A RARE CASE OF CAVERNOUS HAEMANGIOMA FACE

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PRESENTATION OF CASE

A 60-year-old male presented to ENT OPD with a swelling on the left side of face for past 30 years. This was insidious in onset, slowly progressive, not associated with pain, visual disturbance, bleeding from the swelling, facial deformity or impairment of hearing. He had a small ulcer over the swelling for past 2 months and hunchback for past 20 years. He had no history of diabetes mellitus, hypertension, tuberculosis, bronchial asthma, radiation exposure, trauma, surgery in the past or any other intervention.

His personal history was non-contributory with no history of substance abuse or industrial exposure. None of his family members ever suffered from neurofibromatosis or haemangioma or any other congenital anomalies. His general physical examination revealed pallor and kyphosis at upper lumbar region without any lymphadenopathy, café au lait spot or inguinal freckles. His systemic examination was within normal limits and there were no Lisch nodules.

On local examination, the tumour was dark brown multinodular soft tissue mass approximately 12 × 8 cm^2 involving left zygoma, preauricular groove, 2 cm above ramus, the mandible, left angle of mouth, nasolabial fold and infraorbital margin. It was firm, noncompressible with a non-bleeding small ulcer with inverted edge having purulent discharge. No visible blood vessels or any pulsation or bruit. The entire tumour was mobile at subcutaneous plane. ENT and neck was within normal limits.

Routine blood examination revealed normocytic normochromic anaemia with HBsAg positive. X-ray LS spine suggested lumbar kyphosis and other routine investigations and pure tone audiometry were within normal limits.

FNAC from the tumour yielded blood and fibrous tissue, thus inconclusive. Radiology supported a superficial tumour.

With a preoperative differential diagnosis of neurofibroma, fibroma and haemangioma, this case is rare because of the adult-onset multinodularity with atypical pattern, gigantic size and diagnostic surprise.

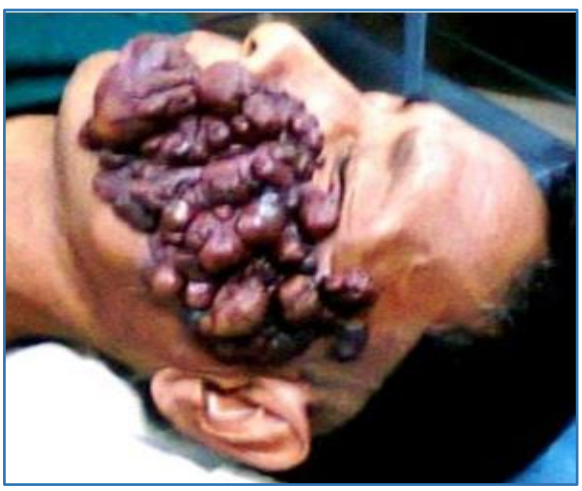

Figure 1. Preoperative Picture

Figure 2. Immediate Post-Operative (wound Closure with SCG)

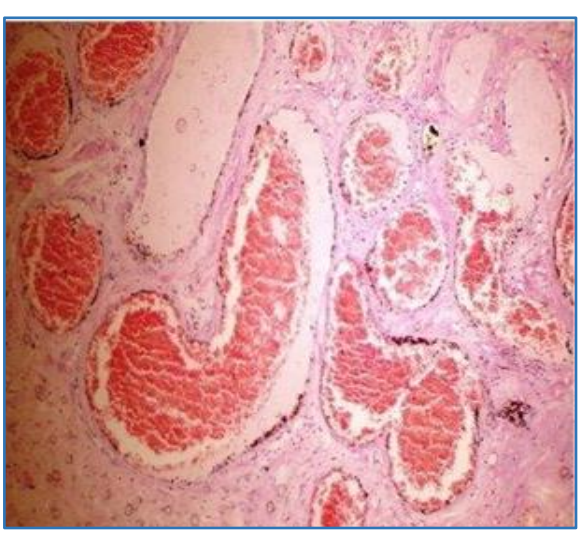

Figure 3. Histopathology

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Figure 4. Postoperative Picture

DIFFERENTIAL DIAGNOSES

1. Neurofibroma.
2. Fibroma.
3. Haemangioma.

CLINICAL DIAGNOSIS

Haemangioma of face.

DISCUSSION OF MANAGEMENT

Haemangiomas are a group of vascular tumours, which are rarely present at birth. They have rapid growth during first six months of life and then mostly involutes with full resolution.[1] They are prominent, red and circumscribed lesions with female preponderance (3:1). Commonly, these are isolated lesions (80%), but might be multifocal.[2] It is seen more commonly in fair skinned female child with low birth weight.[3] Haemangiomas can be classified as superficial haemangioma (capillary haemangioma), deep haemangioma (cavernous haemangioma) or compound haemangioma (capillary cavernous haemangioma). Cutaneous haemangiomas are seen at cephalic extremity (60%), trunk (25%) and extremity (15%). Non-cutaneous haemangioma may involve liver, gastrointestinal tract, larynx, etc.[2]

The diagnosis is usually made by detailed history and clinical examination. Radiology in the form of ultrasound with Doppler or MRI maybe useful.

Management is primarily close observation, but around 40% children require some intervention either medical, surgical or both during its course.[4] Corticosteroids, interferon and vincristine are effective agents for medical management.[5,6,7]

Surgical management includes excision or laser treatment with or without intralesional steroid.[8]

This case was planned for excision with thigh Split Skin Graft (SSG). Incision was given all around the tumour with 1 cm margin. Dissection was done at subcutaneous plane. No major blood vessel encountered during surgery. Tumour was resected completely, which measured 12.5 × 8 cm^2. After mobilisation of facial skin, residual defect was repaired with thigh SSG. Histopathologically reported this as cavernous haemangioma. Patient had an uneventful postoperative recovery.

This case is particularly interesting because of its onset in an adult male, which is very rare as haemangiomas are tumour seen in infancy predominantly in a female child. Most of the haemangiomas involutes by 9 years of age.[8] The gigantic size of this facial haemangioma is also not commonly seen as most of them are around 2-4 cm in greatest dimension.[2]

The diagnostic surprise is also an interesting fact as the tumour in gross appearance closely resembled neurofibroma. It also fulfilled the diagnostic criteria of neurofibroma as patient also had hunchback (lumbar kyphosis).

REFERENCES

[1] Gresham T, Adva BF. International Journal of Pediatrics 2012:1-10.
[2] Sas TN, Boutsiadis N. Facial haemangiomas diagnosis. Current Health Science Journal 2010;36(3):166-170.
[3] Haggstrom AN, Drolet BA, Baselga E, et al. Prospective study of infantile hemangiomas: demographic, prenatal and perinatal characteristics. Journal of Pediatrics 2007;150(3):291-294.
[4] Haggstrom AN, Drolet BA, Baselga E, et al. Prospective study of infantile hemangiomas: clinical characteristics predicting complications and treatment. Pediatrics 2006;118(3):882-887.
[5] Bauman NM, Burke DK, Smith RJH. Treatment of massive or life-threatening hemangiomas with recombinant α2a-interferon. Otolaryngology - Head and Neck Surgery 1997;117(1):99-110.
[6] Perez K, Pardo J, Gomez C. Vincristine-an effective treatment of corticoid-resistant life-threatening infantile hemangiomas. Acta Oncologica 2002;41(2):197-199.
[7] Pope E, Krafchik BR, Macarthur C, et al. Oral versus high dose pulse corticosteroids for problematic infantile hemangiomas: a randomized, controlled trial. Pediatrics 2007;119(6):e1239-e1247.
[8] Buckmiller LM, Francis CL, Glade RS. Intralesional steroid injection for proliferative parotid hemangiomas. International Journal of Pediatric Otorhinolaryngology 2008;72(1):81-87.