

A Rare Case Report of Parotid Haemangioma in a Child Treated by Sclerotherapy Using Bleomycin

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Abstract

Salivary gland haemangioma are rare in adults but more common in children comprising about 50% of salivary tumours in children. The patient may present with a swelling or may be asymptomatic. Diagnosis is made by ultrasound or Magnetic resonance imaging. Treatment is best by excision for small variants but other options of sclerotherapy or lasers or radiotherapy or cryosurgery should also be considered in large ones. We discuss the case of a 7 year child with right sided parotid haemangioma which was treated successfully by sclerotherapy with bleomycin.

Introduction

Salivary gland cavernous haemangiomas are relatively uncommon in children and even rarer in the adult, and provide diagnostic difficulties. This paper demonstrates how the diagnosis can be established and availability of different spectrum of treatment modalities. Our patient was treated with bleomycin sclerotherapy followed by regular follow up for regression..

Case Report

Our patient a 7 year old male child weighing 15 kg resident of Akola, Maharashtra came with chief complaints of right sided cheek swelling since birth gradually increasing in size. However significant increase since last 4 yrs to present size not associated with pain or redness was noted by his mother. There was no history of tuberculosis, convulsions, cyanotic spells, or any significant history. The patient had a full term normal hospital delivery, cried well at birth and immunised till date. No history of similar complaints in the younger female sibling was noted.

He took treatment for the same 4 years back where he was investigated by Ultrasound Neck and treated with 4 cycles of injection sclerotherapy with STD (sodium tetradecyl sulphate) followed by decrease in size (25%) but no complete relief. On examination general condition was fair and he was haemodynamically stable. On local examination there was a soft ill-defined non pulsatile nontender compressible swelling which was slightly red in colour, in right parotid region involving deep and superficial part size 6 x 5 cms (Fig.1). Ultrasonography was suggestive of a parotid lymphangioma/haemangioma. CT scan revealed it as haemangioma/vascular malformation (Fig 2). Patient underwent single sitting of injection sclerotherapy with bleomycin 15 units under sedation. Post procedure course in the ward was uneventful. Patient was given a course of antibiotics and analgesics post procedure There was 75% reduction in the 1st month (Fig 3). The patient was followed up at 1 monthly regular intervals with almost 95% reduction in size with no symptoms/ complications.

Discussion

Salivary gland lesions are uncommon in children and may be related to the parotid, submandibular or sublingual glands. Inflammatory lesions are the most common cause of salivary gland abnormalities in children and can be due to acute viral, acute

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Fig 1 : Prechemotherapy photograph showing right sided parotid swelling.

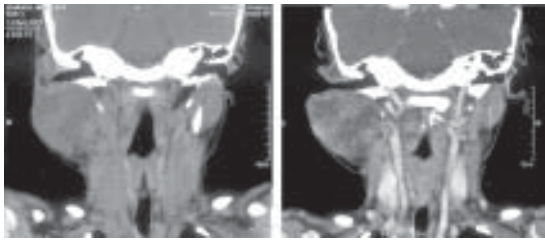


Fig 2 : CT Scan showing the right sided parotid region swelling involving deep and superficial part size 6 x 5 cms with vascular enhancement post contrast confirming a haemangioma.

suppurative or recurrent acute or chronic inflammation. Intraparotid lymphadenitis may also occur, as in cat-scratch disease or in other causes of cervical lymphadenitis. Salivary gland neoplasms are rare in children, and most of them are benign including mainly haemangioma, pleomorphic adenoma, or lymphangioma.

Parotid gland neoplasms are uncommon and account for less than 2% of human tumours. Haemangioma account for about 50% of parotid tumours in infants and children, particularly in females as compared to only 2% in adults. Whereas both capillary and cavernous haemangioma are seen in children, only cavernous haemangioma have as far been reported in adults. As the clinical course of infantile haemangiomas may be self-limiting and the initial treatment is expectant, early and accurate diagnosis is



Fig 3 : Post chemotherapy with bleomycin showing about 75% reduction in the size of the swelling in 1 month.

essential.

Parotid or oral haemangioma cases accounted for about 5 cases in a case study by Weber *et al* in 1990.² The association of juvenile haemangioma of the parotid gland and cytomegalovirus (CMV) is extremely rare: a single case was reported in the literature.³ The pathologic significance of this association is still discussed, although several experimental studies suggested a relationship between the infection by the cytomegalovirus and this tumour.

Clinically, cavernous haemangioma usually present as slowly growing, soft or firm, movable, painless parotid masses. Severe pain and swelling can occur, however, depending on the size of the haemangioma or in particular in the presence of acute haemorrhage or thrombosis.

The diagnosis is suggested by physical examination in all patients and can be confirmed by imaging modalities. Ultrasonography should be the initial imaging study used for the examination of salivary gland lesions in children, given the fact that most of such lesions are benign and picked up clearly by sonography. In most cases, this technique permits the differentiation of intraglandular and

extraglandular lesions, and may suggest the correct diagnosis. Vascular lesions can be demonstrated more clearly using Colour Doppler. Some of the lesions may appear similar, and clinical correlation is important for the differential diagnosis. In such cases it can be confirmed either by histological or imaging modalities like MRI or CTscan either plain or with contrast angiography. MR digital subtraction angiography differentiates a low flow lesion from a high flow lesion that is not always suspected by clinical examination alone (as in our case) and a hypo vascular tumour for which a biopsy can be safely performed as compared to a hyper vascular tumour. MR digital subtraction angiography can be reliably performed in children of all ages without complication, providing a non-invasive assessment of the vascularity of each lesion that could not always have been predicted on the basis of clinical examination or routine MR imaging alone.

The treatment needs to be individualized, usually progressing from less to more invasive, and includes observation, prednisone therapy, interferon,⁴ sclerotherapy, radiotherapy, lasers, cryotherapy, arterial ligation, and resection. Treatment is commonly delayed, as there is a significant likelihood of spontaneous regression as seen at most centres. A review of the clinical, histological, and treatment details of 10 cases of haemangioma and one case of lymphangioma that involved the parotid gland by Mantravadi et al in 1993 advised a watchful expectancy for spontaneous regression and preservation of the facial nerve at surgery.¹ Histologically, the cellularity and increased division figures in these lesions should not be interpreted as a sign of a malignant condition. The indications for surgical intervention are increase in tumour size, rapid tumour growth, failure of tumour size to decrease and haemorrhage

into the lesion. Prior to surgery, magnetic resonance angiography or intra-arterial digital subtraction angiography should be performed to investigate the vascular supply of the tumour. Surgical excision is the treatment of choice for small lesions. Large cavernous haemangioma usually require superficial or total parotidectomy. Especially in the case of extended lesions, the facial nerve may be difficult to identify and should be monitored intraoperatively.

All children are eventually cured, with minimal morbidity. Children with life-threatening haemangiomas can be successfully managed with the use of a variety of techniques. No effective medical treatment has been reported for children with large, deforming haemangiomas of the parotid gland and overlying cheek. Surgical resection of parotid haemangioma provides an aesthetic benefit to young children with low associated morbidity. Early resection by an experienced surgeon should be considered as a treatment option for these disfiguring lesions.⁵

The disadvantage of steroid and radiation treatment is long-term treatment, growth retardation and facial disfigurement. Treatment with interferon has poor results and is costly. Nowadays newer modalities include injection sclerotherapy with bleomycin, lasers and cryotherapy; the latter two requiring a costly setup. Sclerotherapy done with bleomycin is safe and under radiological control. It takes into account the potential of bleomycin to cause fibrosis in the vascular channels followed by regression in the swelling size. Injection sclerotherapy seems to be a better option in that it is easier to perform and is cheaper, done on outdoor basis, done under fluoroscopic or ultrasound guidance.

Conclusion

Vascular neoplasm's of the parotid are

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generally diagnosed with high degree of suspicion and confirmed by USG / CT or MR Angiography. The treatment needs to be individualized; usually progressing from less to more invasive procedures. Injection sclerotherapy including bleomycin can be used for the regression of the haemangioma successfully and safely. Early resection by an experienced surgeon should be considered as a treatment option for disfiguring and life threatening lesions. All children are eventually cured in majority of the cases, with minimal morbidity may it be by watchful expectancy, sclerotherapy or surgical resection.

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CORONARY ARTERY DISEASE IN INDIA : CHALLENGES AND OPPORTUNITIES

In today's Lancet, Denis Xavier and colleagues describe the presentation, treatment, and outcome of more than 20000 patients with acute coronary syndromes admitted to 89 hospitals across 50 cities in India. Patients in India were more likely to be younger and present with ST-elevation myocardial infarction than those in the registry data from developed countries.

Tobacco use, dyslipidaemia, and hypertension are the main determinants of population attributable risk worldwide.

Even a modest change from saturated to polyunsaturated fat can achieve dramatic reductions in cardiovascular mortality, as has been evident in Poland. Control of hypertension and hyperlipidaemia.

Indian hospitals have access to the most modern technologies on a par with the rest of the world and can provide the latest and the best medical care as long as the patient can pay.

Kim Eagle, The Lancet, 2008; 371 : 1394-95.