Complications of suture ligation ablation for ulnar polydactyly: a report of two cases

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Abstract We report two cases resulting in complications following suture ligation treatment for ulnar polydactyly. One case consisted of bilateral, retained gangrene and cellulitis, and a second case consisted of a residual, sensitive skin tag. The case involving gangrene and cellulitis developed after an unsuccessful suture ligation of bilateral pedunculated duplicated digits. The second case developed after suture ligature ablation of a rudimentary digit in the nursery but presented 3 years later with a residual symptomatic nubbin. Both cases were treated by surgical excision of the residual tissue in the operating room. The first case illustrates a morbid complication following unsuccessful ligature while the second case demonstrates the inevitable suboptimal long-term outcome associated with what has traditionally been considered “successful” suture ligation.

Introduction

The management of ulnar polydactyly varies depending on the type of duplication, size, and residual tissue in the underdeveloped digit. Suture ligation at the base of duplicated ulnar digits has traditionally been the most

common treatment implemented for nonfunctional, poorly formed digits lacking proximal bony elements. This procedure is often done in the nursery shortly after delivery. The intended result of the procedure is to produce necrosis of the rudimentary digit followed by autoamputation of the gangrenous extra tissue. Proponents of this management cite its low cost and simplicity [7, 20]. In recent years, the practice of suture ligation has been questioned by several authors who cite associated complications and the frequency of incomplete amputation or painful neuroma [5, 14, 18]. The purpose of this communication is to report two cases that support the contention that duplicated ulnar digits might best be treated with formal excision in the operating suite.

Case 1

A 2-week-old infant was referred for evaluation of both hands after suture ligation of bilateral ulnar duplications that were done in the nursery immediately after birth. The infant was otherwise healthy and there was no family history of congenital hand anomalies. Eighteen days after ligation, the right duplicated digit remained firmly attached and was grossly edematous, inflamed, ischemic, and purple but not necrotic (Fig. 1a). The suture ligature that had previously been placed at the base of the digit had fallen off several days prior to presentation.

The left duplicated digit was dry and necrotic but still firmly attached to the ulnar border of the hand which was erythematous and inflamed (Fig. 1b). The suture ligature on this side had remained in place and was tied firmly around the base of the lesion. At surgery, the operative findings on the right side were consistent with ischemia and venous congestion without obvious necrosis. On the left side, the

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Fig. 1 **a** Right hand demonstrates purple, edematous-congested, and ischemic-duplicated small finger. Suture ligature had fallen off several days prior. **b** Left hand showing dry, necrotic duplicated digits with suture ligature at its base

skin at the base of the necrotic residual digit had developed erythema consistent with evolving cellulitis. Elliptical incisions were made around the base of both duplicated digits, and they were excised along with the raised margins. Electrocautery was required to control bleeding and the skin was then easily approximated with catgut suture. The patient's wounds healed uneventfully and the erythema and cellulitis resolved within the first postoperative week.

Case 2

A 3-year-old child with Antley–Bixler syndrome was referred for evaluation of bilateral radiohumeral synostosis and a skin tag at the ulnar border of the right most ulnar digit (Fig. 2). In addition to elbow synostosis, Antley–Bixler syndrome includes cranial synostosis and upper extremity anomalies. This patient was also diagnosed with ulnar dysplasia of the right forearm and hypoplasia of the right fifth finger. At birth, the fifth digit was round, rudimentary, pedunculated, and nonfunctional. It was ligated with a suture in the early postnatal period, and the digit subsequently auto amputated; however, as the child matured, her mother noticed that she had sensitivity and some discomfort associated with a residual 4–5-mm ulnar skin tag, and the mother considered the nubbin to be aesthetically unacceptable. She eventually requested excision of this lesion. In the operating suite, an elliptical incision was made around the base of the skin tag, and the lesion was fully excised. Electrocautery was required to control bleeding from a main artery and the skin was then easily approximated with catgut suture. Postoperatively the wound healed satisfactorily.

Discussion

Ulnar polydactyly is a one of the most common congenital anomalies and involves the ulnar side of the hand with

duplication of the small finger [8, 11]. Its prevalence is one in 531 live births but varies from one in 143 black infants to one in 1,339 white infants making it roughly ten times more common among black infants [20]. While ulnar duplication may be associated with an underlying syndrome, it is most often an isolated finding, particularly in African Americans where it typically assumes an autosomal dominant mode of inheritance and is bilateral in 70% of cases [1, 20].

Ulnar duplication comprises a spectrum of anomalies ranging from a rudimentary skin tag on the ulnar border of the hand to a fully formed and functional duplicated digit. The simplest classification of ulnar polydactyly was proposed by Temtamy and McKusick [19]; type A duplication consists of a well-formed duplicated digit articulating with the fifth or a sixth metacarpal. Type B duplication is nonfunctional, consisting of either a rudimentary tag or larger, pedunculated digit with the proximal skin bridge lacking bony elements. The latter is the most common form representing approximately 80% of all cases [15, 21]. A more comprehensive classification has been proposed by Rayan and Frey [15] (Table 1). This classification accounts for the small, rudimentary skin tags often observed on the ulnar aspect of the hand, which many authors believe represent the sequelae of pedunculated duplications that have undergone in utero autoamputation. The histopathology of these lesions is identical

Fig. 2 The ulnar border of hand showing skin tag following previous ligature ablation of pedunculated digit in nursery

Table 1 Rayan–Frey classification of ulnar polydactyly (2001)

I	Soft tissue nubbin (wart-like)
II	Pedunculated nonfunctioning digit (lollipop-like)
III	Well-formed functioning digit articulating or fused to the fifth metacarpal
IV	Complete ray duplication with a separate sixth metacarpal
V	Miscellaneous anomalies such as polysyndactyly

to that seen in residual skin tags following postnatal suture ligation [2, 11, 12, 16].

Treatment of ulnar polydactyly falls into the categories of observation, suture ligation in the early postnatal period, excision in the office with a local anesthetic, and formal excision in the operating suite with reconstruction as necessary. Rudimentary skin appendages (Rayan–Frey type I) may be simply observed as their primary detriment has traditionally considered to be cosmetic in nature; however, sensitivity of this lesion caused by neuroma especially after ligation has been documented [13, 14, 18]. It is also not uncommon for the infant to chew on or abrade the skin tag resulting in skin breakdown. The traditional treatment for type II (pedunculated) digits has long been suture ligation in the nursery. This results in necrosis of the distal tissue and autoamputation usually between 10 and 21 days [20]. Several complications have been associated with this treatment, and it has recently come into question. Appropriate treatment of well-formed type III, IV, and V digits, is always carried out in the operating suite and typically requires reconstruction of the adjacent soft tissue in addition to ablation of the digit [4]. Treatment for type II lesions is controversial, yet for reasons illustrated by the preceding cases as well as more recent literature, we recommend formal excision of these lesions in the operating suite.

The first case we report describes a case of bilateral ulnar polydactyly treated with suture ligation at another facility. Both duplicated digits suffered complications associated with this method of treatment. In neither digit did suture ligation result in successful ablation at 2 weeks postligation. The appearance of both digits was distressing to the family, and one digit exhibited clinical manifestations of cellulitis. Our second case report describes a patient following “successful” suture ligation in the nursery whose family eventually requested excision of the resulting sensitive, aesthetically displeasing nubbin.

When suture ligation is employed, the digital nerves of the rudimentary digit are severed at the level of the skin and cannot retract into the soft tissues. This may result in a sensitive scar due to neuroma formation. Several published reports have identified the propensity for ligated digits to develop such neuromas [9, 13, 14]. Leber and Gosain identified three adult patients with five sensitive traumatic

amputation neuromas following suture ligation. Secondary surgical excision of the vestigial amputation stump confirmed the presence of neuroma by histology and successfully achieved relief of symptoms in all patients. In contrast, six patients with 12 type B duplications who underwent formal excision exhibited excellent cosmetic results without postoperative neuroma formation at long-term follow-up. Furthermore, seven of the nine patients in the study had affected family members, all of whom were treated previously with ligature ablation. When carefully examined, all of these patients had signs and symptoms consistent with residual amputation neuromas. Further studies have confirmed the presence of these amputation neuromas at the site of ligature ablation [10, 14, 16, 18]. In their review of 21 infants who underwent suture ligation for type B (pedunculated) polydactyly, Hennrikus and Watson noted that 43% of their patients had a residual “bump” at the site of ligation, but parents typically deemed the stump to be cosmetically acceptable [20]. Mullick et al. reviewed 13 cases in ten patients referred for complications following suture ligation. All of these patients (or their parents) were dissatisfied with the appearance of the residual digit and several complained of associated pain and sensitivity [14]. The recommendation was that suture ligation should be implemented with caution and should not be utilized with lesions having a bony or ligamentous attachment or for lesions with a base greater than 2 mm. Similarly, Frieden suggested that rudimentary digits with greater distal bulk (resulting in more necrotic tissue) and those with a wider pedicle are at an increased risk for infection, scarring, or incomplete removal and should be treated by a specialist [7]. Rayan and Frey reported a 16% rate of a cosmetically unacceptable or tender residual skin tag in a group of 105 patients treated with suture ligation mainly in the nursery [15].

Several less common, but potentially more concerning complications have been described. Watson and Hennrikus noted that the average time for a ligated digit to amputate was 10 days, but that some digits required as many as 21 days and one digit required surgical excision as it remained attached yet necrotic after 4 weeks [20]. Mullick also identified a patient in whom suture ligation ultimately failed to ablate the digit [14]. Obstruction of venous return and edematous enlargement, similar to that observed in our first case, has been described after “home treatment” consisting of digital ligature with suture [3] while rare, soft tissue infection associated with attempted ligation has also been described in the literature [7]. Rayan and Frey documented a 6% infection rate with this practice [15]. Finally, some authors have advised against suture ligation because of the remote possibility of exsanguination from an artery after separation of the ligature [6, 17].

As a consequence of the frequent early complications such as failure of separation, cellulitis, edema, and ischemia

as well as the late complications after separation such as residual sensitivity and suboptimal cosmetic results, we caution against suture ligation for the treatment of ulnar polydactyly. When treatment is requested for type II duplicated ulnar digits, we recommend formal operative excision as the optimal ablative procedure.

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

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