

THE EVOLUTION AND IMPACT OF HEMANGIOMAS OF INFANCY

Petchesi Codruța Diana*, Iuhas Oana Alexandra**, Vesa Cosmin*, Jurca Alexandru*, Kozma Kinga*, Bembea Marius*

*University of Oradea, Faculty of Medicine and Pharmacy, Oradea, Romania,

** Regional Center for Medical Genetics, 12 C. Coposu St, Oradea, Romania, 410469, petchesi_diana@yahoo.com;

Abstract

Hemangiomas are the most frequent tumors in childhood and their incidence has increased lately. This study is a descriptive research, which follows the clinical form of hemangiomas, their evolution and the psychosocial impact of hemangiomas. A total of 326 patients were followed-up for a period of at least 14 years. The majority of hemangiomas were present at birth or appear within the first month; there are predisposing factors like feminine gender, prematurity, low birth weight, twin pregnancy. Most of the hemangiomas regress spontaneously, sometimes leaving residual scars or pigmentary changes. Rarely they present complications or associated malformations, which may require different medical approach. Hemangiomas affect children's self-esteem and relationships less and less every day, due to nowadays possibility of early treatment. Most of the patients had a spontaneous regression, by the age of 10 only about a third of them were still showing local signs and only 88 patients required treatment.

Key words: hemangiomas, prematurity, spontaneous regression, self-esteem, psychosocial impact.

INTRODUCTION

Hemangiomas are the most frequent tumors in childhood and their incidence has increased in the last years (Anderson KR et al, 2016). Although they are benign and self-limited tumors, some of them can cause complications or life-altering disfigurement, can affect children's self-esteem, relationships and performance in school and activities (Vivar K.L. Kruse L, 2018). Most of the hemangiomas regress spontaneously; in some cases, due to the hemangioma form (Bessis D. et al., 2015), location or size, medical or surgical treatment is required (Csoma ZR et al., 2017).

OBJECTIVE

This review follows the clinical evolution and the psychosocial impact of hemangiomas in children.

MATERIAL AND METHOD

The study is a descriptive research, which follows the clinical form, the natural and under treatment evolution, genetic particularities and the familial and psychosocial impact of hemangiomas.

A total of 326 patients with hemangiomas were followed-up from their first presentation up to the present, for a period of at least 14 years. Some cases were followed up retrospective, from 1983 to 2016, and others were studied both retrospective and prospective, from 2016 to 2018.

Since 1984 in the Regional Center for Medical Genetics (previously Genetic Compartment of the Clinical Municipal Hospital of Oradea) 555 patients were diagnosed with hemangiomas. In our study we only included patients which have already reached age 14. For 59 patients we had no follow-up data, so the final study group included 326 patients (59.1% of the total number of hemangiomas diagnosed in the Regional Center).

RESULTS AND DISCUSSION

Hemangiomas are the most common benign tumors (vascular tumors) in children, being found in 5 to 10 % of all infants. From the total number of patients diagnosed in our center (4966), hemangiomas represents 11.17%.

Most hemangiomas occur sporadically without a hereditary component. In a few families, hemangiomas segregate as a highly penetrant, autosomal dominant trait. Gene linkage studies of familial infantile hemangiomas show evidence of linkage to chromosome 5q31-33. (Thuy L. Phung et al., 2005). In our study group only 6 patients (1.84%) had a familial history of hemangioma, which may suggest a polygenic multifactorial inheritance (Jurca C. et al., 2006).

We followed a number of 326 patients, 193 girls and 133 boys; girls being preponderantly affected (sex ratio 1.45: 1) corresponding to the published literature data (Chiller KG et al., 2002). A small percent (11 patients – 3.37%) resulted from a twin pregnancy and in one case both twin were affected (0.3%), which

It is known that prematurity and low birth weight are predisposing factors (Hemangioma Investigator Group et al., 2007). In our case, 57 patients (17.48%) were premature neonates and the other 269 patients (82.51%) had normal gestational age. Birth weight was normal in 261 cases (80.1%), low in 51 cases (15.6%) and big in 14 cases (4.3%).

Usually hemangiomas are present at birth or appear in the first three months (Wananukul S, 2002). In our patients hemangiomas were present at birth in 194 patients (59.5%), within the first month in 85 patients (26.1%) and between the age of 1 -3 month in 47 patients (14.4%).

The number of hemangiomas may vary from a single one in 234 patients (71.8%) to multifocal hemangiomas, in 92 patients (28.2%). More than a third (33.43%) were involving the head and neck, generating

esthetical and sometimes functional problems (Macarthur CJ, 2006). Other locations were: the trunk, abdominal region, upper and lower limbs, genitourinary. More than 23 % of hemangiomas were multifocal, usually involving at least two regions. Fig 1

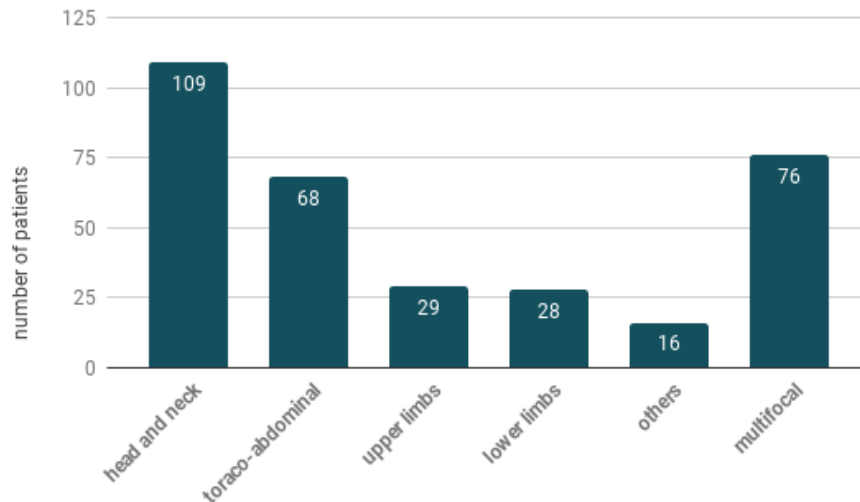


Fig.1. Location of hemangiomas

Morphologically, we can differentiate tuberous forms in 167 cases, cavernous in 32 cases, flat in 69 cases, mixed or combined forms in 45 cases. The maximum dimensions of the hemangiomas may vary from a few millimeters to gigantic, segmental hemangiomas; most of them are small (less than 1 cm) and medium (between 1 and 3 cm). Fig.2.

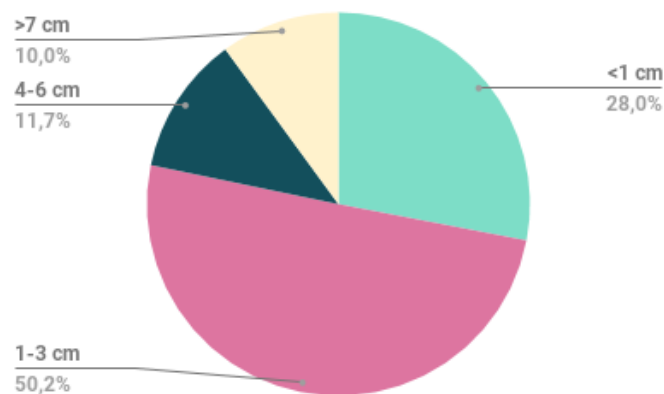


Fig.2. Dimensions of hemangiomas

Evolution of hemangiomas is in most of the cases benign (Marler JJ et al., 2005), appearing within the first weeks of life and rapidly growing

during the first months of life. By twelve months, involution is starting (Suh KY, Frieden IJ, 2010), sometimes slowly over many years (Chang LC et al., 2008). Infantile hemangiomas spontaneously regress over time, in some cases may leave residual scars or pigmentary changes (Couto R.A et al., 2012). In our study group only 88 patients required treatment, due to complication or because of the risk of long-term or permanent disfigurement. The treatment was either medical (general, intralesional and topical) or surgical (Pallag A et al., 2018). The rest of the patients had a spontaneous regression (73%), by the age of 10 only about a third of them were still showing local signs. A small part of the patients presented ulcerations (8.5%), necrosis (0.6%), infections (0.6%) or altered visual, respiratory or digestive function (4.3%).

In 43 cases the patients associated other malformations: cryptorchidism, cafe au lait spots, congenital dysplasia of the hip, cardiac problems, umbilical hernia, pigmented nevi, torticollis, intellectual disability, cleft lip and palate, hydrocephaly, radial hypoplasia, syndactily, upper extremity amputation, Sprengel anomaly, Down syndrome, arthrogryposis, hydrocele, flat foot, deafness and muteness, heterochromia, inguinal hernia, phimosis, hypospadias, hypogonadism, pectus excavatus, clinodactyly (Jurcă MC et al, 2018).

In 6 of the cases (1.84%), the patients were disabled, not thru hemangiomas but thru the associated malformations. They were not properly educated, nor socially integrated.

Sometimes hemangiomas can affect children's self-esteem, relationships and performance in school and activities, due to life-altering disfigurement. In the past, treatment possibilities were limited, so children with severe forms, especially those of the head and neck, were disfigured (Planas-Ciudad S et al., 2017). They felt socially isolated, they were playing alone, they did not feel like they were preferred by the parents (Dieterich-Miller C.A et al., 1992). In the present, guidelines (Reimer A et al., 2016) for hemangiomas management recommend the precocious referral of the patients to the specialist. (Strumila A. Et al., 2018), where he is evaluated and the treatment alternatives are decided, according to a score (Moyakine AV et. al., 2017). In our study group, due to precocious referral to the specialist and precocious treatment, patients were not disfigured, and by the age of 10, only a third had residual scars or pigmentary changes.

CONCLUSIONS

1. The majority of hemangiomas were present at birth or appeared within the first month (85,6%).

2. There are predisposing factors like feminine gender, prematurity, low birth weight, twin pregnancy which increases the incidence of hemangiomas.

3. Although there has been reported evidence of linkage to chromosome 5q31-33, most of our cases are sporadic, without significant family history.

4. The natural evolution of hemangiomas is self-limited with spontaneous regression: in most of the cases it only requires observation (73%).

5. A small part of the patients presents complications and requires medical or surgical treatment.

6. Rarely, they do associate other signs: malformations, syndromes (13.19%), which may require different medical approach.

7. Patients with hemangiomas should be referred to a specialist at the first sign.

8. By the age of 10, only a third of our patients had residual scars or pigmentary changes.

9. Hemangiomas affect children's self-esteem, relationships and performance in school and activities, less and less everyday, due to nowadays possibility of early treatment.

REFERENCES

1. Anderson KR, JJ Schoch, CM Lohse, JL Hand, DM Davis, MM. Tollefson, 2016, Increasing incidence of infantile hemangiomas (IH) over the past 35 years: Correlation with decreasing gestational age at birth and birth weight, *J Am Acad Dermatol*, 74(1):120-6.
2. Bessis D, M Bigorre, C. Labrèze, 2015, Reticular infantile hemangiomas with minimal or arrested growth associated with lipoatrophy, *J Am Acad Dermatol*, 72(5):828-33.
3. Chang LC, AN Haggstrom, BA Drolet, E Baselga, SL Chamlin, MC Garzon, KA Horii, AW Lucky, AJ Mancini, DW Metry, AJ Nopper, IJ Frieden, 2008, Hemangioma Investigator Group, Growth characteristics of infantile hemangiomas: implications for management, *Pediatrics*, 122(2):360-7.
4. Chiller KG, D. Passaro, IJ Frieden, 2002, Hemangiomas of infancy: clinical characteristics, morphologic subtypes, and their relationship to race, ethnicity, and sex, *Arch Dermatol.*, 138(12):1567-76.
5. Couto R.A., R.A. Maclellan, D. Zurakowski, A.K. Greene, 2012, Infantile hemangioma: Clinical assessment of the involuting phase and implications for management, *Plast Reconstr Surg.*, 130:619–624.
6. Csoma ZR, S. Dalmády, R. Ábrahám, T. Rózsa, K. Rácz, L. Kemény, 2017, Infantile haemangioma: clinical and demographic characteristics, experiences in the treatment, *Orv Hetil.*, 158(39):1535-1544.
7. Dieterich-Miller C.A., B.A. Cohen, J. Liggett, 1992, Behavioral adjustment and self-concept of young children with hemangiomas, *Pediatr Dermatol.*, 9:241–245.

8. Hemangioma Investigator Group, AN Haggstrom, BA Drolet, E. Baselga, SL Chamlin, MC Garzon, KA Horii, AW Lucky, AJ Mancini, DW Metry, B. Newell, AJ Nopper, IJ Frieden, 2007, Prospective study of infantile hemangiomas: demographic, prenatal, and perinatal characteristics, *J Pediatr. Mar*, 150(3):291-4.
9. Jurca MC., M. Bembea, I. Romocea, S. Bembea, 2006, Hemangioamele la copil-evolutie si tratament, *Revista Medicala Oradeana*, 13 (1): 61-70
10. Jurcă MC, M. Bembea, K. Kozma, M.I. Șandor, R.A. Negrean, L. Dobjanschi, E.A. Cuc, C.D. Petchesi, A.D. Jurcă, 2018, Empty sella associated with growth hormone deficiency and polydactyly, *Rom J Morphol Embryol*, 59(1):381–384
11. Macarthur CJ, 2006, Head and neck hemangiomas of infancy, *Curr Opin Otolaryngol Head Neck Surg.*, 14(6):397-405.
12. Marler JJ, JB Mulliken, 2005, Current management of hemangiomas and vascular malformations, *Clin Plast Surg*, 32(1):99-116
13. Moyakine AV, B. Herwegen, CJM van der Vleuten, 2017, Use of the Hemangioma Severity Scale to facilitate treatment decisions for infantile hemangiomas, *J Am Acad Dermatol* 77(5):868-873.
14. Pallag A, T Jurca, V Sirbu, A Honiges, C Jurca, 2018, Analysis of the Amount of Polyphenols, Flavonoids and Assessment of the Antioxidant Capacity of Frozen Fruits, *Rev. Chim.*, 69,(2), pg 445-448
15. Planas-Ciudad S, E. Roé Crespo, I. Sánchez-Carpintero, IJ Frieden, A. Martín-Santiago, P. Redondo Bellón, E. Moreno Artero, E. Osier, L. Puig Sanz, E. Baselga Torres, 2017, Infantile hemangiomas with minimal or arrested growth associated with soft tissue hypertrophy: a case series of 10 patients, *J Eur Acad Dermatol Venereol.*, 31(11):1924-1929.
16. Reimer A, M Fliesser, PH Hoeger, 2016, Anatomical patterns of infantile hemangioma (IH) of the extremities (IHE), *J Am Acad Dermatol.*, 75(3):556-563.
17. Strumila A, RV Dagilytė, V Beiša, 2018, Recommendations and a guideline for referral of infantile haemangioma to tertiary centres, *Acta Med Litu.*, 25(1):38-44.
18. Suh KY, IJ Frieden, 2010, Infantile hemangiomas with minimal or arrested growth: a retrospective case series, *Arch Dermatol.*, 146(9):971-6.
19. Thuy L. Phung, M Hochman, MC Mihm, 2005, Current Knowledge of the Pathogenesis of Infantile Hemangiomas, *Arch Facial Plast Surg*, 7(5):319-321.
20. Vivar K.L., L. Kruse, 2018, The impact of pediatric skin disease on self-esteem, *Int J Womens Dermatol*, 4(1): 27–31.
21. Wananukul S., 2002, Clinical manifestation and management of hemangiomas of infancy, *J Med Assoc Thai*, 85(1):280-5.