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RAPIDLY INVOLUTING CONGENITAL HEMANGIOMA (RICH) AT THE ORO-FACIAL REGION - A CASE REPORT

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ABSTRACT

Hemangiomas are the most common vascular tumor. Congenital hemangiomas are rare, benign vascular tumors that, unlike infantile hemangiomas, are present and fully grown at birth. They present as bossed plaques or exophytic masses located on the head, neck, or limbs. This report describes the case of a 9 months old female baby with progressively increasing hemangioma of the right cheek not involving the corner of the lip. This hemangioma promptly responded to Cryosurgery -“The Cold-Method”. A gradual improvement in clinical picture was observed in just a few days after the beginning of the treatment. The regression of 1/4 of the lesion was noticed as compared to the original size in 45 days of treatment, and to less than 1/10 after 8 months, free of any major side effects. The case was followed for more than 3 years. The procedure was quick, simple, bloodless.

KEYWORDS

Congenital hemangioma, Cryosurgery, Cold method and Involuting.

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INTRODUCTION

Background

Congenital hemangiomas are clinically present as fully developed lesions at birth, and either rapidly involutes during the first year of life or may never show involution¹⁻⁶. Congenital hemangiomas are rare, benign vascular tumors that, unlike infantile hemangiomas, are present and fully grown at birth. They present as bossed plaques or exophytic masses located on the head, neck, or limbs. According to their occurrence of natural history, two major subtypes of CH have been recognized: rapidly involuting congenital hemangiomas (RICH) and non-involuting congenital hemangiomas (NICH). In most cases, RICH involute completely by the age of

14 months, whereas NICH never regress but grow in proportion with the child and may require eventual excision. A third intermediate subtype, the so-called partially involuting congenital hemangioma (PICH), shows overlapping features of RICH and NICH³.

Juvenile hemangioma is an outdated term. Infantile hemangiomas are not present at birth and arises during the first 8 weeks of life^{1,2,4-7}. Initially the lesion appears as a circumscribed area of discoloration or telangiectasia of facial skin.

Hemangiomas are one of the most common neoplasm of infancy with an estimated prevalence of 1-3% of all the neonates and 10% by one year of age. 60% of hemangiomas arise in the head and neck. Prematurity is well defined risk factor especially in the neonates below 1500grams in weight. Hemangiomas show female predilection in the ratio of 3:1. A new study reveals that oxygen depletion in placenta can cause infantile hemangioma. Hemangiomas manifest by one month with a rapid growth during infancy and involution during childhood. They can occur anywhere in the head and neck but more common in the parotid, lip, oral cavity, perinasal region and the larynx².

Treatment approach of various types of oral haemangiomas varies considerably after consideration of the age of the patients, and the extent and type of the lesion. The range of treatment includes surgery⁸, laser therapy, use of sclerosing agents, and embolisation. The treatment approaches in the past include - Cryotherapy, compression, and radiation therapy¹.

The haemangiomas which are occurring in infancy should be left alone, for allowing the possibility for natural involution to occur². Immediate treatment considerations should be undertaken in case of psychological reasons and when the lesions at eyelids affecting vision. Angiography, computerised tomography and magnetic resonance imaging are useful in delineating the vascular supply and extent of the haemangiomas when their sizes are large or when jaw bones are involved². Management of such extensive lesions often requires a team approach consisting of radiologist, maxillofacial surgeon and plastic surgeon.

Case presentation

A nine month old female baby was reported to our department. A well circumscribed, round, bluish-red lesion on the right cheek not involving the corner of the lip was noticed. History given by the parents revealed presence of reddish peanut sized lesion of about 2.5cm in diameter, round raised soft non-tender, bluish in color since birth with gradual increase in size to the present size of about 3cm diameter. Figure No.1.

Investigations

The diagnosis of a hemangioma is best made by clinical history and physical exam. In cases of unclear diagnosis, the best radiographic modalities to use are either a Doppler ultrasound or MRI. On palpation there was no pulsation or phleboliths, easily compressible and blanch under finger. Normal delivery. At birth it was noticed as a small blue patch and started increasing in size as the infant grew. CT angio, radiation exposure on infant was a concern. Complete blood picture and basic lab tests for the procedure.

Treatment

Cryosurgery- The "Cold Method"

Under general anaesthesia, nitrous oxide cryosurgery was executed. Cryonization is based on tissue destruction by necrotizing temperatures (-20°C to -196°C) causing irreversible necrosis followed by elimination of the necrotic tissue. The cryo-cycle must usually be repeated. The depth of necrotization is up to 10mm. Each thaw cycle is 30-40 seconds at -186°C. With the help of cryogun, thawing of the lesion was carried out. Lubricating medium used was KY water based jelly. Starting from one corner, every area of the lesion was covered.

Intra-orally also the lesion was thawed. The mechanism of cell death by cryo is through the formation of intracellular and extracellular ice crystals immediately after cryotherapy. The extracellular ice reduces extracellular water, increasing solute concentration and osmolality, which causes a fluid shift and disrupts the cell membranes. Further damage is produced during the thawing process when intracellular ice damages mitochondria and the endoplasmic reticulum decreases the cell survival. Large ice crystals are

more damaging than small ones. Also slow thawing is associated with the recrystallization of ice and is more destructive than rapid thawing. The whole procedure was totally bloodless and took 60 minutes. Good anaesthetic team was a great help in handling the baby intra-operatively and post-operatively. The procedure was totally safe as it was a closed procedure. Post-operatively facial deformity and sloughing was extensive.

Out-come and Follow-up

The patient was followed up for more than 3years. The lesion was decreased with very small area of haemangioma still persisting. There was slight scarring with hypo-pigmentation of the skin. There was no regrowth of lesions in any case after cryotherapy during the follow-up period. All of the adverse effects were minor and negligible. It was reported that melanocytes are more affected by cold than keratinocytes. This may explain the hypopigmentation as a complication of cryotherapy, but were self-limited and improved during the follow-up period. The procedure was quick, simple, bloodless. Nevertheless, the operator has no control on the post-operative sequelae and the total eradication of the disease.

DISCUSSION

Hemangiomas are the most common benign tumors of infancy occurring in about 5-10% of newborns and infants, being more common in female children and in premature infants⁹.

Although most of them grow rapidly during the first years of life with spontaneous involution, 10-20% require active intervention because of their aggressive growth, especially those involving the periorbital area, parotid region, airways, and anogenital area¹⁰.

Different treatment modalities have been adopted over the years. However, each of these modalities has its own side effects and sometimes potential serious complications. Hemangiomas may have great psychological impact on the parents of the affected children; they may feel panic, fear, sadness, guilt, personal shame, and a sense of loneliness.

A few stubborn, problematic hemangiomas may result in serious disfigurement and dysfunction, and even become life-threatening. In order to not leave disfigurement and psychological sequelae, it is suggested that active treatment should be taken rather than observation. With the advances in modern technology, active treatments not only have definite therapeutic effects, but could also minimize the psychosocial distress caused by the lesions. The present modern and advanced treatment methods of head and neck hemangiomas mainly include drug therapy, laser therapy, and surgery. The treatment plan of hemangiomas should be individual and depend on the primary sites, extent, growing phase of the lesions and techniques available. There is no "gold standard" treatment applicable to all patients, and multidisciplinary management is often needed for best efficacy.



Figure No.1: Pre-operative picture



Figure No.2: Use of Cryoprobe on the lesion



Figure No.3: Extra-oral ice formation



Figure No.4: Intra-oral application of cryoprobe



Figure No.5: Intra-oral ice formation



Figure No.6: First post-operative day sloughing (Extra-oral)



Figure No.7: First post-operative day sloughing (Intra-oral)



Figure No.8: 1 week post-operative



Figure No.9: 1 month post-operative



Figure No.10: 1 yr post-operative



Figure No.11: 3 yrs post-op follow up

CONCLUSION

The cold application treatment has been in use since ancient Egyptian times (3500 BC). Liquid nitrogen is the most popular and effective cryogen used today. It is the most effective because of its low temperature (-195.6°C), easy availability from medical and commercial sources, low cost, and safety of use. On the use of cryotherapy for treatment of cutaneous hemangioma incidence of is seemingly rare. The authors conclude that despite various other advanced treatment modalities Cryotherapy could be still a alternative choice with least available resources, economic constraints for the welfare of the child.

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CONFLICT OF INTEREST

We declare that we have no conflict of interest.

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