

# Prevalence and characteristics of congenital hand and foot anomalies requiring surgery in remote areas of Thailand

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**Background** : *To identify patients in remote regions of Thailand who suffer from congenital anomalies, including cleft lip, cleft palate, and various types of congenital anomalies, in November 1998, the Relief and Community Health Bureau, Thai Red Cross Society, under the HRH Princess Maha Chakri Sirindhorn initiated the “cosmetic surgery project for cleft lips and cleft palates and other deformities” to solve this problem. The surgery mission had been a successful charity project for the last 18 years. Most of the patients are cleft lip and cleft palate patients along with all anomalies of hand and foot.*

**Objective** : *The purpose of this study was to analyze the prevalence and characteristics of congenital hand and foot anomalies requiring surgery in remote areas of Thailand.*

**Methods** : *Data of surgical patients who presented with congenital hand and foot anomalies form 2007 - 2014 were retrospectively reviewed.*

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**Results** : Four hundred and three cases of congenital hand and foot anomalies were operated on: 202 males (50.10%) and 201 females (49.90%). The average age at surgery was 10.90 years (two months – 67.00 years). The most common anomaly was polydactyly (205 cases, 50.90%), followed by syndactyly (142 cases, 35.20%), constriction ring syndrome (19 cases, 4.70%), camptodactyly (11 cases, 2.70%) and macrodactyly (7 cases, 1.70%). Rare conditions (<1.00%) were radial club hand (2 cases), congenital trigger thumb (2 cases), clasped thumb (two cases), and clinodactyly (1 case).

**Conclusions** : Polydactyly (50.90%) and syndactyly (35.20%) were the two most common congenital hand and foot anomalies that required surgery in a remote area of Thailand. The prevalence of congenital hand and foot anomalies that required surgery was 1.15 per 1,000 live births.

**Keywords** : Congenital hand anomalies, congenital foot anomalies, prevalence, Thailand.

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**เหตุผลการทำวิจัย** : ผู้ป่วยที่มีภาวะความพิการแต่กำเนิดในพื้นที่ห่างไกลและทุรกันดารของประเทศไทย มักจะไม่สามารถรับบริการทางสาธารณสุขที่เหมาะสม ในปี พ.ศ. 2541 สำนักงานบรรเทาทุกข์และประชานามัยพิทักษ์ สภากาชาดไทย ได้ริเริ่มโครงการศัลยกรรมตกแต่งแก้ไขปากแหว่ง เพดานโหว่ และความพิการอื่น ๆ เพื่อให้บริการผ่าตัดแก้ไขความพิการแต่กำเนิด รวมถึงความพิการอื่น ๆ ของประชากรไทยในถิ่นทุรกันดารที่ไม่สามารถเข้าถึงบริการทางสาธารณสุขได้ตลอด 18 ปี ของโครงการ ภาวะความพิการแต่กำเนิดของผู้ป่วยที่ได้รับการผ่าตัดแก้ไขส่วนใหญ่นั้น ประกอบด้วย ภาวะปากแหว่ง เพดานโหว่ และความพิการแต่กำเนิดของมือและเท้า เป็นหลัก

**วัตถุประสงค์** : เพื่อศึกษาอัตราความชุกของโรค และลักษณะเฉพาะของความพิการแต่กำเนิดบริเวณมือและเท้าที่จำเป็นต้องรักษาด้วยการผ่าตัดของกลุ่มผู้ป่วยในถิ่นทุรกันดารของประเทศไทย จากโครงการศัลยกรรมตกแต่งแก้ไขปากแหว่ง เพดานโหว่ และความพิการอื่นของสำนักงานบรรเทาทุกข์และประชานามัยพิทักษ์ สภากาชาดไทย

**วิธีการศึกษา** : ศึกษาข้อมูลจากเวชระเบียนของผู้ป่วยในโครงการศัลยกรรมตกแต่งแก้ไขปากแหว่ง เพดานโหว่ และความพิการอื่น ๆ ที่มีภาวะความพิการแต่กำเนิดบริเวณมือและเท้าที่เข้ารับการผ่าตัดรักษาในช่วงปี พ.ศ. 2550 – 2557

**ผลการศึกษา** : ผู้ป่วยที่เข้ารับการผ่าตัดรักษาภาวะความพิการแต่กำเนิดบริเวณมือและเท้าทั้งสิ้น 403 คน เป็นชาย 202 คน หญิง 201 คน โดยอายุเฉลี่ยที่เข้ารับการผ่าตัดคือ 10.90 ปี ความพิการแต่กำเนิดบริเวณมือและเท้าที่พบได้บ่อยในถิ่นทุรกันดารของประเทศไทยเป็นดังนี้ ภาวะนิ้วมือและนิ้วเท้าเกินร้อยละ 50.90 (205 คน), ภาวะนิ้วมือและนิ้วเท้าติดกันร้อยละ 35.20 (142 คน), ภาวะพังผืดหดร้งบริเวณมือและเท้าร้อยละ 4.70 (19 คน ), ภาวะนิ้วมือและนิ้วเท้าโค้งงอแต่กำเนิด ร้อยละ 2.70 (11 คน) และภาวะนิ้วมือและนิ้วเท้าโตผิดปกติ ร้อยละ 1.70 (7 คน) นอกจากนั้นยังพบความพิการอื่น ๆ ที่พบได้น้อยกว่าร้อยละ 1.00 ได้แก่ ภาวะขาดการเจริญเติบโตของกระดูกเรเดียส, ภาวะนิ้วมือนิ้วมือแต่กำเนิด, ความผิดปกติของนิ้วหัวแม่มือจากการขาดเส้นเอ็นเหยียดนิ้ว และปลายนิ้วคดงอแต่กำเนิด

- สรุป** : ความชุกของความพิการแต่กำเนิดของมือและเท้าที่ต้องได้รับการผ่าตัดในถิ่นทุรกันดารของประเทศไทยอยู่ที่อัตรา 1.15 ต่อ 1,000 คนของอัตราการเกิดมีชีพ โดยพบว่าชนิดของความพิการแต่กำเนิดในบริเวณมือและเท้าที่บ่อยที่สุด 2 อันดับแรก ได้แก่ ภาวะนิ้วมือและนิ้วเท้าเกิน และภาวะนิ้วมือและนิ้วเท้าติดกันแต่กำเนิด
- คำสำคัญ** : ความพิการแต่กำเนิด, ความชุก, นิ้วมือและนิ้วเท้าเกิน, นิ้วมือและนิ้วเท้าติด, ประเทศไทย.

In Thailand, the prevalence of major congenital anomalies was 26.1 per 1,000 live births. The five most common birth defects were congenital heart defects, limb anomalies, cleft lip and/or cleft palate, Downs' syndrome and congenital hydrocephalus.<sup>(1)</sup> In Thai newborns, common orthopaedic problems were calcaneovalgus (60:1,000 live births), metatarsus adductus (7.6:1,000), polydactyly and/or syndactyly (2.6:1,000), talipes equinovarus (2.4:1,000), brachial plexus injury (1.5:1,000), developmental dysplasia of the hip (0.6:1,000), osteogenesis imperfecta (0.6:1,000), congenital vertical talus (0.3:1,000) and clavicular fracture (0.3:1,000).<sup>(2)</sup>

Children born with congenital anomalies rely on early evaluation and surgical interventions to limit morbidity. However, people in rural and remote regions of Thailand lack access to receive adequate primary health care service, causing delay and neglect for proper treatment. To address this problem, the Relief and Community Health Bureau, Thai Red Cross Society, under the patronage of HRH Princess Maha Chakri Sirindhorn initiated the "Cosmetic surgery project for cleft lip and cleft palate and other deformities" in November 1998. With project objectives to identify patients in remote regions of Thailand who suffer from congenital anomalies including cleft lip, cleft palate and other various types of congenital anomalies in need of surgical treatment. Subsequently, mobile surgical units were set up in hospitals in several provinces of Thailand to provide surgical operations for these patients. Since its inception 16 years ago, a total of 4,753 patients have received surgery. The objective of this present study is to analyze the prevalence of various types of

congenital hand and foot anomalies that required surgical intervention in remote areas of Thailand.

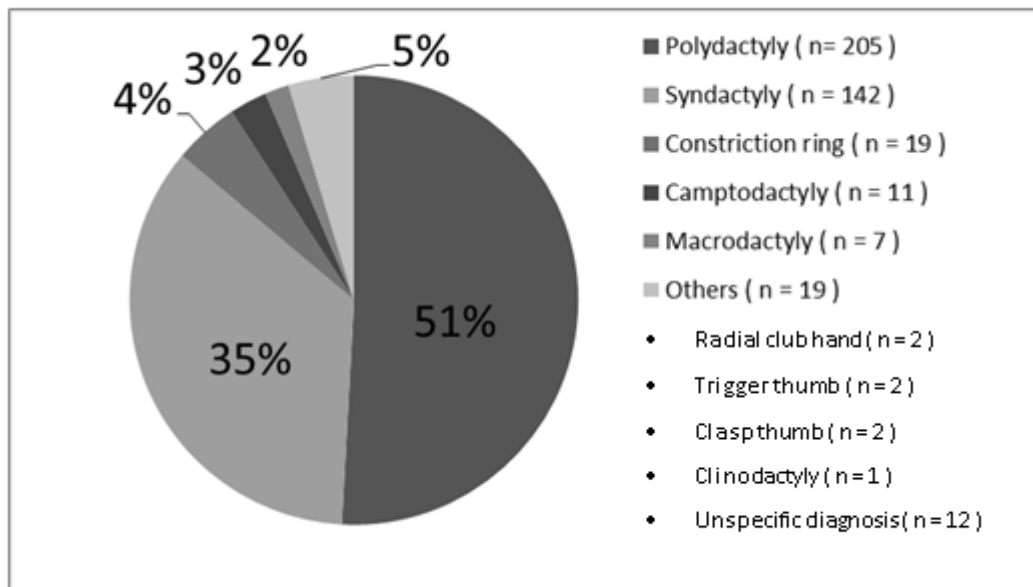
## Methods

This retrospective study focused on congenital hand and foot anomaly cases in remote areas of Thailand that required surgical intervention from the medical mission "Cosmetic surgery project for cleft lip and cleft palate and other deformities" between January 2007 and August 2014. During this 8-year study period, mobile surgical units were set up in 40 provinces. The number of populations and live births in each province of Thailand were collected. In this study, we analyzed the prevalence of various types of hand and foot anomalies, age at operation, gender, pattern of limb involvement in each anomaly and reviewed the types of surgical operations for each deformity.

## Results

Between 2007 and 2014, 403 patients, 202 males (50.10%) and 201 females (49.90%), with congenital hand and foot anomalies were operated on. The average age of surgery was 10.90 years (age range two months – 67.00 years).

The most common congenital hand and foot anomaly found in our study was polydactyly (205 cases, 50.90%), followed by syndactyly (142 cases, 35.20%), constriction ring syndrome (19 cases, 4.70%), camptodactyly (11 cases, 2.70%) and macrodactyly (7 cases, 1.70%). Rare conditions (less than 1.00%) included radial club hand (2 cases), congenital trigger thumb (2 cases), clasped thumb (two cases), and clinodactyly (1 case) (Figure 1).



**Figure 1.** Types of congenital hand and foot anomalies that required surgery in remote areas of Thailand between 2007 and 2014 (a total of 403 surgical patients).

The total population in 40 provinces of Thailand during 2007 - 2014 were 28, 952, 588 and total live births were 351,135.<sup>(3)</sup> The overall prevalence of congenital hand and foot anomalies that required surgical intervention from during 2007 - 2014 was 1.15 per 1,000 live births. As for the two most common anomalies, the prevalence of polydactyly that required surgery and prevalence of syndactyly was 0.58 per 1,000 and 0.40 per 1,000 live births, respectively (Table 1). The overall prevalence of hand and foot anomalies including polydactyly and syndactyly have shown a slight decrease over the 8-year period. (Figure 2)

Remote areas of Thailand were divided into four geographic regions, namely: northern, north eastern, central and southern Thailand. Patients with congenital anomalies were most commonly found in the northeast region (211 cases). There were 61 patients found in the northern part of Thailand, 66 patients in the central region, and 65 patients in the southern region. In total, the most common type of congenital anomaly in all regions was polydactyly,

followed by syndactyly. Only in the southern Thailand syndactyly was found more common than polydactyly (Table 2). Moreover, no patients with macroductyly were observed in the southern part of Thailand.

#### **Polydactyly patients**

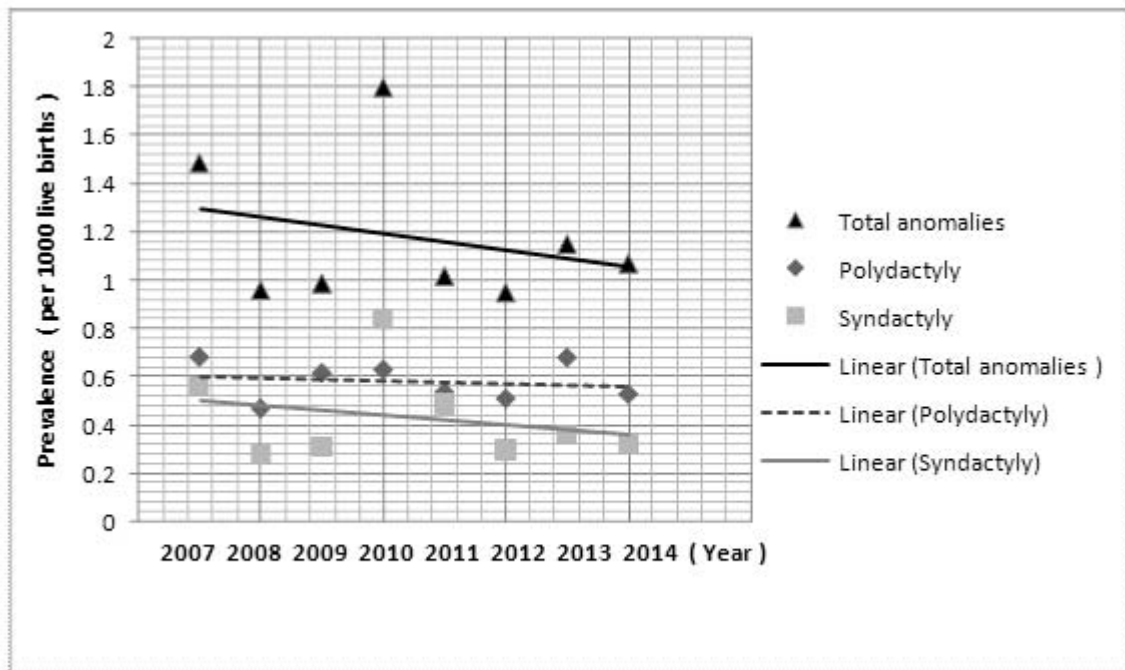
Polydactyly was the most common congenital hand and foot anomaly that required surgery in remote areas of Thailand: a total of 205 polydactyly patients (50.90%) from 403 patients. There were 103 males and 102 females who suffered from polydactyly. The prevalence of polydactyly was 0.58 per 1,000 live births. Mean age upon receiving the operation was 10.80 years. (age range 2 months – 56 years)

Compared with the other types of deformities, polydactyly prevalence and proportion was highest in the northern region of Thailand (0.76: 1,000 live births; 42/61 cases (68.90%), followed by the northeast region (0.66: 1,000 live births; 118/211 cases (55.90%), central part (0.65:1,000 live births; 28/66 cases (42.40%) and then the south (0.23: 1,000 live births;17/65 cases (26.20%)(Table 1, 2).

**Table 1.** The prevalence of congenital hand and foot anomalies requiring surgery in remote areas of Thailand during 2007 - 2014.

Region	Total Population (person)	Total Live births (person)	Number of cases ( n ) and Prevalence per 1,000 live births (p)																	
			All Anomalies			Polydactyly			Syndactyly			Constriction Ring			Camptodactyly			Macroductyly		
			n	p	n	p	n	p	n	p	n	p	n	p	n	p	n	p		
North	4,551,725	55,278	61	1.10	42	0.76	13	0.24	2	0.04	1	0.02	1	0.02	1	0.02				
North-East	14,688,405	177,932	211	1.19	118	0.66	71	0.40	4	0.02	5	0.03	4	0.02	4	0.02				
Central	3,522,659	42,827	66	1.54	28	0.65	19	0.44	7	0.16	4	0.09	2	0.05	2	0.05				
South	6,189,799	75,098	65	0.87	17	0.23	39	0.52	6	0.08	1	0.01	0	0.00	0	0.00				
Total	28,952,588	351,135	403	1.15	205	0.58	142	0.40	19	0.05	11	0.03	7	0.02	7	0.02				

Abbreviation : n = Number of case, p = Prevalence per 1,000 live-births



**Figure 2.** The prevalence of congenital hand and foot anomalies that required surgery in remote areas of Thailand during 2007 - 2014.

**Table 2.** Distribution of congenital hand and foot anomalies in 4 regions of Thailand.

(Total 403 patients)	North	Northeast	Central	South	Total
All deformities	61	211	66	65	403
Polydactyly	42	118	28	17	205
Syndactyly	13	71	19	39	142
Constriction ring	2	4	7	6	19
Camptodactyly	1	5	4	1	11
Macroductyly	1	4	2	0	7
Others	2*	9**	6	2	19

\* Trigger thumb (1 case), unclassified ( 1 case )

\*\* Trigger thumb (1 case), Radial club (2 cases) ,Clasp thumb ( 2 cases), Clinodactyly (1 case) , unclassified ( 3 cases )

Polydactyly cases usually affected the hand (157 cases from 205, 76.60%), while only 41 cases (20.00%) affected the feet. The ratio between the hand and foot involvement was 3.8:1. Only seven cases (3.10%) involved both hand and foot (Table 3).

In 157 hand polydactyly patients, 155 cases (98.70%) suffered pre-axial polydactyly. Post-axial polydactyly was rare (2 cases). Unilateral hand polydactyly was found in 148 cases (94.30%). Bilateral hand involvement was found in only nine



cases (5.70%). Foot polydactyly was less common (41/205 cases, 20.00%). Most were unilateral involvement (24 cases, 58.50%) but the prevalence of bilateral polydactyly in feet was much higher than in hands (17 cases, 41.50%). The proportion between unilateral and bilateral polydactyly in hands was significantly higher in comparison to unilateral and bilateral polydactyly proportion in feet (16.4:1 vs 1.4:1).

Most polydactyly patients (187 cases) were successfully treated by soft tissue procedures alone including some simple excision or excision with reattachment of intrinsic muscle and collateral ligament from extra-digit. Only 18 cases in our series called for bony procedures such as corrective osteotomy or insertion of Kirchner wire fixation to correct severe angular deformity of the remaining thumb.

### Syndactyly

In total, there were 142 syndactyly patients,

75 males and 64 females. The prevalence of syndactyly was 0.40 per 1,000 live births. The mean age at surgery was 10.10 years. (age range 6 months to 67.00 years)

Compared with other types of anomalies, the prevalence of syndactyly was highest in southern Thailand (0.52: 1,000 live births); followed by the central (0.44: 1,000 live births); northeast (0.40: 1,000 live births) central and northern parts of Thailand (0.24 : 1,000 live births) (Table 1).

Isolated syndactyly, without other associated anomalies, were found in 119 cases. In this group, hand syndactyly was more common than foot syndactyly (70 cases, 58.80% vs. 43 cases, 36.10%). The proportion between hand and foot involvement was 1.6:1. Combined hand and foot syndactyly was present in six cases (5.00%) (Table 3). Acrosyndactyly was uncommon (4 cases, 3.40%). There were three cases of hand acrosyndactyly and one case of foot acrosyndactyly.

**Table 3.** Characteristics of polydactyly and syndactyly patients that required surgery in remote areas of Thailand during 2007 - 2014 (Total 347 patients).

	<b>Polydactyly (n = 205)</b>	<b>Syndactyly (n = 142)</b>	
		<b>Isolated (n = 119)</b>	<b>Associated anomalies (n = 23)</b>
Hand involvement	157	70	
Unilateral	148	58	Syndactyly with Amniotic band
Bilateral	9	12	(n = 10)
Pre-axial	155	-	Syndactyly with Polydactyly
Post-axial	2	-	(n = 8)
Foot involvement	41	43	Symbrachydactyly
Unilateral	24	35	(n = 4)
Bilateral	17	8	Syndactyly with Torticollis
Hand & Foot involvement	7	6	(n = 1)

Most of the syndactyly patients were unilaterally involved. In 70 cases of hand syndactyly, unilateral and bilateral hand involvement was found in 58 and 12 cases, respectively. In 43 cases of foot syndactyly, 35 cases were with unilateral foot involvement and eight cases with bilateral feet involvement. No significant difference was found in the proportion between the unilateral and bilateral in hand and foot syndactyly (4.8:1 and 4.4:1, respectively).

Interestingly, we found that 16.50% (23 cases) of syndactyly cases were associated with other congenital deformities. Furthermore, the most commonly associated condition with syndactyly was constriction ring syndrome (10 cases), polydactyly (eight cases), symbrachydactyly (four cases) and syndactyly with torticollis (one case).

Our technique for syndactyly separation of the hand included commissure reconstruction and webspace coverage by dorsal flap design (Figure 3). We used a dorsal and volar zigzag incision along the finger. Defects of the finger were covered by full thickness skin grafts from the groin region. Fingertip and perineuchium areas were created by long seagull flap design. In less severe syndactyly

cases that involved only webspace, separation of the syndactyly was performed without skin graft by Ostrowski flap design. In the event where syndactyly cases presented with other associated problems, such as constriction band or polydactyly, an additional simultaneous operation was performed along with the syndactyly separation.

### **Constriction ring syndrome**

Constriction ring syndrome (CRS) was the third most common hand and foot anomaly in our study. Isolated constriction ring syndrome was found in 19 cases. Prevalence of isolated CRS was 0.05 per 1,000 live births. Geographical distribution of CRS among Thailand found 2 cases in the north, 4 cases northeast, 7 cases central and 6 cases in the south. The mean age at operation was 7.60 years (age range 8 months - 26.25 years). There were seven males and 12 females in this group. Single ring lesion was found in 10/19 cases and multiple rings involvement were present in 9/19 cases. Constriction ring syndrome surgery was carried out by releasing the constriction ring with z-plasty, skin graft and separation of syndactyly in eleven, one and five cases, respectively.



**Figure 3.** Flap design for syndactyly separation.

### **Camptodactyly**

Camptodactyly was found in 11 cases, in our series: six females and five males. The prevalence of camptodactyly was 0.03 per 1,000 live births. The mean age at operation was 11.05 years (age range 2.00 – 53.00 years). Unilateral camptodactyly was found in nine cases. Single digit involvement was found in five cases (one index and four small fingers). Unilateral multiple digits involvement was found in two cases (middle and ring finger involvement). Bilateral camptodactyly was found in two cases (each case involved both small fingers). Camptodactyly was found in the north (one), northeast (five), central (four) and southern (one) of Thailand.

There were several surgical operations for camptodactyly in this study including: 1) release with lateral finger flap (five cases); 2) release with primary closure (five cases); 3) release with full-thickness skin graft (one case); and 4) Kirchner wire fixation (one case).

### **Macroductyly**

Macroductyly was found in seven cases (1.70%); all seven cases were female. The prevalence of macroductyly was 0.02 per 1,000 live births. The mean age at surgery was 9.55 years (age range 4.00 - 17.00 years). Hand macroductyly was present in two cases (one case was right index and middle finger macroductyly, the other was bilateral). Toe macroductyly was found in five cases. By geographic distribution, macroductyly was found in the north (one), northeast (four) and central (two) parts of the country.

All cases were operated by debulging procedures, with or without osteotomy or Kirchner wire fixation.

### **Other rare condition (less than 1.00% of total cases)**

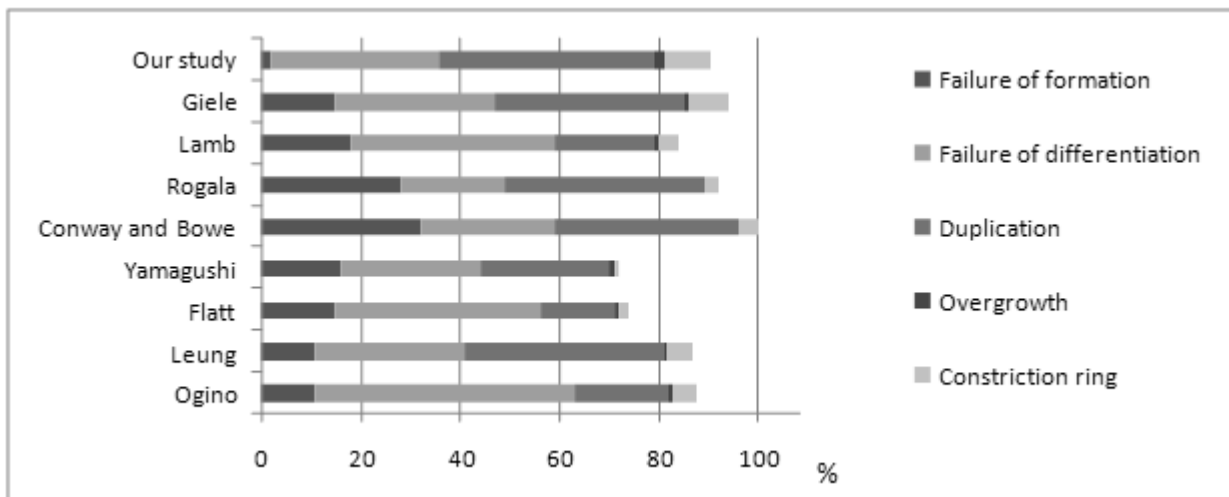
- Radial club hand (two cases), operated by centralization;
- Congenital trigger thumb (one case), operated by opened A1 pulley release;
- Clasp thumb (two cases), operated by first web release, extensor indicis proprius transfer, local flap coverage and full-thickness skin graft;
- Clinodactyly (one case), operated by extensor tendon rerouting at left ring finger;
- Unclassified categories (12 cases).

### **Discussion**

As for children born with congenital hand and foot anomalies, early surgical intervention limits morbidity and returns normal patterns of hand and foot functions. From our study, the prevalence of some congenital hand and foot anomalies that were treated by surgery was far less than prevalence found in newborns. For example, the prevalence of polydactyly and syndactyly treated by surgery in this study was 0.98:1,000 compared with prevalence of polydactyly and syndactyly found in newborns in a previous study (2.6:1,000 live births).<sup>(2)</sup> If possible, surgical corrections before the age of 1.00 - 1.50 years yields the most desirable outcome that allows children to start developing hand skills. Unfortunately, in Thailand, the treatment is usually delayed or neglected due to geographical constraints and lack of adequate healthcare providers. We found that the mean age at time of surgery in remote areas of Thailand was much higher than the recommended (10.90 years for overall anomalies, 10.80 years for polydactyly and 10.10 years for syndactyly). Causes of the delayed or neglected treatments of these conditions consist of

inadequate access to healthcare service, lack of knowledge about possibility of treatment and in some rural areas, refusal of surgical treatment due to specific cultural or religious beliefs. However, the prevalence of congenital hand and foot anomalies that required surgery in remote area of Thailand was slightly decreased from 1.48 : 1,000 live births in 2007 to 1.07:1,000 live births in 2014 after this medical mission started. (Figure 2 ). This effect might come from 1.) The overall prevalence of congenital anomalies may be decreased in Thai population, 2.) Some of the cases were operated in our mobile surgical units every years and 3.) Some of the cases were earlier detected by parents or health care provider and already received proper surgical treatments from local or referral hospitals.

In this study, we found that the most common hand and foot anomaly that required surgery in remote areas of Thailand was polydactyly (205 cases, 50.90%), followed by syndactyly (142 cases, 35.20%), constriction ring syndrome (19 cases, 4.70%), camptodactyly (11 cases, 2.70%) and macrodactyly (seven cases, 1.70%). According to International Federation of Societies for Surgery of the Hand (IFSSH) classifications, our patients were classified in the following categories: 1) duplication anomaly (polydactyly); 2) failure of differentiation (syndactyly, camptodactyly, clinodactyly, trigger thumb, and clasp thumb); 3) overgrowth anomaly (macrodactyly); and, 4) failure of formation (radial club hand and symbrachydactyly).<sup>(4-7)</sup> This classification allows us to compare types of congenital upper limb anomalies with other studies (Figure 4).<sup>(8)</sup>



**Figure 4.** Comparison of the type of congenital upper limb anomalies with previous studies (Classified by IFSSH classification\*)

\* According to IFSSH classification, "polydactyly" was classified as a duplication anomaly, "syndactyly, camptodactyly, clinodactyly, trigger thumb, and clasp thumb" as a failure of differentiation; "macrodactyly" as an overgrowth anomaly; and "radial club hand, symbrachydactyly.

As seen in previous studies, the two most common categories of congenital upper limb anomalies were duplication and failure of differentiation. In our study, the third most common category was constriction ring syndrome which was in contrast to many previous studies which found the failure of formation as the next most common class.

In terms of geographic distribution, we found that the prevalence of overall congenital hand and foot anomalies requiring surgery were highest in the central region of Thailand. Polydactyly prevalence was highest in the northern region and tended to be lower in the central and areas. This is in contrast with syndactyly where prevalence was higher in the southern part of Thailand and less frequent in the north (Table 1). We suspect that this distribution might be attributed to several factors including some genetic distributions, lack of access to adequate healthcare services and religious beliefs facilitating the refusal of surgery in the southern areas.

### Conclusion

Our study shows that the prevalence of congenital hand and foot anomalies that required surgical intervention in remote areas of Thailand in 2007 - 2014 was 1.15 per 1,000 live births. The most common type of anomaly was polydactyly (50.90%), followed by syndactyly (35.20%), constriction ring syndrome (4.70%), camptodactyly (2.70%) and macrodactyly (1.70%). Other rare congenital anomalies (less than 1.00%) found in this study were radial club hand, congenital trigger thumb, clasped thumb and clinodactyly. The data collected allows healthcare providers to better understand the situation and the problem of congenital anomalies treatment

in remote area such as delayed or undiagnosed, late surgical treatment and inability to access to the standard hospital and in order to offer most appropriate surgical intervention to reduce morbidity rates in the rural areas of Thailand.

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