

## Case Reports

### CYSTIC HYGROMA: A PRESENTATION OF THREE CASES IN THE OROFACIAL REGION

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ثلاث حالات للورم الكيسي الزلالي عولجت في قسم جراحة الفم والوجه والفكين وتم شرح طرق تشخيصها ومناقشة أساليب معالجتها.

Three cases of cystic hygroma treated at the Department of Oral and Maxillofacial Surgery are presented. Methods of diagnosis and treatment are discussed.

#### Introduction

Hygroma cysticum coli or cystic hygroma is a division of lymphangiomas that presents mostly at birth, it is therefore a congenital malformation of the lymphatic system.<sup>1,2</sup> Most of these tumors are present by the time the patient reaches the age of 3 to 5 years.<sup>2</sup>

The most common site for the tumor is the posterior triangle of the neck and it may involve vital structures, such as the sympathetic chain, carotid sheath content, and branches of the hypoglossal, lingual, and the facial nerves.<sup>2</sup> Depending on the anatomical site, cystic hygroma may cause dysphagia or airway obstruction and respiratory distress that necessitates immediate surgical interference.<sup>3,4,5</sup>

The cystic hygroma in the neck is manifested as large, deep, diffuse swelling.<sup>1,5</sup> On palpation, it is often doughy and is usually transilluminant. These lesions are composed of lymph containing endothelial spaces that vary in size from capillary dimensions to cysts of several centimeters in diameter.<sup>6</sup> Although, the embryologic origin is still unclear, failure of primary lymph spaces to join the central lymphatic system (thoracic duct and right lymphatic duct) and the venous system may be the basis for their development. Cystic hygroma is usually multiloculated and is composed of spaces.

These spaces contain straw colored fluid which may be discolored with secondary infection of intracystic hemorrhage.<sup>7</sup>

It may also cause obstructive sleep apnea. Such apnea is a different sign of presentation than the ones that are commonly observed. Commonly, the hygroma manifests itself as a uni- or bilateral swelling of the neck that causes asymmetry.<sup>8</sup> Although surgical excision of the tumor, is the treatment of choice for most clinicians, local injection of bleomycin fat emulsion could be another success-ful alternative.<sup>9</sup>

In this report, we are presenting three cases of cystic hygroma that were treated since August 1990. Two of the cases were completely excised, while the third one was partially excised. Facial nerve palsy was not noticed in any of them post-operatively. Hospitalization period ranged from 5-17 days.

#### Case 1

A ten-year-old Saudi female referred to our department at Riyadh Dental Center (RDC) with a large swelling of the right parotid and submandibular regions. The referral diagnosis was residual buccal fat. The swelling existed for 5 years. When the patient was 6 years old, the lesion was diagnosed as "lipoma" and was operated by a general surgeon in order to remove it through an intra-oral approach, but the treatment was not successful.

Examination of the patient revealed soft, diffuse, mobile large swelling over the right parotid and submandibular regions [Fig. 1]. Transillumination test was positive. Clinical diagnosis of cystic hyg-

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roma was made. CT scan showed the extent of the tumor [Fig. 2].



Figure 1. Photograph showing big diffuse swelling over the parotid and submandibular regions.

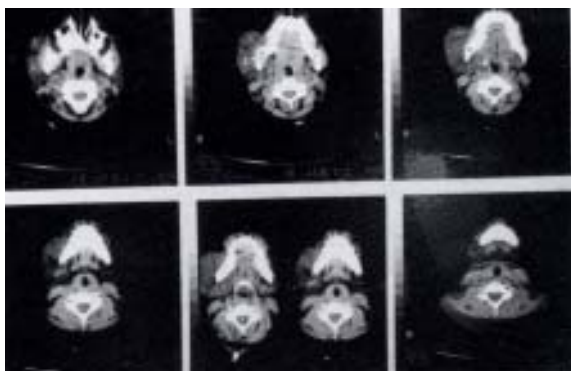


Figure 2. CTscan showing the extent of the lesion and its boundaries.

The patient was then admitted to RDC for surgical removal of the lesion. Through a parotidectomy incision, about 70% of the lesion was excised, leaving the deep (medial) portion. The facial nerve branches were identified and preserved. The patient was hospitalized for 17 days and was discharged in a satisfactory condition. There were no facial nerve deficit. Unfortunately, the central part and tip of the elevated skin flap suffered a deep second degree burn due to application of hot compress during the first two days after surgery which required further management.

### Case 2

A three-year-old Kuwaiti female presented to the Oral and Maxillofacial Surgery Clinic, RDC with a large swelling of her left submandibular and pre-

auricular region. The swelling was asymptomatic except for the disfigurement [Fig. 3]. It was present at birth and increased in size subsequently. Upon examination, the swelling was soft, diffuse, about 6 cms. in diameter. Transillumination test was positive. Aspiration of the lesion gave clear, yellow fluid. A CT scan was requested and showed a cystic lesion with clear boundaries. A diagnosis of cystic hygroma was made. A decision was made to surgically remove the tumor. The patient was admitted to RDC, and the lesion was excised completely through a modified pre-auricular incision. The facial nerve branches were identified and protected after exposing the trunk just after its exit from the stylo-mastoid foramen [Figs. 4a and b]. The lesion involved the skin and was not dissected easily. The incision was closed in layers after the insertion of a vacuum drain.



Figure 3. Huge swelling of the left side of the face causing severe facial asymmetry.



Figure 4a. Photograph showing the main trunk of the facial nerve dissected out intact with its branches



Figure 4b. The tumor being excised in one piece.

**Case 3**

A thirteen-year-old Saudi boy presented to our department complaining of large swelling over the left preauricular region. The swelling started two years earlier after the patient received trauma to that area. A general surgeon aspirated the lesion on two occasions after which the swelling subsided, to regain its size within a few weeks.



Figures 5a,b. Photographs showing the size and the extent of the tumor.

Clinical examination revealed soft, diffuse swelling involving the left parotid and submandibular regions [Figs. 5a and b]. Aspiration of the lesion gave brownish fluid [Fig. 6]. CT scan was requested and showed large cystic lesion with definite boundaries [Fig. 7].



Figure 6. Brownish fluid that was aspirated from the lesion

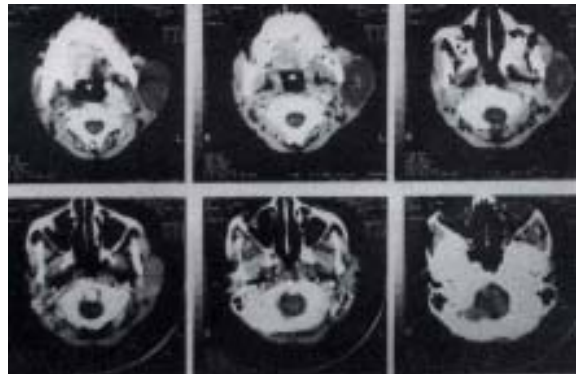


Figure 7. CT scan showing the cystic cavity and its boundaries



Figure 8a. Photograph showing the parotidectomy incision and the tumor uncovered with flap reflected.

The facial nerve was identified, its branches were preserved and the lesion was completely excised through a parotidectomy incision [Figs. 8a and b], and the incision was closed in layers after drain insertion. Post-operatively, there was slight weakness of the frontal branch which recovered completely, in four weeks [Fig. 9]. The patient has had a satisfactory hospital course and left the center on the 5th day post-operatively.



Figure 8b. Complete excision of the tumor



Figure 9. Four weeks post-operative photograph showing complete recovery with minimal post-operative swelling of the left side of the face.

### Conclusion

Surgical excision is the treatment of choice for cystic hygromas. Cystic hygroma in the orofacial region is a difficult lesion to excise. Although the tumor is benign, the intimate relation to the

various important anatomic structures makes its dissection a tedious task.

Involvement of the skin makes it a risk during raising the skin flap and even after successful elevation, the sensibility of the skin is jeopardized which may be dangerous if the patient is instructed to apply hot compresses. Extension of the lesion and involvement of muscle and glandular structure make complete excision sometimes impossible. When complete excision is sought, unnecessary sacrifice of muscles will leave the patient with a deformity which is more difficult to treat as it constitutes a deformity in the form of a defect and a functional one in the form of loss of muscle function. Partial excision may be the solution, even though recurrence is possible following incomplete excision. The goal in the management of cystic hygromas must be primarily to relieve obstruction upon vital tissues and a good cosmetic result.

### References

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