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WEDGE RESECTION AS A SOLUTION TO AIRWAY AND FEEDING PROBLEMS IN CYSTIC HYGROMAS INVOLVING THE TONGUE

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SUMMARY: Two cases of cystic hygroma of the neck involving the tongue are presented and the literature reviewed. A wedge resection of the tongue was performed in both cases to over-come feeding and airway problems.

RESUME: On a rapporté 2 cas d'hygrome cystique de cou qui comporte la langue en meme temps et on a observe la litterature. Pour resoudre les problemes d'alimentation et de la voix respiratoire, on a appliqué a la langue "wedge resection" a tout les deux cas.

ZUSAMMENFASSUNG: Zwei zystic-hygroma Falle mit Zungeninvazion sind in diesem Artikel vorgestellt und die Literatur ist durchgesehen. In jeden Fallen wedge rezektion von der Zunge durchgeführt, um Ernahrung- und Atmungsprobleme zu beseitigen.

The most common site for cystic hygroma is the neck (6,7,9). They usually lie in the supraclavicular fossa of the posterior triagnle. Less frequently they are located in the anterior triangle just below the angle of the jaw, where they can present significant airway and feeding problems (6). Tongue is involved much less, causing serious airway and feeding problems. In the cases presented the tumours were located in the anterior triangle and the tongues were involved.

CASES

The two cases were 1.5 month old and 1 month old respectively. They both presented with a huge mass in the neck and the tongues were exceeding out of the mouth. The mothers, both, had an uneventful pregnancy and delivery. The routine blood and urine analyses were of no significance. The patients' feeding were through oral feeding tubes and they were to be kept in intensive care unit for airway problems.

The first patient, F.Y., 1.5 month old, presented with a large mass of about 12x8 cm. in the neck and the tongue was exceeding out of the mouth, making feeding impossible and narrowing the airway. An extirpation of the mass without sacrifying vital structures and a wedge resection to the tongue was performed at the operation. The post-operative course was uneventful and the patient was discharged at the seventh post-operative day able to be fed and breathe normally. The routine controls during the last two years revealed no recurrence (Figures 1, 2).

The secon patient, M.G., 1 month old, presented with a huge tongue exceeding out of his mouth, causing airway and feeding problems. He was operated for the mass in the left side of his neck in pediatric surgery department of our faculty. A wedge resection of the tongue was performed and the patient was discharged on the fifth post-operative day. There was no recurrence in the follow-up period of about 1 year (Figures 4, 5, 6).

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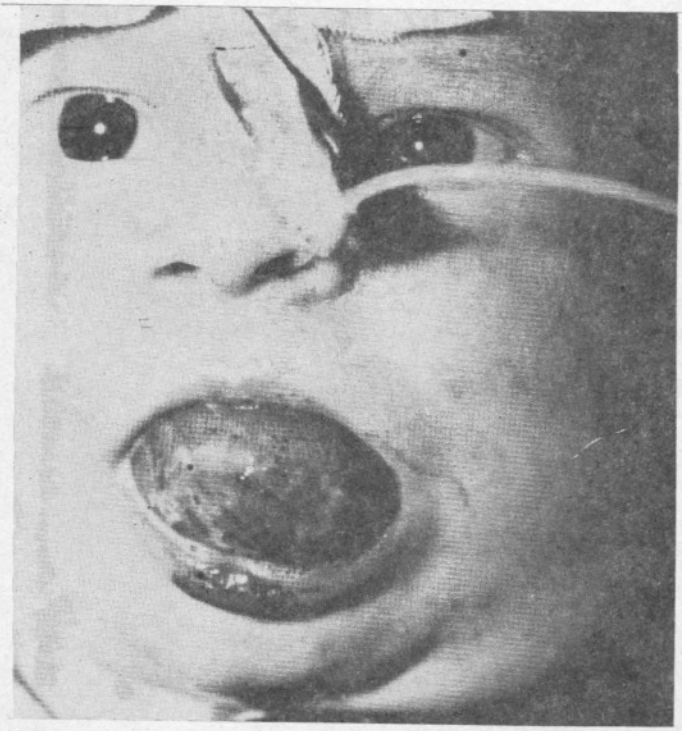

Figure 1. Pre-operative view of the first patient.

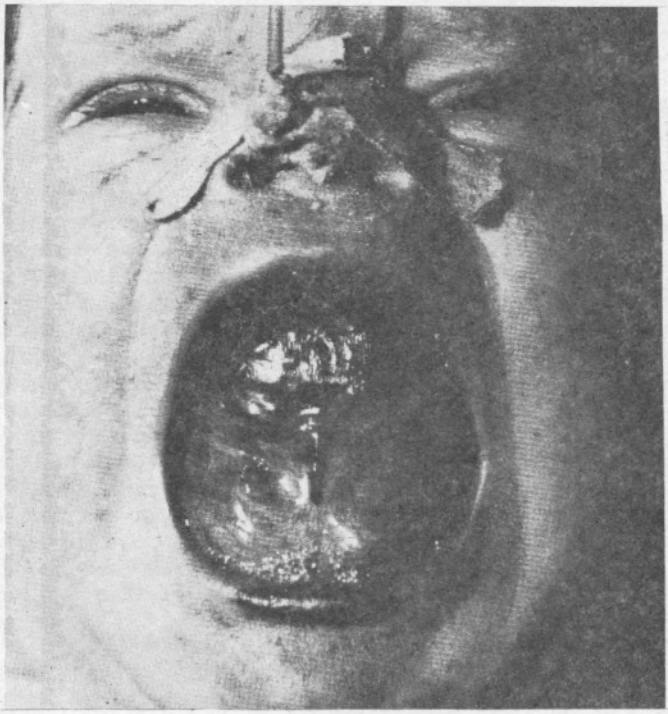

Figure 2. Post-operative view of the same patient.

DISCUSSION

The lymphatic system makes its appearance during the sixth week of embryonic development (7). There are two theories about its development. According to Sabin, it arises as an outgrowth from the venous system while the others claim it differentiates de novo from adjacent mesenchyme (5,6,7). In every case, it is clear that the lymphatic

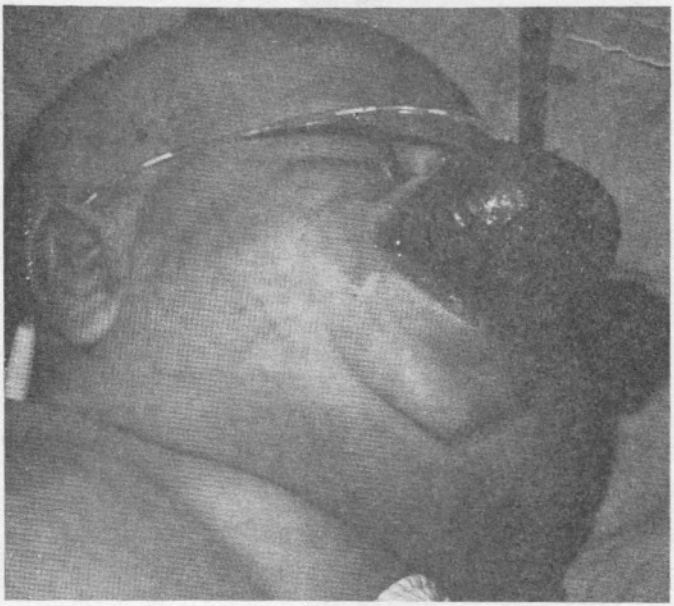

Figure 3.
Pre-operative view of the second patient.

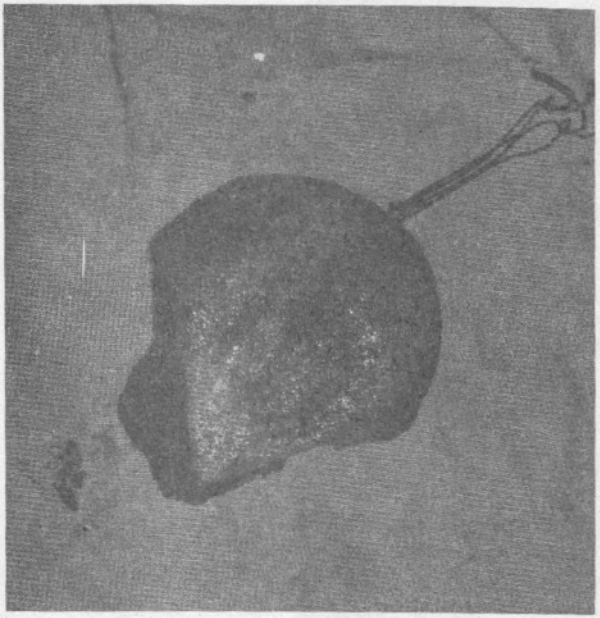

Figure 4. Pre-operative view of the resected tongue piece.

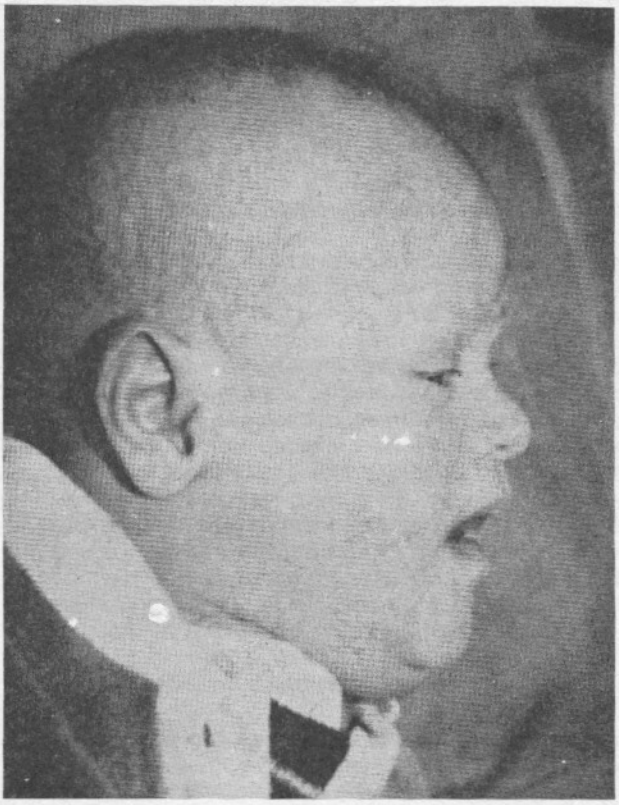

Figure 5. Post-operative view of the same patient.

system and parallels the venous drainage of an organ (7).

The definition of lymphangiomas can present differentiations as with hemangiomas. It is controversial as to whether they must be defined as true neoplasms, hamartomas or just lympangiectasias. Actually, since they are all benign lesions and the therapy is largely dictated by their location and clinical extent, these distinctions are of little practical value. Generally, they are regarded as malformations which arise from sequestrations of lymphatic tissue that lack normal communication with the lymphatic system. These remnants, possessing also some capacity to proliferate, accumulate great amounts of fluid.

Lymphangiomas are divided into three groups, viz: lymphangioma simplex or capillary lymphangioma, cavernous lymphangioma and cycstic lymphangioma. But, this classification has no clinical value. Some lymphangiomas have both cystic and cavernous components and long-standing cavernous lymphangiomas can be converted to cystic lymphangiomas. Cystic lymphangiomas arise in areas such as neck and axilla where loose connective tissue is suitable for expansion while cavernous lymphangiomas develop in the mouth, lip, cheek, tongue where dense connective tissue and muscle prevent expansion (10).

Lymphangiomas are relatively rare. Their incidence is about % 0.14-6 (1,2,5,6,7, 11). The most common site for cystic hygroma is the neck. Less frequent tsites are the mediastinum, axillary, inguinal and retroperitoneal regions. The sex inci-dence is nearly equal

(6,7). It is estimated that % 50-65 of these tumours are present at birth, and as many as % 90 may be manifest by the end of second year. Mousattas tas and Baffes found 27 cases among 267 patients treated surgically for cervical masses (6).

The common clinical feature is a soft mass in the posterior triangle of the neck, in the supraclavicular fossa. It is usually fluctuant, lobulated and not attached to skin but fixed to the deep structures and generally transilluminates. Less frequently tumour can be located in the anterior neck triangle and in this location it can give rise to airway and feeding problems. The mass may enlarge, remain the same size or regress. Sudden enlargement following infection or trauma may occur. Differential diagnosis with branchial cleft cysts and lipomas must be made.

Many of the cystic lymphangiomas involving the face are cavernous type. Cystic hygromas can occur in association with hemangiomas (Maffucci's syndrome). They have been shown to be associated with hydrops fetalis, Turner's syndrome and in utero high death rate (3,4,7). Because of this, in utero detection by ultrasound is helpful. Noonan's syndrome, familial pterygium colli, fetal alcohol syndrome and several chromosomal aneuploidies may also be associated with cystic hygroma. We have not detected any sign in favor of these syndromes in our cases. Treatment of choice is early surgical excision as it is easier to remove the tumour before it further invades normal tissue. The dissection is difficult and tedious and it is wiser to leave small bits of cysts then to divide important structures especially when it invades the jugular vein, carotid artery and nerves of the carotid sheath, although remaining cysts increase the recurrence rate, which is said to occur within one year. Recurrence can be minimized by opening any cysts left behind. Ravitch and Rush reported a recurrence rate of % 10-15, when portions of the cysts left behind (6). The cavernous lymphangiomas in the tongue have a tendency to insinuate between muscle fibers and are complicated by a high recurrence rate and nerve palsies (2).

RT should not be used as there care cases of malignant transformations of previously irradiated lymphangiomas (8).

A wedge resection of the tongue was performed in both cases to overcome feeding and airway problems with extirpation of the mass in one case. The other case was operated for his tumour in pediatric surgery department. The results were satisfactory, the patients were able to be fed orally 3 days after the operation and breathe normally. We have not met any recurrence neither in the necks nor in the tongues during the follow-up period of one year. The tongues are of normal function. As a conclusion we can state that in cystic hygromas treatment of choice is early conservative surgical excision and wedge resection of the tongue gives satisfactory results in the presence of airway and feeding problems caused by its involvement.

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