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Case Report

Fronto-orbital infantile haemangioma: case report and literature review

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Abstract

Background: Haemangiomas are a benign overgrowth of blood vessels in the skin, made up of cells that form the inner lining of blood vessels. They are soft, elevated swellings on the skin, often characterised by a bright, red surface. **Objective:** The paper aims to report a rare case of fronto-orbital haemangioma and review relevant literature. **Case presentation:** We managed a five-month-old female child who presented with fronto-orbital swelling noticed since birth, and progressive growth has become worrisome to the mother. On examination, it is a bluish mass with slightly discoloured overlying skin, lobulated, non-tender, non-pulsatile, soft, blanching, and compressible. The child was optimised and offered total excision. Histopathological assessment confirmed a cavernous haemangioma. The mother reported an excellent cosmetic appearance. **Conclusion:** Cavernous haemangioma in infants may require surgical excision if rapid growth is observed, with a good cosmetic and psychological outcome.

Keywords: *Haemangioma, Neurosurgery, Paediatric, Scalp lesion.*

Introduction

Haemangiomas are a benign overgrowth of blood vessels in the skin, made up of cells that form the inner lining of blood vessels. They are soft, elevated swellings on the skin, often characterised by a bright, red surface. [1] They may present at birth, express rapid growth in the first six months of life, involute with time, and may or may not cause infiltration or destruction of surrounding tissues. [2,3]

Epidemiology

Their prevalence has been estimated to be between 22% and 30% in preterm babies weighing less than 1 kg, 2% and 3% in the neonates, 5% of all infants, and 10% under age 1 year. [4-6] Haemangiomas are more frequent in girls than boys, ranging from a 3:1 to 5:1 ratio.[1,7-9] They are also common among twins and Caucasians. [7] Location-wise, it may be cutaneous or extra-cutaneous. Haemangiomas can be found in all regions of the body, but they occur most commonly in the head and neck region (60%), followed by the trunk (25%) and then the extremities (15%).[10] Approximately 80% of them are solitary, and 20% can proliferate in multiple sites.

Aetiology

The aetiology remains unknown; they are typically non-inherited, though they may be present in other family members. However, some risk factors have been found to be implicated. It is reported that childbearing age, gestational hypertension, and the infant's birth weight may be related to the formation of haemangioma. [2, 11]

Pathophysiology and Clinical Course

Development of haemangioma is the result of genetic mutations, overexpression of angiogenic factors, and downregulation of inhibitors of angiogenesis. Haemangioma is characterised by endothelial cell proliferation. The pathophysiology has not been well

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elucidated, but there are theories explaining the pathology. The first theory supports the notion that there is overexpression of angiogenic factors, such as vascular endothelial growth factor, basic fibroblast growth factor, and matrix metalloproteinases, with downregulation of some inhibitors of angiogenesis, such as tissue inhibitor of metalloproteinase-I. The second theory is that the presence of liver haemangiomas involves a genetic background of mutations.[12] Genetic errors in growth factor receptors have also been shown to affect the development of haemangiomas. Metalloproteinases can accumulate in the endoplasmic reticulum of the tumour cells, causing self-digestion and vacuole formation. Cavernous haemangioma cells can downregulate Derlin-1. Derlin-1 is a protein that, when overexpressed, induces the dilated endoplasmic reticulum to return to its normal size. The third theory suggests that haemangioma endothelial cells arise from disrupted placental tissue embedded in foetal soft tissues during gestation or birth.[12] Markers of haemangiomas have been shown to coincide with those found in placental tissue. This is further supported by the fact that they are found more commonly in infants following Chorionic villus sampling, Placenta previa, and pre-eclampsia. [13,14] Haemangioma may be associated with POEMS syndrome, PHACES syndrome, and Castleman disease. On gross pathology, a spongy appearance with vascular compartments of many sizes separated by fibrous tissue are findings of haemangioma. On microscopic histopathological analysis, channels lined by benign endothelium containing red blood cells are findings of haemangioma.

Pathology

The histologic appearance of haemangiomas fluctuates with the stage of the life cycle of the tumour and can be divided into the proliferative phase and the involutive phase. The proliferative haemangiomas show a proliferation of endothelial cells. The basal lamina is thickened and multilaminate underneath the endothelial cells and forms syncytial masses with or without lumens. Light micrographic findings demonstrate that a large number of vascular plexuses consist of capillaries, venules, and small veins. The proliferative endothelial cells are active with hypertrophy and a pale staining nucleus. The nuclei show occasional mitotic figures, and the number of mast cells is much higher than in normal tissue. The involuting phase haemangioma demonstrates diminished cellularity with a flattening of the lining endothelial cells. As the endothelium flattens, there is a relative dilation of the vessels supplying the tumour and progressive deposition of perivascular, intralobular, and interlobular fibrous tissue. The basement cell membrane is still multi-laminated, and the number of mast cells gradually returns to normal. Complete involuted haemangioma has a ''sponge-like'' structure, with scattered thin-walled blood vessels lined with flat endothelial cells. The basement membranes remain multi-laminated, and the number of mast cells returns to normal amounts. [13,14]

Clinical Presentation and Course

Scalp haemangiomas are typically non-life-threatening or function-impairing, but a few stubborn, problematic haemangiomas may result in serious disfigurement and dysfunction. [1]

If it grows on the forehead, it could impair vision in either or both eyes, depending on the width of expansion. Disruption of the anterior hairline may occur as the lesion involutes leaving behind a large region of alopecia, and the disruption of the aural anatomy that can become permanent if the lesion is not removed early large scalp IHs are often associated with serious sequelae such as ulceration, bleeding, cardiac failure, as well as long-term outcomes such as alopecia, conspicuous scarring, and even malformation of the cranial vault. It can also cause psychological distress to the patient, parent, or guardian. [12] The natural course can be divided into: Rapid proliferating phase (0-1 yr), Involuting (Quiescence) phase (1-5 yr), and the Involuted phase (5-10 yr) [12]

Proliferation Phase

Rapid growth during the neonatal period is the historical hallmark of infantile haemangiomas. Eighty per cent of proliferation occurs by three months of life, but may last longer.

The most growth occurs during the first 4-6 months of life. The haemangioma becomes elevated and dome-shaped, lobulated, plaque-like, tumoral, or any combination of these morphologies.

During proliferation, rapid growth can lead to exhaustion of the blood supply with resulting ischemia, necrosis, ulceration, and bleeding.[12]

Quiescence Phase

Proliferation slows between 6 and 12 months of life considerably. Following proliferation, haemangiomas enter a slower or no growth phase, known

as quiescence.[12] This phase typically lasts from nine to twelve months of age.

Involution Phase
The final and unique phase of the haemangioma life cycle is involution.[12] This phase is marked by greying of the overlying skin and shrinking of the deeper components. At the final stages of involution, a fibrofatty protuberance may remain. Complete involution in 50% of infantile haemangiomas by age 5 years, 70% by age 7 years, and 90% by age 9 years. [1]

Classification [7,15]

1. Cutaneous
2. Extra-cutaneous

1. Based on the depth of involvement, the simple classification by Waner and Suen of Cutaneous haemangiomas is:

A. superficial haemangioma (located in the papillary dermis),
B. the deep-seated haemangioma (located in the reticular dermis or subcutaneous tissue), and
C. the compound (mixed) type (with both superficial and deep haemangioma characteristics).

2. Sites of Extracutaneous manifestations of haemangiomas include the Liver, Gastrointestinal tract, Larynx, Central nervous system, Pancreas, Gallbladder, Thymus, Spleen, Lymph nodes, Lung, Urinary bladder, and Adrenal glands.

Case presentation
We managed a 5-month-old female who presented to our clinic with a history of a right fronto-orbital swelling since birth. This rapidly progressively increased in size over the ensuing months. Swelling was said to reduce in size temporarily whenever the mother massages it with warm water. It was non-painful, non-ulcerated, nor discharging, no associated head enlargement, no associated swelling on the cranio-spinal axis, nor on other parts of the body. No history of associated deformities. No history suggestive of poor vision, excessive crying, poor feeding, vomiting, seizures, or abnormal body movement.
She was born at term to an 18-year-old primiparous mother. Her antenatal period was not eventful, with no history suggestive of the use of teratogenic drugs in pregnancy or native concoctions.
When examined, we found a cheerful infant, not pale, acyanosed, afebrile, not dehydrated, anicteric, and no pedal oedema. A Right Fronto-orbital mass (figure 1), 10cm x 8cm x 3cm, smooth surface, and a wide base, non-tender, no differential warmth, soft, fluctuant, non-pulsatile, non-emptying, with a mobile skin above it and not fixed to the underlying cranium with an underlying bony defect. There was Ptosis of the Right eye induced by the swelling. Occipito-Frontal Circumference (including the swelling) was 37cm, fontanelles were patent and flat child had normal tone and reflexes for age. Other systemic examinations were normal.

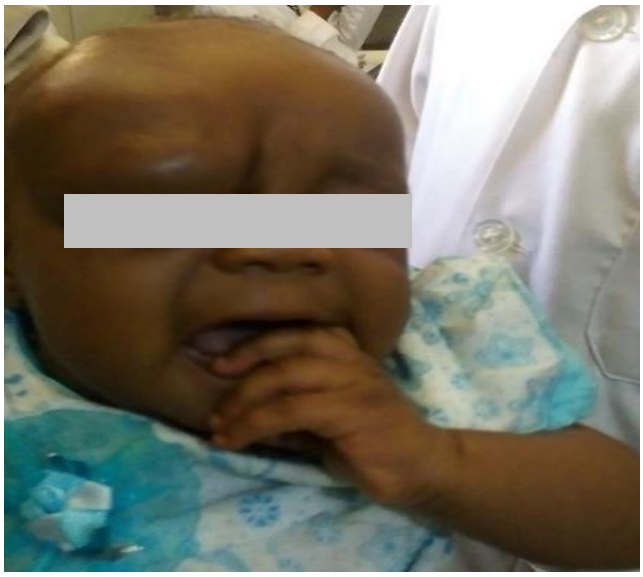
Figure 1A: right fronto-orbital swelling

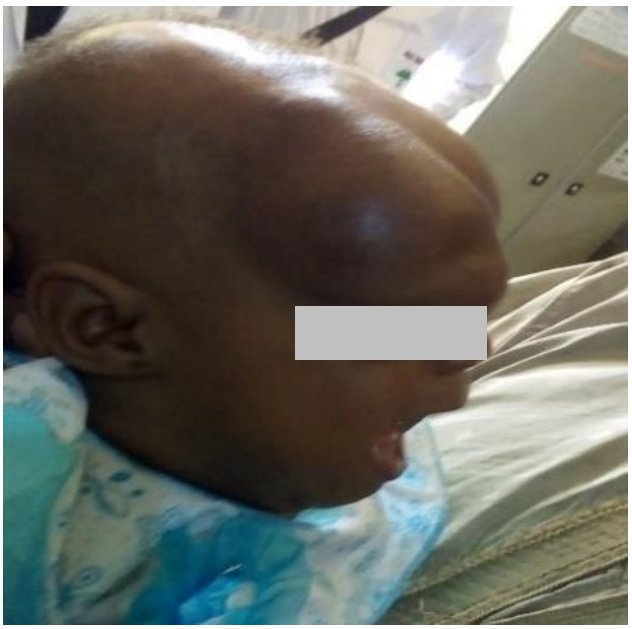
Figure 1B: right fronto-orbital swelling

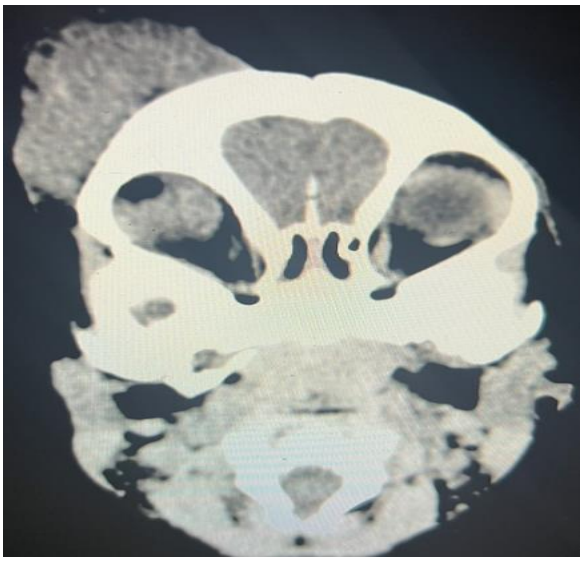

Figure 1C: Axial cut CT scan showing right fronto-orbital isodense mass.

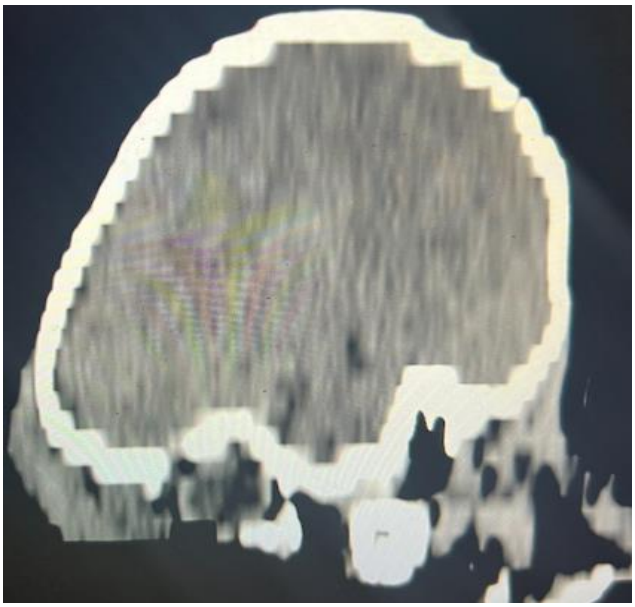

Figure 1D: Sagittal view CT scan showing right fronto-orbital isodense mass.

Brain CT scan (figure 2A & B) showed a soft tissue swelling at the right fronto-orbital region, having a heterogeneous density but isodense, with minimal uptake of contrast, an intact underlying bone, and normal brain. A diagnosis of fronto-orbital haemangioma was made and had total excision.

Figure 2A: intraoperative image just before surgery.

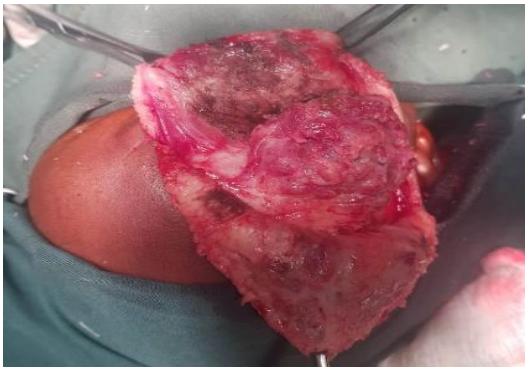

Figure 2B: Dissected tissue

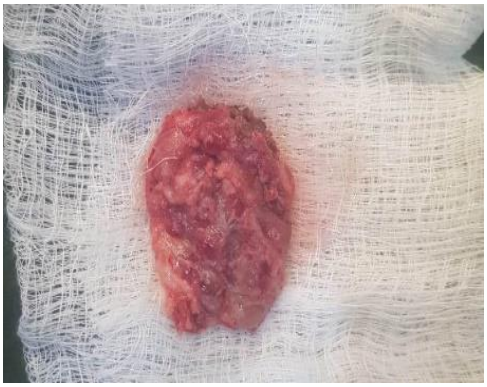

Figure 2C: Excised tissue.

Figure 2D: Immediate post-surgery image

The child had a normal postoperative recovery, histopathology confirmed cavernous haemangioma and was regular on follow-up visits (figure 3).

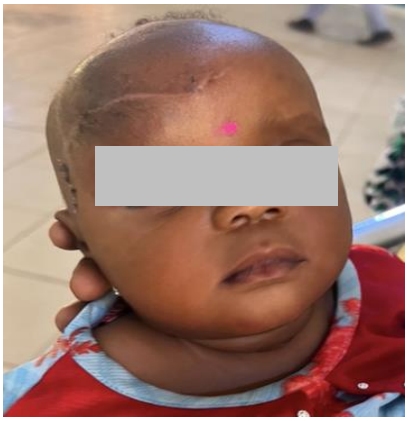

Figure 3: six (6) weeks post-operative period.

Discussion

Infantile haemangiomas are said to be the commonest tumours of infancy. They are benign vascular neoplasms. They commonly occur as birthmarks but grow progressively before regressing. Most times, no treatment is needed unless it ulcerates, gets infected, or causes functional impairment. They are rarely noticed at birth. Our patient, however, was noticed at birth and was located on the fronto-orbital region, causing ptosis of that eye and impairing the child's vision. As reported in the literature, haemangioma could involve the eyelids or part of the craniofacial area. [15] The rate of growth of our index case was quite worrisome to the mother, who, on presentation, was crying at the clinic due to the gross cosmetic disfigurement, the fear of its continuous progressive enlargement, and the fear of it affecting the child's brain and other functions. This was her first child, and she was already getting depressed.

Cranial computed tomography scan shows fronto-orbital iso-hyperdense lesion with no intracranial or intra-orbital extension. The child was noted to be anaemic. Anaemia was corrected by blood transfusion. Treatment aims to prevent life-threatening complications, disfigurement, and psychosocial stress to parents. Several treatment options were suggested depending on the characteristics of the haemangioma and associated complications. Treatments include pharmacologic agents such as timolol and propranolol, and surgical extirpation. [18,19] In the index case, surgery had to be considered first because of the functional impairment, gross cosmetic disfigurement, and psychological compromise to the mother. At surgery, the findings on clinical assessment and radiological investigation were confirmed, the lesion was completely excised with an incision made from the midline at the level of the glabella extending laterally towards the right squamo-temporal area, and the adjoining normal skin was closed primarily.

The outcome was a happy infant who could see clearly with even happier parents.

Conclusion

Scalp haemangiomas are benign vascular neoplasms that are commonly seen in infancy. Their presentation may mimic other congenital abnormalities such as encephaloceles, dermoid cysts, etc. They seldom require treatment as the majority regress spontaneously. However, once a diagnosis is confirmed and surgery is indicated, surgical excision confers an excellent outcome.

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