

Evaluation of hand anomalies in children admitted to a tertiary health center in eastern Anatolia

Doğu Anadolu bölgesinde bir üçüncü basamak sağlık merkezine başvuran el anomalili çocukların değerlendirilmesi

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Abstract

Aim: Identification and treatment management of congenital or acquired hand anomalies are serious problems for orthopedic and plastic surgeons. With the start of medical education in hand surgery as a subspecialty, the number of hand surgeons has increased, and this problem has partially been removed. This study aimed to contribute to the epidemiologic data of our country by sharing admission times and types, treatment management, and demographic data of children admitted with hand anomalies to a tertiary health center in the light of the literature.

Methods: Pediatric patients who were admitted with hand anomalies to the hand surgery clinic between 01.12.2018 and 01.12.2019 were included in this cross-sectional study. They were classified by extended OMT (Oberg, Manske, and Tonkin) classification by using the data obtained from the hospital registry. Patients' ages, genders, diagnoses, admission times, educational statuses, treatment plans and decisions of parents on the treatment were evaluated by a single hand surgeon available in the region.

Results: Out of approximately 1500 patients admitted to the hand surgery clinic, 49 patients between the ages of 0 and 18 with pediatric hand anomalies were included in the study. Out of 49 patients, 7 had acquired and 42 had congenital hand anomalies. The most common congenital anomaly was trigger finger. Eleven (22.5%) of the patients were at school age, 12 (24.5%) at pre-school age, and 26 (53%) were infants, aged 2 years and below.

Conclusion: It is highly important for patients with hand anomalies to reach the appropriate physician at the right time so that their treatment may be planned accordingly, the present anomaly does not delay the growth of the child, the deformity does not progress further and that these individuals can be brought into the society earlier. Therefore, we believe that the number of physicians should be increased due to the following reasons: Hand surgery is a new branch and these patients should be referred to subspecialists in accordance with the demands of the patients' parents. This branch is vital for the region, and there are patients still waiting for their already planned surgery.

Keywords: Congenital anomaly, Pediatric patient, Hand surgery

Öz

Amaç: Konjenital veya edinsel el anomalilerinin tanımlanması ve tedavi yönetimi ortopedik ve plastik cerrahlar için ciddi problemlerdir. El cerrahisinde tıbbi eğitimin bir alt uzmanlık olarak başlamasıyla, el cerrahlarının sayısı artmış ve bu sorun kısmen giderilmiştir. Bu çalışma, el anomalisi ile üçüncü basamak bir sağlık merkezine başvuran çocukların kabul tiplerini, tedavi yönetimini, kabul sürelerini ve demografik verilerini literatür eşliğinde paylaşarak ülkemizin epidemiyolojik verilerine katkıda bulunmayı amaçlamıştır.

Yöntemler: 01.12.2018 - 01.12.2019 tarihleri arasında el cerrahisi kliniğine el anomalisi ile başvuran pediatrik hastalar bu kesitsel çalışmaya alındı. Hastaların dosya kayıt sistemlerinden elde edilen veriler kullanılarak uzatılmış OMT (Oberg, Manske, and Tonkin) sınıflandırmasına göre sınıflandırıldı. Hastaların yaşı, cinsiyeti, tanısı, kabul süresi, eğitim durumu, tedavi planlaması ve ebeveynlerin tedaviye ilişkin kararları bölgede mevcut tek el cerrahisi tarafından değerlendirildi.

Bulgular: El cerrahisi kliniğine başvuran yaklaşık 1500 hastadan 0 ile 18 yaşları arasında çocuk el anomalisi olan 49 hasta çalışmaya dahil edildi. 49 hastanın 7'sinde edinsel ve 42'sinde konjenital el anomalisi vardı. En sık görülen konjenital anomali tetik parmak idi. Hastaların 11'i (%22,5) okul çağında, 12'si (%24,5) okul öncesi yaşta ve 26'sı (%53) 2 yaş ve altı bebeklik döneminde idi.

Sonuç: El anomalisi olan hastaların uygun zamanda uygun doktora ulaşmaları, deformitenin uygun zamanda tedavi olması, mevcut anomalinin çocuğun gelişimini engellememesi, deformitenin daha da büyümemesi ve bu bireylerin topluma erkenden kazandırılması açısından çok önemlidir. El cerrahisinin bölgede yeni bir oluşum olması, bu hastaların yan dal olan el cerrahisine yönlendirilmesi, ebeveynlerin bu yönde taleplerinin olması, halen planlanan ameliyatlarını bekleyen hastaların olması nedeniyle bölgede el cerrahinin olması çok önemlidir ve bu nedenlerden dolayı hekim sayısının artırılması gerektiği kanaatindeyiz.

Anahtar kelimeler: Konjenital anomali, Pediatrik hasta, El cerrahisi

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Ethics Committee Approval: Ethics approval was obtained from the Non-Interventional Clinical Research Ethics Committee of Van Yüzüncü Yıl University (Date: 27 December 2019, no: 18-09).
Etik Kurul Onayı: Etik onay Van Yüzüncü Yıl Üniversitesi Girişimsel Olmayan Klinik Araştırmalar Etik Kurulu'ndan alınmıştır (Tarih: 27 Aralık 2019, no: 18-09).

Conflict of Interest: No conflict of interest was declared by the authors.

Çıkar Çatışması: Yazarlar çıkar çatışması bildirmemişlerdir.

Financial Disclosure: The authors declared that this study has received no financial support.

Finansal Destek: Yazarlar bu çalışma için finansal destek almadıklarını beyan etmişlerdir.

Published: 1/31/2020
Yayın Tarihi: 31.01.2020

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Introduction

Upper extremity has functionally had a crucial role in people's daily life, even in the development of civilization throughout history. Identification of functions and structure of the hand, which plays the main role in the functional structure of human, has always been an object of interest and many studies on identification of hand deformities (congenital or acquired), determination of their frequencies among different races and societies, and treatment modalities have been performed. This study aimed to identify the frequency of patients admitted to the hand surgery center of our hospital with hand anomalies (after the specialist hand surgeon was appointed) and share our knowledge in light of the literature. Upper and lower extremity anomalies vary widely and these anomalies, whether congenital or acquired, cause loss of function and many psychological problems, thereby impairing the quality of life [1]. A study [2] evaluating the distribution of patients admitted with upper extremity injuries to the hand surgery clinic in terms of whether they were emergent or elective was performed in the region, however, epidemiologic evaluation of pediatric patients with hand anomalies according to patient group was not performed. Many studies on hand anomalies and different incidences among different races and societies were reported in the literature [3-7].

This study aimed to identify the types of patients admitted with congenital [8] and acquired [9] anomalies within a year to the hand surgery clinic of a tertiary care university hospital serving approximately five cities. We believe that the data obtained from this study will be useful for future epidemiological studies and that knowledge of sociodemographic characteristics of these patients will be a guide in planning treatment services.

Materials and methods

This cross-sectional study was performed on patients admitted with congenital and acquired anomalies to our hand surgery clinic between December 2018-December 2019. Patients who were under the age of 18 and who did not complete the growth curve were included in the group. A total of 3 patients who were under the age of 18 with closed physes were excluded. In compliance with the distribution of the patients, patients admitted at the age of 1 month or below were evaluated as 1-month old patients as there would not be any delay at the time of treatment. The study was performed in accordance with the Declaration of Helsinki and approved by the local Ethics committee (no: 2019/18-09).

Data collection tools

Patients' ages, genders, diagnoses, etiologies, treatment statuses, the city where they came from, admission times, and whether there was any sequela due to delayed admission were recorded. Ages of all patients in the study were calculated in months. Diagnoses were classified according to the "Refinement of Oberg, Manske, and Tonkin (OMT) Classification (Extended Version)" (Table 1) [10,11]. Based on the treatment plan, patients were classified as the ones who were operated, the ones who refused treatment, the ones whose future surgery was planned and the ones with managed conservatively. Admission timing of the patients was divided into early, late, and normal

timing for each patient group. Clinical evaluation was performed by a single physician with physical examination and comparative x-ray monitoring of both upper extremities.

Table 1: Refinement of Oberg, Manske and Tonkin (OMT) Classification (Extended Version)

1. Malformations	
A. Failure of axis formation/differentiation—entire upper limb	
1. Proximal-distal axis	
i. Brachymelia with brachydactyly	
ii. Symbrachydactyly	
iii. Transverse deficiency	
Amelia	
Clavicle	
Long/short above elbow	
Wrist	
Proximal/distal carpal row	
Metacarpal	
Proximal/middle/distal phalanx	
iv. Intersegmental deficiency	
Phocomelia (total/proximal/distal)	
2. Radial-ulnar (anteroposterior) axis	
i. Radial longitudinal deficiency	
Thumb hypoplasia (with proximal limb involvement)	
ii. Ulnar longitudinal deficiency	
iii. Ulnar dimelia	
iv. Radioulnar synostosis	
v. Humeroradial synostosis	
Elbow ankyloses	
3. Dorsal-ventral axis	
i. Nail-patella, Fuhrmann, and Al-Awadi syndromes	
ii. Arthrogryposis	
iii. Absent/hypoplastic extensor/flexor muscles	
4. Unspecified axis	
i. Undescended shoulder (Sprenzel)	
ii. Abnormal shoulder muscles	
B. Failure of axis formation/differentiation—hand plate	
1. Proximal-distal axis	
i. Brachydactyly	
ii. Symbrachydactyly	
iii. Transverse deficiency	
Wrist	
Proximal/distal carpal row	
Metacarpal	
Proximal/middle/distal phalanx	
2. Radial-ulnar (anteroposterior) axis	
i. Radial (thumb) deficiency (no radius involvement, absent thumb, absent/hypoplastic thenar muscles)	
ii. Ulnar deficiency	
iii. Radial polydactyly	
iv. Triphalangeal thumb	
v. Ulnar polydactyly	
3. Dorsal-ventral axis	
i. Dorsal dimelia (palmar nail)	
ii. Hypoplastic/aplastic nail	
iii. Arthrogryposis	
4. Unspecified axis	
a. Soft tissue	
i. Syndactyly	
ii. Camptodactyly	
iii. Thumb in palm deformity	
iv. Deviated finger without skeletal deformity	
b. Skeletal deficiency	
i. Clinodactyly	
ii. Kirner deformity	
iii. Metacarpal and carpal synostosis	
c. Complex	
i. Cleft hand	
ii. Synpolydactyly—central	
iii. Apert hand	
2. Deformations	
A. Constriction ring sequence	
B. Trigger digits	
C. Not otherwise specified	
3. Dysplasias	
A. Hypertrophy	
1. Whole limb	
i. Hemihypertrophy	
ii. Aberrant flexor/extensor/intrinsic muscle	
2. Partial limb	
i. Macroductyly	
ii. Aberrant intrinsic muscles of hand	
B. Tumorous conditions	
1. Vascular	
i. Hemangioma	
ii. Malformation	
2. Neurological	
i. Neurofibromatosis	
3. Connective tissue	
i. Juvenile aponeurotic fibroma	
ii. Infantile digital fibroma	
4. Skeletal	
i. Osteochondromatosis	
ii. Enchondromatosis	
iii. Fibrous dysplasia	
iv. Epiphyseal abnormalities	
4. Syndromes	

Statistical analysis

The required statistical analyses were performed with SPSS 23 statistical software. Numerical variables were defined as mean (standard deviation) while categorical variables were presented in number and percentile. One-way analysis of variance (One-way ANOVA) was used to determine whether there was a difference between the ages in different classifications. Duncan multiple comparison test was used to determine the classifications that were important after analysis of variance. Fisher Exact test was used to determine whether there was a relationship between classifications and admission times, classifications and treatment status, treatment status according to the cities and between admission times according to the cities.

Results

Forty-nine patients were included in our study. Seven of the patients had acquired and 42 had congenital anomalies. According to the extended OMT classification, only one of the patients among the ones with acquired anomalies had camptodactylia and was suitable for Benson’s classification type II. Four (8.16%) patients had an anomaly related to the wrist-forearm and 45 (91.84%) patients had an anomaly on their hand. The youngest patient was newborn and the oldest was 15 years old. There were no patients over 15 years of age in the included age range (0-18). The difference between classifications in terms of mean month was statistically significant ($P<0.001$). The lowest and highest mean month values were obtained in 1.A.3.ii (Arthrogyposis) classification (mean: 1 month) and 3.A.2.i (Macroductyly) classification (180 months/15 years), respectively. Eleven (22.5%) of the patients were at school age, 12 (24.5%) at pre-school age, and 26 (53%) were infants aged 2 years and below. Thirty-five (71.4%) patients were referred by another specialist physician, among which 13 were referred from pediatrics, 14 from plastic surgery and 8 from orthopedics. Other patients did not have any referral history. The evaluations of age distributions according to the classification are presented in Table 2.

Table 2: Age (month) distribution of our patients according to OMT Classification (Extended Version)

Diagnosis	n	Mean	SD	95% Confidence Interval for Mean		Minimum	Maximum
				Lower Bound	Upper Bound		
1.B.4.a.i acquired	4	44.500	45.8	-28.367	117.367	10	108
2.B	6	42.167	24.6	16.406	67.927	1	72
1.B.2.iii	8	18.500	12.4	8.173	28.827	5	36
1.A.2.i	3	104	81.7	-98.910	306.910	12	168
1.A.2.ii	2	4.500	2.1	-14.559	23.559	3	6
1.A.3.ii	1	1	1	1	1	1	1
1.A.1.ii	3	39	26.7	-27.238	105.238	9	60
3.A.2.i	2	180	180	180	180	180	180
1.B.4.a.ii	2	102	25.5	-126.712	330.712	84	120
3.B.1.i	2	168	168	168	168	168	168
1.B.2.iv	2	84	84	84	84	84	84
3.B.4.i	2	96	96	96	96	96	96
2.A	2	60	60	60	60	60	60
1.B.4.c.i	2	2	2	2	2	2	2
1.B.2.v	2	20	20	20	20	20	20
1.A.2.iii	2	4	4	4	4	4	4
1.b.1.I	2	6	6	6	6	6	6
Total	49	50.3	54.6	34.609	65.962	1	180

A statistically significant relationship was found between the classification and admission times ($P<0.001$). Patients with 2.A (Constriction ring sequence) and 1.B.2.v (Ulnar polydactyly) classifications were generally at normal times, those with 1.A.2.i [Radial longitudinal deficiency, Thumb hypoplasia (with proximal limb involvement)], 1.A.2.iii (Ulnar

dimelia), 1.A.3.ii (Arthrogyposis), 2.B (Trigger digits), 1.B.4.a.i (Syndactyly), and 1.B.4.c.i (Cleft hand) were early and patients with acquired anomalies, namely, 3.B.4.i (Osteochonromatosis), 3.B.1.i (Hemangioma), 3.A.2.i (Macroductyly) and 1.B.2.iv (Triphalangeal thumb) were late in presentation. The evaluation of treatment status according to the classification is presented in Figure 1. The relationship between the classification and sequela occurrence was significant ($P<0.001$). While there was sequela in patients with acquired anomalies, 3.A.2.i (Macroductyly), 3.B.1.i (Hemangioma), 1.B.2.iv (Triphalangeal thumb), 3.B.4.i (Osteochonromatosis), and 1.B.4.c.i (Cleft hand), due to late admission, there was no sequela in other classifications. The relationship between classification and sequela is presented in Figure 2.

A significant relationship was found between the cities and admission times ($P<0.001$) (Figure 3). Patients coming from Bitlis and Igdır were gradually operated and those coming from Van and Hakkari were generally operated, or had their surgeries planned. The patient who were admitted with thumb hypoplasia and who came from Agri refused treatment, hence was not operated.

A statistically significant difference was found between the cities and admission times ($P<0.001$). Patients from Agri were admitted late, patients from Hakkari were mostly admitted early and patients from Van were mostly admitted late (Figure 4).

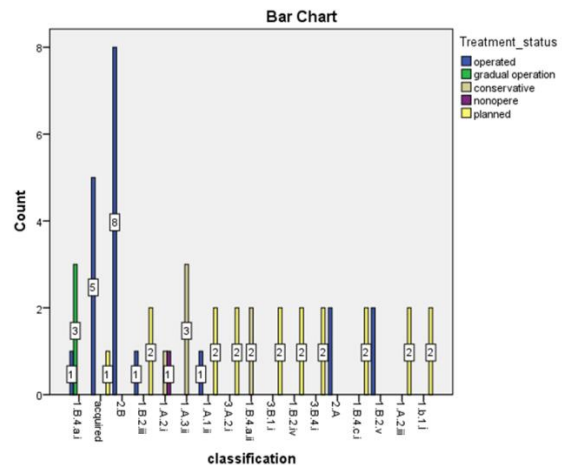


Figure 1: The evaluation of treatment status according to the classification

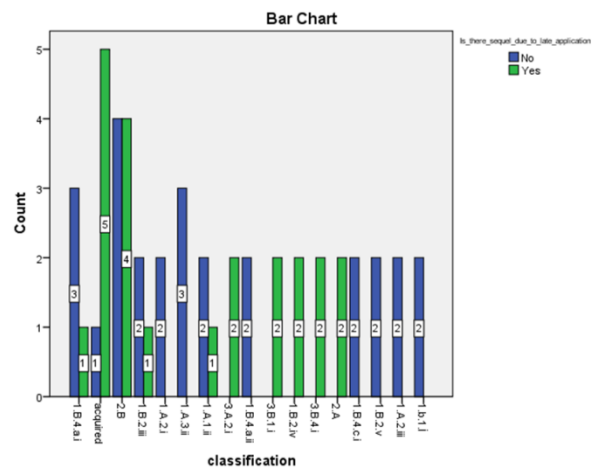


Figure 2: The relationship between the classification and occurrence of sequela

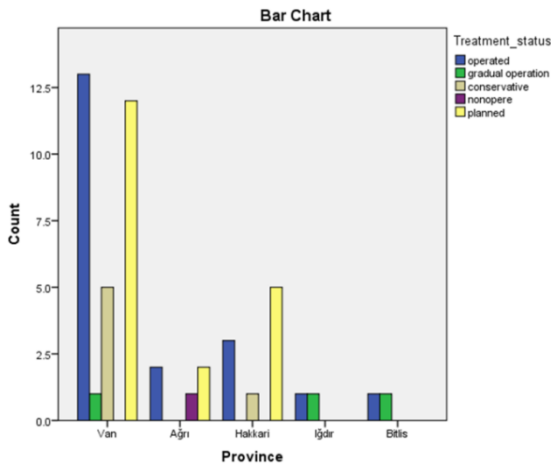


Figure 3: Relationship between the cities and admission times

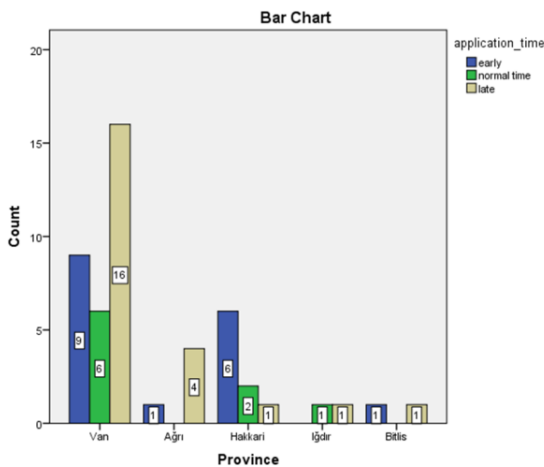


Figure 4: Difference between the cities and admission times

Discussion

The department of hand surgery provides the diagnosis and treatment of hand, wrist, forearm, shoulder, brachial plexus and peripheral nerve injuries and diseases and congenital anomalies of upper extremity [9,12]. The first hand surgery clinic at the level of Ministry of Health and University in our country was founded at Ankara Numune Training and Research Hospital in September, 2009. When a specialist hand surgeon was appointed to the Ministry of Health University in Van in September 2017, a hand surgery clinic was founded. Around 1500 patients are admitted to the Hand Surgery Clinic annually [2].

Hand Surgery is a field that has recently become a subspecialty in our country. Although studies identifying the clinical and sociodemographic characteristics of the patients admitted to hand surgery clinic have been performed in detail, there is not any detailed study on pediatric patients with hand anomalies, especially in the Eastern Anatolia Region of Turkey. Many age groups from birth to the end of childhood were admitted to our clinic with hand and wrist anomalies. Of these patients who had to be mostly operated at pre-school ages or in infancy, 22.5% were admitted at school ages. One hundred percent could not yet be reached in this patient group, who must be examined by a specialist physician and referred without missing the diagnosis. Admission times significantly differ according to patient groups, which is also related to the fact that the diagnosis can be recognized by both the patients' relatives and physicians. When the children with congenital hand

anomalies are admitted later than the average surgical times of their present diseases, the functional and cosmetic success rate of the surgery decreases [13]. In the region, 22 (44.9%) of these patients with congenital hand anomalies exceeded this time. Sequelae were seen in patients with Madelung deformity, burn, contracture, swan neck deformity secondary to mallet finger, hemangioma in the wrist, triphalangeal thumb, osteochondromatosis and cleft hand due to late presentation. As patients in Hakkari, Van, Bitlis and Iğdir preferred our clinic, most of them were either treated or their operations were planned. There was only one physician and some patients had to wait for their operations due to busyness. Patients from Hakkari were admitted early, while patients from Van and Agri were admitted late to our clinic in Van, which reveals that patients' relatives and/or physicians who would refer the patients are not aware enough. As the rate of being recognized in patients with additional pathology and syndrome was much, higher no delay was seen in their presentation. Presence of health insurance and parents being educated above high school have a positive effect on admission times. Congenital bilateral trigger finger was the most common anomaly. While especially grade 4 (Green Classification) presentations were more frequent, grade 2 and 3 were usually seen in the other finger. The possibility that this anomaly may be bilateral should be kept in mind in order to administer a complete treatment.

Limitations

The study was performed in a single center (as our clinic was the only hand surgery clinic in the region, no significant loss in the potential of patients was considered). Main limitations of our study may be that the number of patients was low, the 1-year duration of the study was short, and patients did not come from all Eastern Anatolia cities (due to probable conditions of transportation and closeness of other hospitals to other centers). Further studies are needed to contribute to the epidemiological data of our country in the future.

Conclusion

The facts that hand surgery is a new branch, patients in need of hand surgery should be referred from general examination to subspecialists, parents have demands on this direction, this branch with high work load and long learning-curve is extremely necessary for the region and that there are patients still waiting for their already planned surgery, suggest the need to increase the number of physicians. It is highly important for patients with hand anomalies to reach the appropriate physician at the right time so that their treatment can be planned to prevent the present anomaly from delaying the growth of the child without further progression. The aim is to bring these individuals into the society earlier. Therefore, we believe that identifying the frequency of these anomalies, increasing the awareness of the parents by improving the geographical as well as socioeconomic conditions, especially in rural areas, increasing the number admissions to the hospitals, evaluating the neonates in this regard and referring them to the related center early will contribute to the prevention of some progressive deformities and to the socioeconomic development of our country.

References

1. Doğan E, Gül S, Çullu N, Doğan MM. Case of incomplete fibular hemimelia with tarsal coalition, pes planus, ball and socket ankle. *J Surg Med.* 2019;3(3):271-3.
2. Arık H, Çoşkun T. Van İlindeki Bir Üniversite Hastanesinin El Cerrahisi Kliniğine Başvuran Hastaların Profili. *Van Tıp Derg.* 2018;25(4):502-7.
3. Ekblom AG, Laurell T, Arner M. Epidemiology of congenital upper limb anomalies in 562 children born in 1997 to 2007: a total population study from stockholm, sweden. *J Hand Surg Am.* 2010 Nov;35(11):1742-54.
4. Goldfarb CA, Wall LB, Bohn DC, Moen P, Van Heest AE. Epidemiology of congenital upper limb anomalies in a midwest United States population: an assessment using the Oberg, Manske, and Tonkin classification. *J Hand Surg Am.* 2015;40(1):127-32.
5. Koskimies E, Lindfors N, Gissler M, Peltonen J, Nietosvaara Y. Congenital upper limb deficiencies and associated malformations in Finland: a population-based study. *J Hand Surg Am.* 2011;36(6):1058-65.
6. Pinto HB, Pais AP, Vitorio SC, Brandão R, Moreira AAD, Molinaro LR. Case study of congenital anomalies of the upper limb in reference ambulatory care facility. *Acta Ortop Bras.* 2018;26(5):325-7.
7. Parker SE, Mai CT, Canfield MA, Rickard R, Wang Y, Meyer RE, et al. National Birth Defects Prevention Network. Updated National Birth Prevalence estimates for selected birth defects in the United States, 2004-2006. *Birth Defects Res A Clin Mol Teratol.* 2010;88(12):1008-16.
8. Oberg KC, Feenstra JM, Manske PR, Tonkin MA. Developmental biology and classification of congenital anomalies of the hand and upper extremity. *J Hand Surg Am.* 2010;35(12):2066-76.
9. Chu A, Chan J, Baxi O. Congenital Deformities of the Hands. *Pediatr Clin North Am.* 2020;67(1):85-99.
10. Iba K, Horii E, Ogino T, Kazuki K, Kashiwa K. Congenital Hand Committee of Japanese Society for Surgery of the Hand. The Classification of Swanson for Congenital Anomalies of Upper Limb Modified by the Japanese Society for Surgery of the Hand (JSSH). *Hand Surg.* 2015;20(2):237-50.
11. Tonkin MA, Tolerton SK, Quick TJ, Harvey I, Lawson RD, Smith NC, et al. Classification of congenital anomalies of the hand and upper limb: development and assessment of a new system. *J Hand Surg Am.* 2013;38(9):1845-53.
12. Leblebicioğlu G. Hand surgery in Turkey. *The Journal of Hand Surgery.* 2017;42(3):323.
13. de Almeida CEF. Analysis of surgical results and of residual postoperative deformities in preaxial polydactyly of the hand. *J Plast Reconstr Aesthet Surg.* 2017;70(10):1420-32.

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The National Library of Medicine (NLM) citation style guide has been used in this paper.

Suggested citation: Patrias K. Citing medicine: the NLM style guide for authors, editors, and publishers [Internet]. 2nd ed. Wendling DL, technical editor. Bethesda (MD): National Library of Medicine (US); 2007-[updated 2015 Oct 2; cited Year Month Day]. Available from: <http://www.nlm.nih.gov/citingmedicine>