doubtful and suspicious cases (14/69). Minor cases of linear and/ or circular scar hyperpigmentation are only followed up for 6 months for any progression or regression. Topical corticosteroids tried for cases with anxious and worried parents, surgical excision done for cases associated with other MC complications; like skin bridges or cicatricial phimosis, and neodymium:yttrium-aluminum-garnet (Nd: YAG) laser technique is used effectively in 4 cases at or above the age of 8 years with extensive lentigo and nevi. Data were analyzed by Fisher's exact probability test, two-tailed, as well as non-parametric tests including the Mann-Whitney U test. P-value less than 0.05 was considered significant.

Results

Total cases examined were 550 males aged from 6 months to 14 years (mean 5 years), and 69 cases (12.5%) had 72 confirmed pigmentary complications (three of them had combined lesions), all PC were discovered at 2 to 36 months after commencement of circumcision (mean 18 months). Parents are aware of the existence of PC in only 28 cases, 12 patients were mindful about the lesion himself, but 29 cases were detected anciently during the study.

The most common lesion detected is a circular hyperpigmented scar proximal to the coronal sulcus (n = 29); eighteen of them had high loose circumcision scar, (Fig 1) 4 had a redundant excess prepuce and two of them had a full picture of cicatricial phimosis, and 5 of those cases had variable degrees of skin bridges between the penile shaft and the glans.

Another 13 cases had a linear pigmentation along the glans penis or the shaft of the penis, (Fig 2) or a combination of circular scar hyperpigmentation and a scared hyperpigmented ventral surface of the penile shaft, glanular hyperpigmentation is commonly seen around the meatus (Fig 3A), different sizes of melanocytic nevi (glandular melanosis) on the glans or corona diagnosed in 7 cases (Fig 3B-C), multiple small dark melanocytic nevi at the circumcision scar diagnosed in 2 cases (Fig 4A). Different forms of a raised exogenous hyperpigmentations diagnosed in 9 cases, (Fig 4B), combined lesions are not rare (Fig 4 C). One case had balanitis xerotica obliterans (BXO); diagnosed histologically and presented with a hyperpigmented glanular scarring with skin bridges (Fig 5). Different forms of hypopigmentation are diagnosed in 3 cases (Fig 6), but kissing or divided nevus is the rarest complication (2 cases) (Fig 7). History of post MC meatitis and different forms of balanitis subjectively confirmed in 23 cases, minor post MC bleeding in 6 cases, circumcision redo in 6. All cases reported that the child had a gauze bandage after the procedure, and in 41 cases the bandage was left for more than one day and the rest had an overnight bandage.

As much as the parents could not remember any abnormality or associated pathological conditions existed before circumcision, also all of them denied any similar pigmentary lesion detected or reported prior to circumcision. 45 cases with PC were circumcised in the 1st month of life; 32 of them at the 1st week, 14 at 6 months, and 10 after 12 months

of life. The circumcision methods in most cases were the bone cutting clamping guillotine method (42/69) under local anaesthetics in 28, but without anaesthesia in 14 cases, Gomco clamp method used in 18, and plastibell used in 9 cases.

Betadine solution (povidone-iodine 10%) is an antiseptic non-sterile solution used by the mother for dressing the circumcision wound in 85% of the cases, which were accompanied with a topical antibiotic ointment in 65% of the cases, in another 10 cases the family was unacquainted about the method of post MC dressing. Topical corticosteroid was tried as management for minor PC with partial success in 15 cases, surgical excision of pigmented disfigured scar were done for 19 cases, and one session of Nd: YAG local laser used for 4 cases with a complete success in three of them. Only reassurance and follow up applied for the rest of the cases.

Discussion

Circumcision procedure may be undertaken for modification of the genitals, to change the appearance of the penis and to appeal to more aesthetics. Despite that, it may leave a permanent change of the natal characteristics of the genitalia. Pigmentary complications may occur as a result of non-aesthetic preputial cutting or the untidy healing of the circumcision wound ⁶.

Pigmentary complications may appear after months or years of the procedure. As well, it may result in early family dissatisfaction and affect the psychological impact of a man's contentment with his penis and may lead to loss of self-esteem ⁷.

Genital skin is relatively thin when compared with other sites, and the balanopreputial epithelium is incompletely keratinized, causing enhanced susceptibility to damage from irritants, resulting in several pigmentary complications, which may be transient or permanent, localized or segmental, or more rarely a combined lesions ⁸. Cullen ⁹ proved that about 9% of males have at least one acquired melanocytic nevus during the lifetime in the genital area. Genitalia pigmentation is particularly influenced by sex hormone impregnation before birth, which leads to the normal dark color of the scrotum and median raphe. Other factors may partly explain hyperpigmentation as friction or chronic irritation which may be manifested later on after puberty.

Hyperpigmentation caused by post-inflammatory conditions may coincide with an accelerated stimulation of genital skin melanocytes that leads to an increase in the melanin concentration in basal and suprabasal layers. Mediators as cytokines and growth factor which stimulate the melanocyte released by inflammatory cells of the epidermal infiltrate ¹⁰.

Postinflammatory hyperpigmentation may leaves a scar that is darker than the surrounding unaffected skin. The mechanisms behind this are not fully understood but may involve activation of melanocytes by inflammatory mediators or reactive oxidative species released by damaged skin ¹¹. Also, additional melanin is produced in the response to injury and this is engulfed by macrophages which are involved in the inflammatory response. The macrophages then retain the melanin in the dermis until the cells and melanin are degraded, enabling the pigment to persist within the dermis for some time post-injury, increased and prolonged activation of melanogenesis within these lesions may rarely lead to uncontrolled proliferation and melanoma formation later on ¹². Certain factors are predisposing to a poor cosmetic appearance that following MC, these factors including insufficient hemostasis, failing to recognize anatomical diversities or abnormalities. Also, using thick sutures with long absorption time, impertinent tissue handling, too tight dressing, and excessive resection of prepuce are considered ¹³. Generally, the two-toned penis and penile pigmentation variations are not rare among circumcised men, wherein the penis has two distinct

colours, often divided by a circular circumcision scar usually proximal to the coronal sulcus. This two-toned penis may be considered ugly or of aesthetic concern for some men. In a survey completed by the National Organization of Restoring Men in the UK; 74% of the respondents were dissatisfied with the appearance of their circumcised penises, and particularly, 26% complained about the variation in skin color, in each case, the circumcision scar is clearly visible, this scar, likely not of concern except after adulthood¹⁴. Freckles are commonly detected obviously at the glans penis, after MC, but rare forms are difficult to diagnose and may mislead with other venereal diseases. Yazdanpanah et al. reported that genital warts were the most common disease diagnosed after MC among their cases, but inflammatory dermatoses such as psoriasis and balanitis were less common compared with other studies¹⁵. A lentigo is a small pigmented spot on the skin with a clearly defined edge, surrounded by normal-appearing skin, it is benign hyperplasia of melanocytes that is linear in its spread. This means the hyperplasia of melanocytes is restricted to the cell layer directly above the basement membrane of the epidermis where melanocytes normally reside. This is in contrast to the "nests" of multi-layer melanocytes found in moles (melanocytic nevi). Stromal melanophages can occasionally be seen, especially in penile lentiginosis¹⁶. Lentigos are distinguished from freckles based on the proliferation of melanocytes, freckles have a relatively normal number of melanocytes but an increased amount of melanin. Penile lentiginosis is characterized by the presence of multiple hyperpigmented small-to-medium-size lesions with uniform or variegated pigmentation¹⁷.

Diffuse hyperpigmentation which resulting from chronic inflammation or postinflammatory conditions may be presented as multiple macules that giving a 'spotty' appearance to the skin of the genitalia. A remarkable pigmentary loss in vitiligo occurs mainly in patients with dark complexions, and it may be overlooked in people with fair skin. Characteristically, it may appear as white patches on the glans penis, but spontaneous repigmentation has been known to occur. Hypopigmentation associated with postinflammatory conditions may be seen after any form of dermatophyte infection, genital ulcer, intertrigo, or chronic dermatitis¹⁸. In this series of patients vitiligo is diagnosed in only 3 cases (Fig 6)

Divided or kissing nevi, mostly located in the genitalia, are very rare and can only be seen on parts of the body where the separation occurs during embryogenesis. Commonly, these nevi are believed to be benign pigmented tumors. On the other hand, malignant transformation has not yet been reported¹⁹. Divided naevus has been reported with one component located on the dorsal or dorsolateral side of the glans and the other on the distal penile shaft or inner face of the prepuce, and separated by uninvolved skin across the coronal sulcus. It has been hypothesized that melanoblasts migrate to the lesion before the embryological separation of epithelial preputial placode²⁰. It is difficult to incriminate circumcision as an etiology of such type of nevus, our finding of 2 cases of divided nevus may be a coincidental diagnosis. Penile melanosis had a particular clinical and microscopic appearance characterized by pigmented macules plus basal layer hyperpigmentation. It may be associated with or without melanocytic hyperplasia. Some patients with penile melanosis are associated with local irritation, the injuries as occurred with circumcision, or diabetes mellitus²¹. Post MC pigmentary complications can be differentiated from cases of fixed drug eruption, which may occur 1 to 2 weeks after initial exposure to oral medication. These lesions are mainly solitary, violaceous inflammatory plaques located on the penile shaft or glans. Also, these lesions have clear borders and may blister or eroded. Furthermore, lesions may be associated with pain or pruritus and heal with time but may leave hyperpigmented patches. The circumcision procedure inherently limits superficial vascular supply either through the mechanism of local vasospasm induced by the preputial excision itself, or alternatively, the pressure or tumescence of the homeostatic dressing wrapped around the penile shaft following MC which may hinder the blood supply to some degree²².

This cohort study was carried on all circumcised boys attending the outpatient clinic during the period from June 2016 to March 2020. Common pigmentary complications diagnosed in this group of patients are circular and linear hyperpigmentation proximal to the coronal sulcus, which gives the penis the look of the two-toned penis (Fig 1). In 48 cases the pigmentary complications are proved to be directly related to MC, linear, circular and freckles hyperpigmentation are the common lesion in this group, also the hyperpigmentation around the urinary meatus definitely acquired after circumcision. 11 cases the were probably related to circumcision; and the common lesions of this group is the hypopigmentation and patients wit multiple small dark melanocytic nevi. In 10 cases the lesion is unrelated to MC; mainly the patients with dividing nevus. Overall the most common lesion is the circular hyperpigmented scar (29 cases); liner hyperpigmented scar in 13, spotted exogenous melanosis in 18 cases, melanocytic nevi (7), hypopigmentation diagnosed in 3 cases, but kissing nevus is the rarest finding (2).

Topical corticosteroid was tried in 15 cases, surgical excision of pigmented scar were done for 19 cases, local laser used for 4 resistant cases and reassurance with follow up for the rest.

Treatment of genital pigmentary lesions depends on the type of the nevi, its location, and abnormal features detected in the dermatoscopic examination. Penile melanosis is generally harmless and does not require treatment, but some people may choose to have cosmetic procedures to remove the spots. In hyperpigmented lesions, melanosomes are destroyed by the laser, preventing further melanogenesis and melanin transfer. Laser therapy can achieve good results but may require multiple treatments²³.

In this study the prolonged dressing for 2 or more days is significantly associated with hyperpigmentations (60% of the cases), using 10% povidone-iodine is significantly associated with PC; especially circular and linear hyperpigmentation, also post MC infection in the form of balanitis, meatitis, or BXO are significantly associated with PC (24/69); P-value less than 0.05.

This study intends to arouse awareness towards this hitherto of unrecognized complications, better knowledge regarding it may assist in formulating an educated approach towards this complication, in order to employ preventive measures to eliminate it or at least provide reassurance wherever it is possible.

Study limitations: Lacking previous documented similar observations of these findings postulate that either inherent qualities of the penile skin or external factors induce the damage to the penile soft tissue are responsible for such complications.

Conclusion

Post MC penile hypo or hyperpigmentation may appear after several conditions affecting the penis; like balanitis, secondary to topical medications applied after MC or a tight bandage, which may have been the cause of the spotty pigmentation of the glans. Biopsy and histology are indicated when history is unclear or the causative agent is no longer present.

Abbreviations

Male circumcision (MC)

Pigmentary Complications (PC)

Neodymium

yttrium-aluminum-garnet (Nd:YAG)

Balanitis Xerotica Obliterans (BXO).

Declarations

Disclosure of potential conflicts of interest: Authors state that there is no any conflict of Interest in this research.

This research not involving human or animals participants.

Informed consent: Parents of all patients gave a written consent for photographing their children and they also consented use of those photos for publication.

Statement on consent for publication: All authors consent publication of this manuscript in the World Journal of Urology, and the corresponding author is responsible for contact with the editorial board of the journal.

Statement on Ethical approval and informed consent: This paper was approved by the ethical committee of Faculty of Medicine for Girls, Al-Azhar University. The written approval is available upon request.

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Figures



Figure 1

Circular hyperpigmented scar proximal to the coronal sulcus in a high lose circumcision scar.



Figure 2

A linear hyperpigmented scar with a skin bridge over the glans.

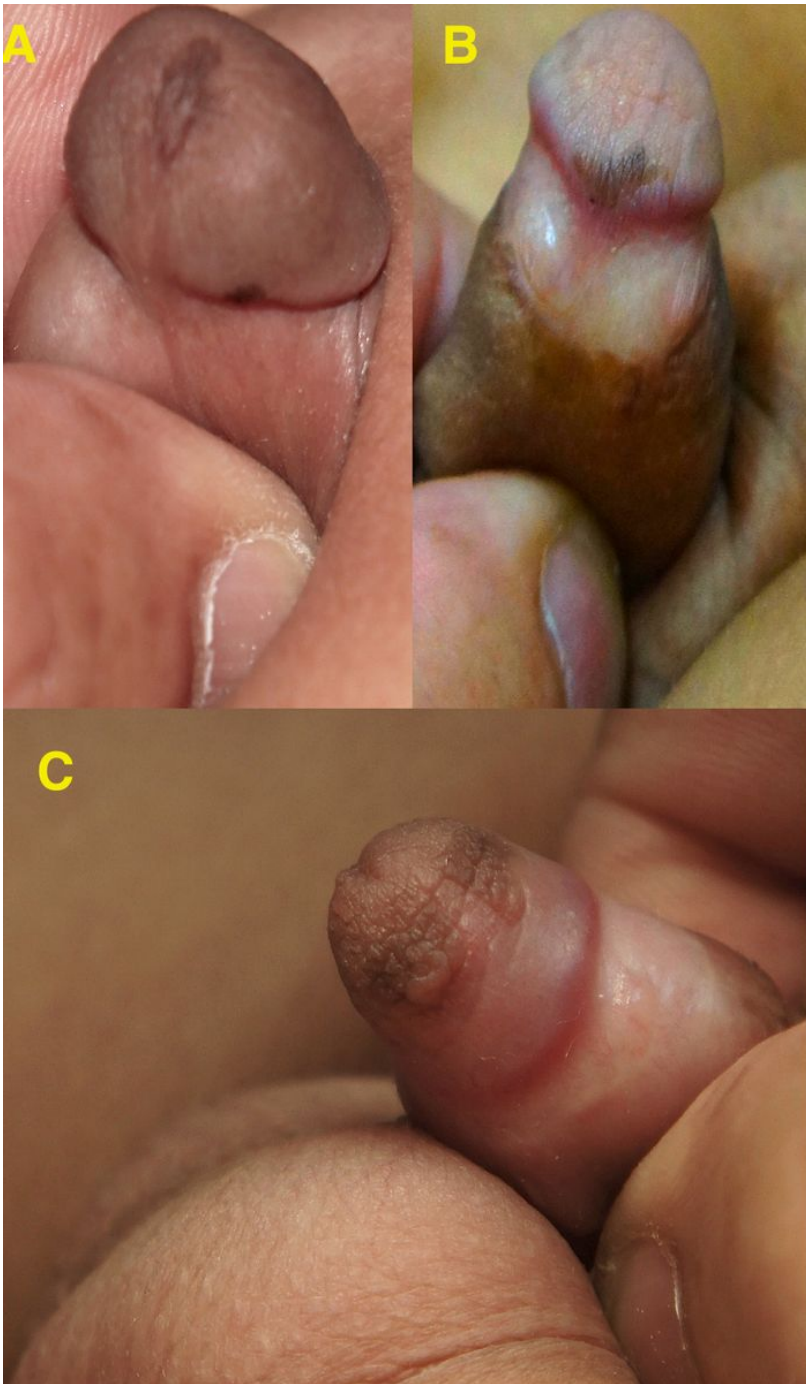


Figure 3

(A) Hyperpigmentation around the urinary meatus, which may be secondary to meatitis. (B) Proximal nevus at the dorsum of the glans near the coronal sulcus, and (C) larger nevus over the glans.

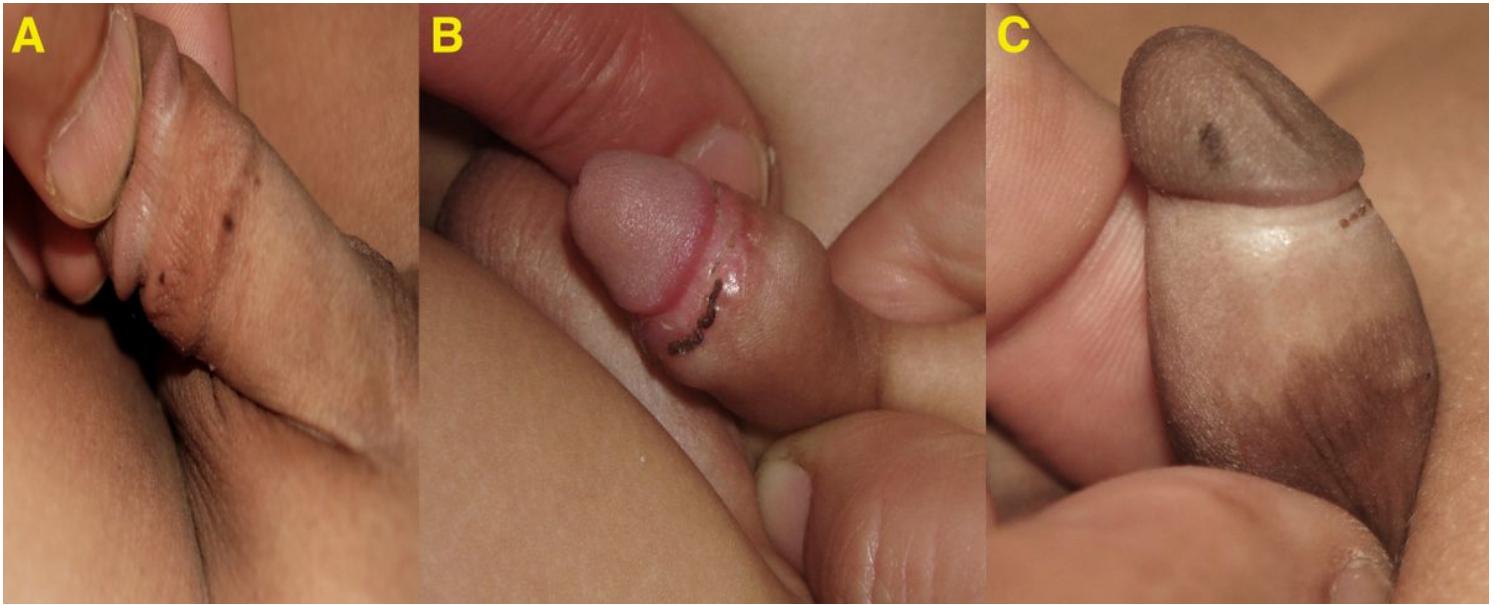


Figure 4

(A) Multiple small dark melanocytic nevi at the circumcision scar. (B): Hyperpigmented raised exogenous hyperpigmented scar at the preputial remnants. (C): Combined glandular melanosis, exogenous hyperpigmentation and circular preputial remnant hyperpigmentation.



Figure 5

Post MC balantitis xerotica obliterans (BXO) presented with hyperpigmented glans scaring.



Figure 6

Hyperpigmented glans and proximal penile shaft with a distal distinctive hypopigmentation.



Figure 7

Kissing nevus at both the glans and the preputial remnants.