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Multiple hand anomalies with unusual dermatoglyphic pattern in a Nigerian female.

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Abstract
Congenital hand anomalies of the hand is a myriad of deformities resulting from gene mutation and intrauterine environmental insult occurring during the period of limb embryogenesis. Each entity may exhibit a peculiar dermatoglyphic pattern which is genetically predetermined.
During routine embryology practical demonstration, we came across a 20 year old female medical student with bilateral post axial polydactyly, bilateral incomplete simple syndactyly between the middle, ring and little fingers and non aligned ulnar border of the hand. Dermatoglyphic studies revealed abnormally high Total Finger Ridge Count (TFRC), abnormally high atd angle, occurrence of whorl & twin whorl patterns in all but one fingers and palmar crease lines on the thenar eminence, longitudinal to the thumb.
Knowledge of anomalies of the hand, their combination and genetic basis is important for improving the outcome of surgical correction and genetic counseling.
Keywords: Multiply hand anomalies, Dermatoglyphic patterns, Syndactyly, Polydactyly.

Introduction

Congenital anomalies of the hand encompass a myriad of deformities ranging from undergrowth, overgrowth, duplication or differentiation of limb parts to total absence (Swanson *et al*., 1968). The embryogenesis of the limbs occurs during 4^{th} to 8^{th} week of intrauterine life and the limb develops through development of limb bud, formation of apical ectodermal ridge, growth of the limbs and apoptosis aided separation of the digits (Moore & Persuad, 2008).
Exposure to teratogens during this vulnerable period may result in failure of separation of the digits (syndactyly) and formation of extra digits (polydactyly) (Moore & Persuad, 2008). Polydactyly is the most frequently observed hand anomaly (Castilla *et al*., 1998). It can occur in isolation or as part of a syndrome. It may be autosomal dominant or recessive in inheritance (Castilla *et al*., 1998). Polydactyly may be classified as preaxial, postaxial or central denoting an extra digit attached to the thumb, little digit and other digits respectively (Elliot & Evan, 2006). Based on the extent of webbing, syndactyly can be classified as complete or incomplete. The term simple syndactyly denotes soft tissue involvement while complex syndactyly denotes bony involvement in the webbing. Syndactyly involving the middle and ring finger is the most frequent while that involving the thumb and index finger is the least frequent (Kettelkamp & Flat, 1961; Nylew, 1957).
Palmar creases and finger prints are established during the very early weeks of gestation and remain unchanged throughout life. Specific dermatoglyphic patterns have been known to be associated with genetically predisposed diseases and congenital anomalies. (Floris and Marini, 1998, Ziegler *et al*., 1993, Suzuki, 1979).

Case Report

During routine embryology practical demonstration in the Department of Anatomy, University of Ibadan, Nigeria, we observed multiple hand anomalies and unusual dermatoglyphic patterns in a-20-year-old female medical student. Examination of the hand revealed a bilateral incomplete simple syndactyly between the middle and ring fingers, and between the ring and little fingers. The syndactyly ended proximal to the proximal interphalangeal joint (Fig. 1 & 2).
A rudimentary sixth digit was present on the ulnar side of both hands (postaxial polydactyl) (Fig 3). There was nonalignment of the ulnar border of the 5^{th} digit (line A) with the ulnar border of the rest of the hand (line B) (Fig 4). X-ray of the hand showed no bony involvement in the syndactyl (Fig 5). An incidental finding of a sesamoid bone located close to the base of proximal phalanx of the first digit of both hands was spotted on x-ray (Fig 5). There was perfect alignment of 5^{th} metacarpal bone with the phalanges of the 5^{th} digit on xray. Dermatoglyphic study using the ink method revealed a TFRC of 80 on the Right Hand (RH) and 84 on the Left Hand (LH), atd angle of 50^{o} (RH) and 52^{o} (LH) and a-bRC of 44(RH) and 41(LH) (Fig 6). The predominant digital ridge pattern observed was the rare twin whorl and whorl pattern which was present in all the digits of both hands except the middle finger (Fig 4).The digital pattern present in the middle finger was ulnar loop

bilaterally. An unusual palmar crease on the thenar eminence running in the direction of the longitudinal axis of the thumb was observed bilaterally. Normal palmar creases: radial longitudinal line, proximal and distal transverse lines were present in both hands of this subject. There was a history of bilateral polydactyly in the mother of the subject.

Discussion

Congenital hand anomalies have an estimated incidence of 2.3 per 1000 total births (Lamb and Loynne-Dave, 1998). Polydacyly and syndactyly ranked as leading congenital anomalies of the hand occurring in isolation or as part of over 300 syndromes. In this case, we reported a bilateral rudimentary polydactyly occurring alongside incomplete syndactyly and non-aligned ulnar border of hand and 5^{th} finger. The observed polydactyly type was post axial thus up holding the previously reported black predisposition (Glois *et al*., 2002). Polydactyly has been linked with mutation of 39 different genes located on chromosome 7q36 (Bisecker, 2002; Hang *et al*., 2005). Lu (2006) implicated the expression of fibroblast growth factor 4 and 8 on mouse limb bud in the etiology of polydactyly. The reported case has a positive family history of polydactyly, with mother having bilateral post axial polydactyly.

Isolated post axial polydactyly is transmitted through autosomal dominant inheritance with reduced penetrance while syndromic post axial polydactyly is transmitted via autosomal recessive inheritance (Castilla *et al*., 1998).

The syndactyly in the present case was observed between the middle and ring finger and between the ring and the little digit. This is in tandem with the previous findings that the spaces between 3^{rd} and 4^{th} fingers and 4^{th} and 5^{th} fingers are the most commonly affected, constituting 50% and 30% of the total web respectively (Waters and Bae, 2012). Webbing is due to failure of apoptosis of the inter digital cell columns (Moore and Persaud, 2008). Though no family history of syndactyly was present in this case, syndatyly between 3^{rd} and 4^{th} fingers can be inherited as an autosomal dominant trait (Rao and Bhavani, 2012). Webbing at this site has been reported to be due to mutation on chromosome loci 2q34-q36 (Rao and Bhavani, 2012).

The common digital ridge pattern observed among Igbo tribe in Nigeria was ulnar loop (Adetona *et al*., 2008). Ulnar loop was only present on the middle finger of our subject, who is a descendent of the Igbo tribe in Nigeria. All other fingers had twin whorls and whorl patterns. The observed TFRC (RH 80, LH- 84) and atd angle (RH-50, LH-52) were higher than the mean values reported for normal Igbo females in previous studies (Adetona *et al*., 2008). Adetona et al (2008) reported a mean TFRC value of 53.70 and mean atd value of 52^0 in normal Igbo females. Variation in palmar creases is under hereditary and environmental influence with the later acting only during intrauterine line (Aleksnadrowicz *et al*., 1996).

The observed oblique lines on the thenar eminence of both are a variant reported in a previous study on palmar flexion in Nigerians (Adetona *et al*., 2012). The X-ray of the hand showed perfect alignment of the fifth phalanx with the 5^{th} metatarsal bone. It can be hypothesized that the non alignment of the ulnar border of 5^{th} digit and that of the rest of the hand may be due to expansion of the thenar space rather than non alignment of the bones. Thenar space expansion may be due to hypertrophy of one or more of thenar muscles or/and the presence of an extra variant muscle. As the study was done on living subject, dissection of hand to ascertain the exact cause of thenar expansion was impossible.

Conclusion

Co existence of polydactyly poses a functional, cosmetic and psychological challenge which requires a multidirectional approach in management. Knowledge of congenital anomalies of the hand, their combination and genetic basis is important for improving the outcome of corrective hand surgeries.

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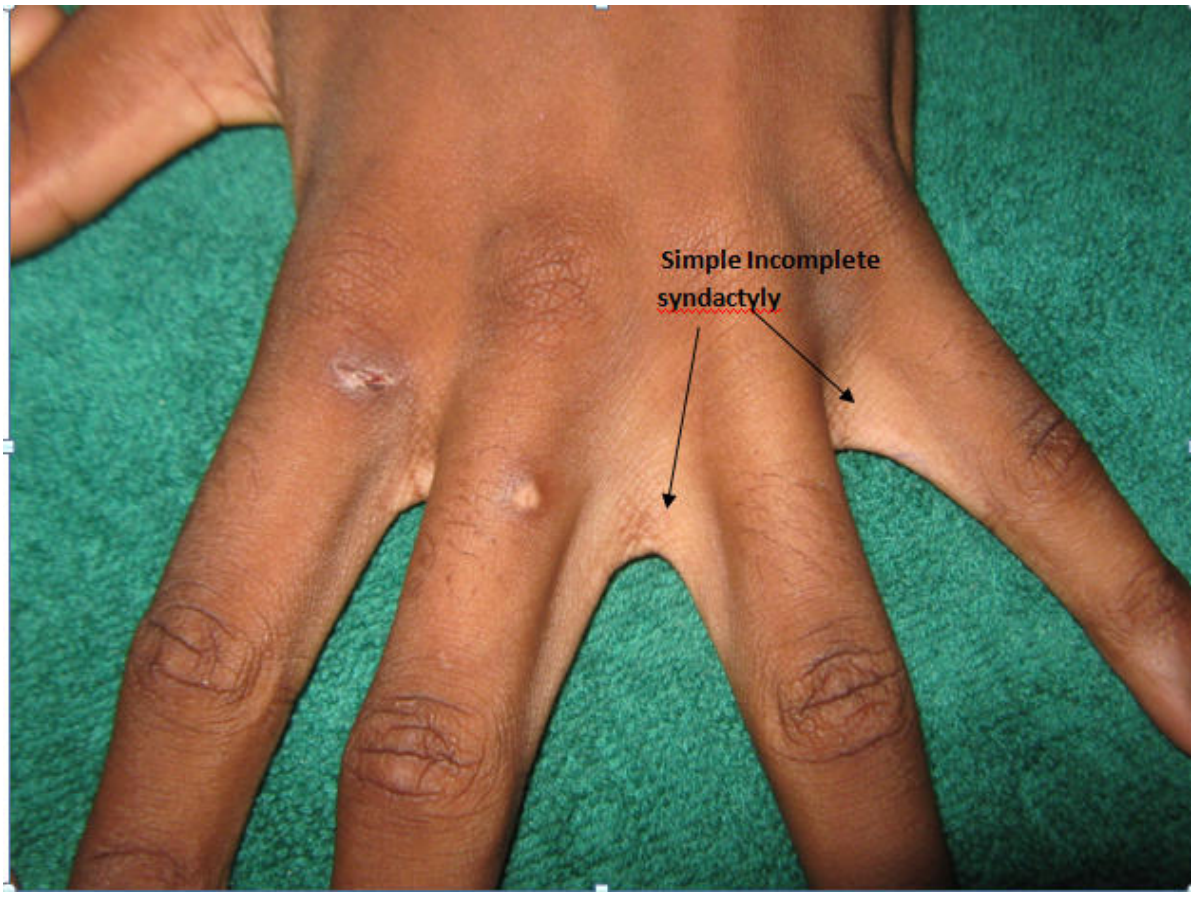

Fig 1: showing simple incomplete syndactyly on the left hand

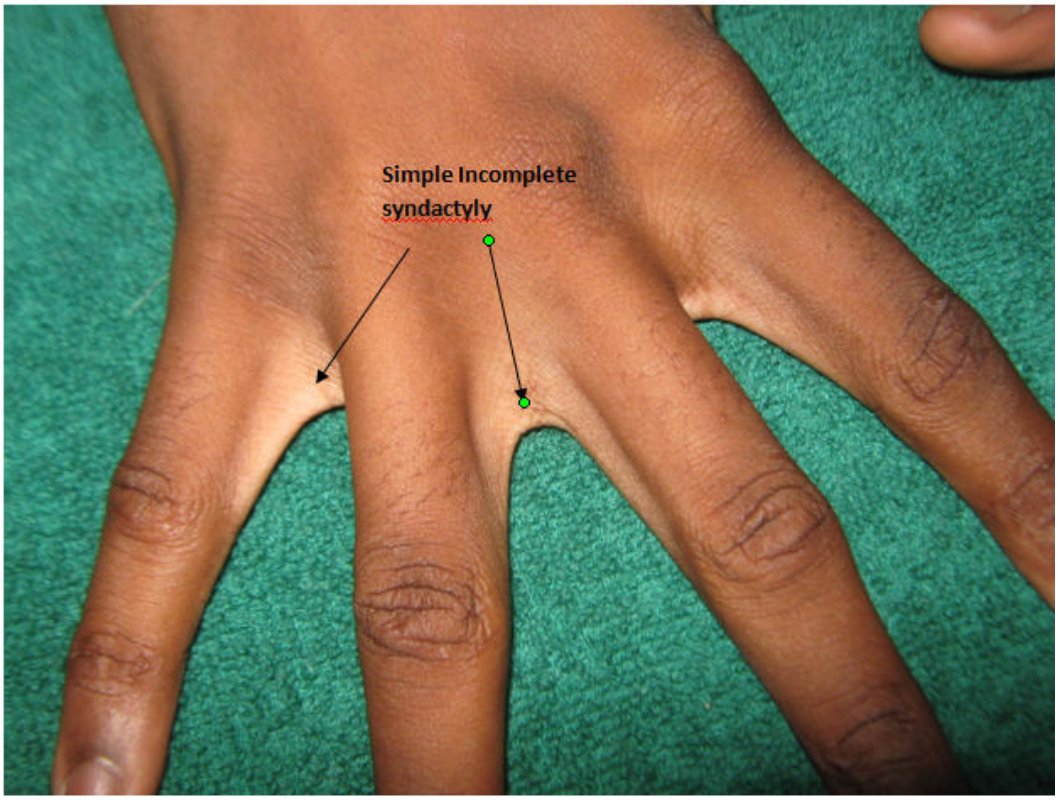

Fig 2: showing simple incomplete syndactyly on the right hand

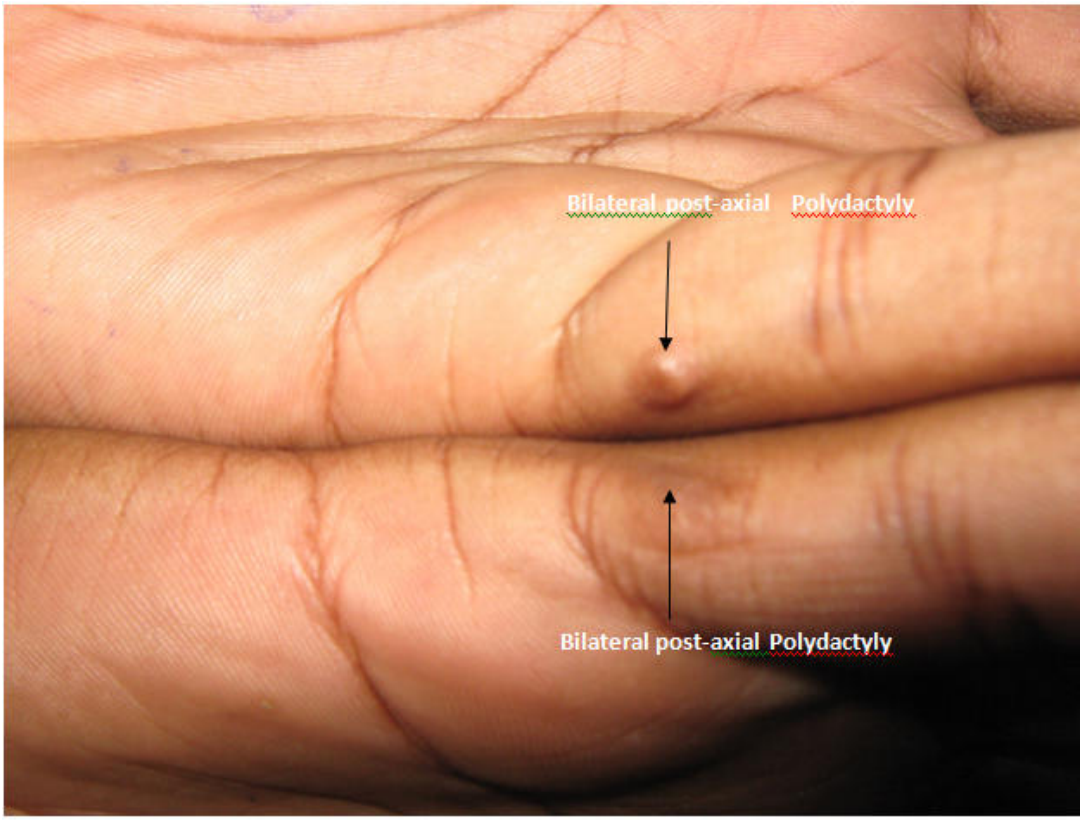

Fig 3: showing rudimentary bilateral post-axial polydactyly on both hands

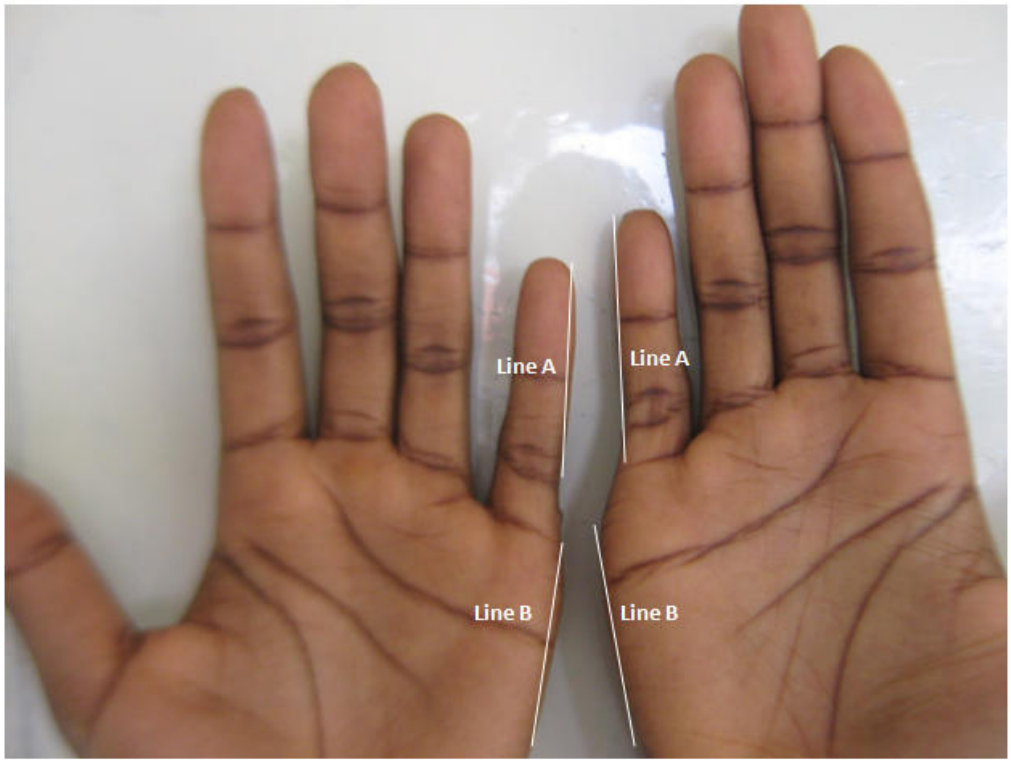

Fig 4: showing non-alignment of ulnar borders of the 5th digit (line A) and ulnar border of the rest of the hand (line B).

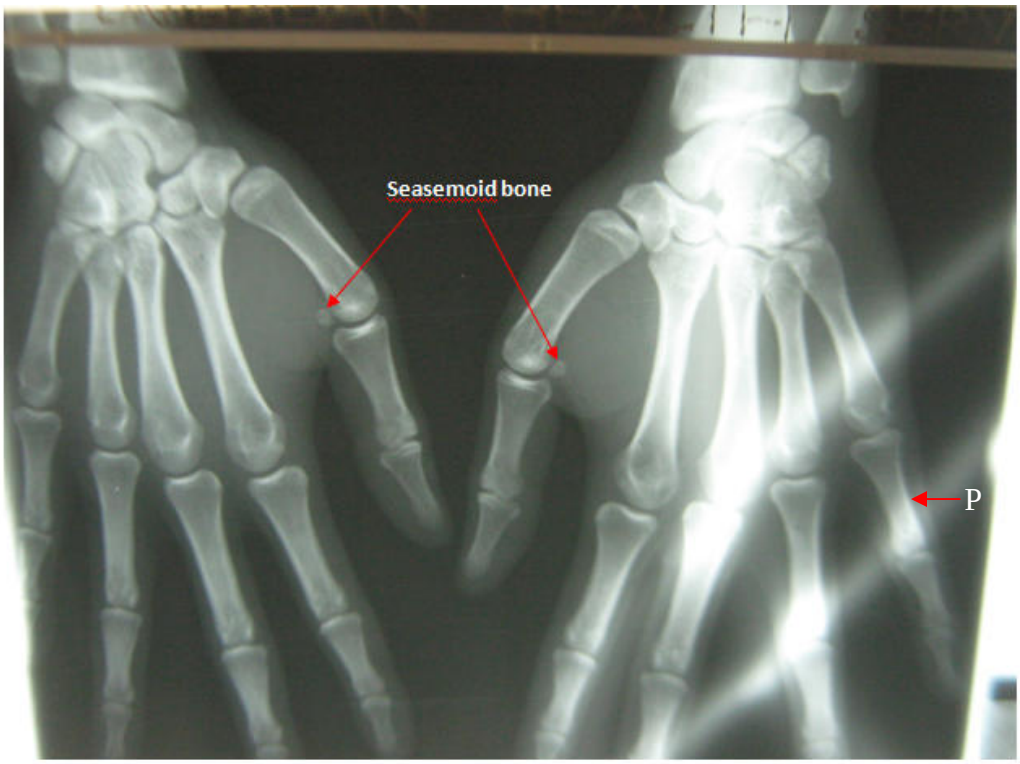

Fig 5: X-ray showing no bony involvement in the polydactyly (P) and presence of seasamoid bones

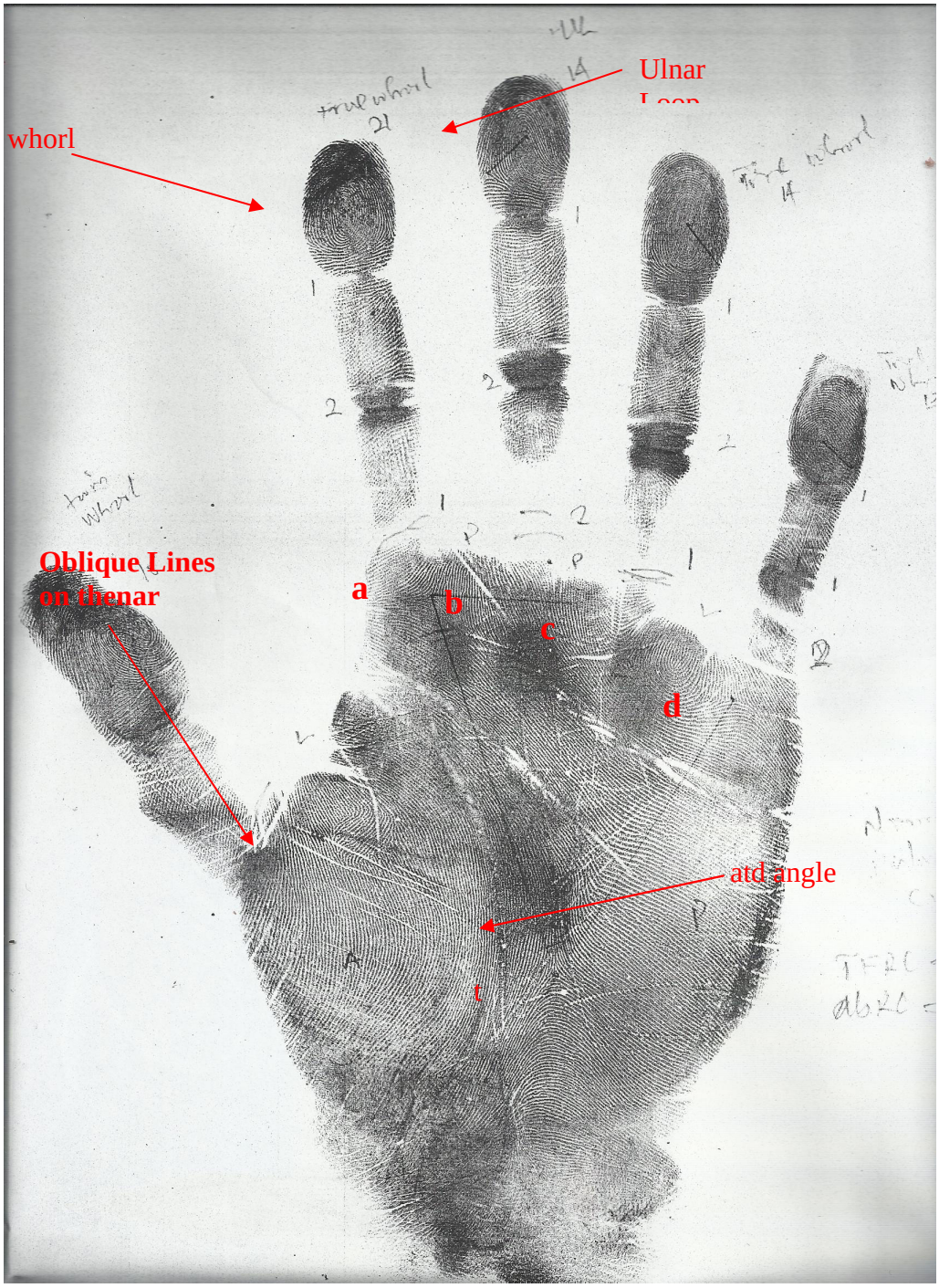

Fig 6: Showing the dermatographic patterns of the right hand

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