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Cystic Lymphangioma of the Parotid Gland in Children

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Case Report

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Abstract: Cystic lymphangioma of the parotid gland is an uncommon congenital lymphatic malformation that rarely appears in adults; it's usually detected in the first two years of life. We report and discuss two cases of intra-parotid cystic lymphangioma that occurred in children. A young boy of 12 years old and a seven-year girl were admitted in our unit because of an indolent and renitent isolated mass occupying parotid region. The diagnostic of an intra-parotid cystic lymphangioma was clinically advocated on echographic and CT imaging findings and then definitely proven after histopathologic studies of the removed tumour. A total parotidectomy was made, removing the entire tumour. The parotid localization of cystic lymphangioma makes their surgical management difficult because of the development of lymphangioma between the branches of the facial nerve. The question of the benefit / risk ratio makes the therapeutic decision more complex because the aggressiveness of the radical tumour resection encounters a benign tumoral origin. It opposes two fundamental principles, first that of being radical on the tumour which is very recurrent and on the principle of remaining functional while preserving the function of the facial nerve.
Keywords: Cystic, lymphangioma, parotid, children, parotidectomy.

INTRODUCTION

Cystic lymphangiomas of the parotid gland are benign vascular tumours occurring rarely in the adult population. They can be seen anywhere in the body, but most often in the cervicofacial region, especially in the posterior triangle of the neck [1, 2]. They are more frequent during the first 2 years of life and both sexes are affected equally.

Their occurrence is due to an embryonic developmental anomaly of the lymphatic system [2]. Herein we report on two cases of intra-parotid cystic lymphangioma that occurred in teen age.

CASE REPORTS

Case 1

A young boy of 12 years age, with no specific history, was admitted because of a mass over the angle of the left mandibula (6cm), the tumour has been present since the first months of birth and has apparently increased in size without the development of obivious symptoms referable to its presence (figure 1a). Computer tomography studies exhibited a cystic structure in the left parotid (figure 1b,c). A left total parotidectomy was done removing the entire tumour together with a flap positioning (SMAS). Post-operative course was uneventful and after a follow-up period of 18 months, the patient still remains in good condition (figure 1d).

Case 2

A seven-year girl was admitted in our unit because of an indolent and renitent isolated mass occupying the right parotid region, present at birth and characterized by iterative inflammatory episodes (figure 2a). The diagnostic of an intra-parotid cystic lymphangioma was clinically advocated on echographic and CT imaging findings (figure 2b,c) and then definitely proven after histopathologic studies of the ablation fragments. A total parotidectomy was made, removing the entire tumour associated with a flap positioning (SMAS). Post-operatively, the child presented with labial paresis, which was recovered during a few weeks, after a 24-month follow-up with no recurrence or facial paresis (figure 2d).

Case 1

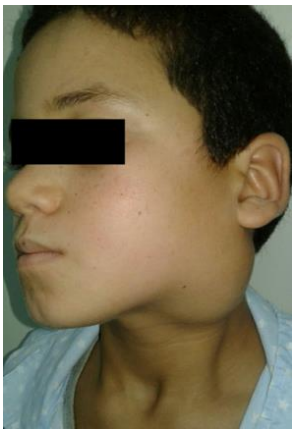


Fig-1a : Masse of the left parotid region

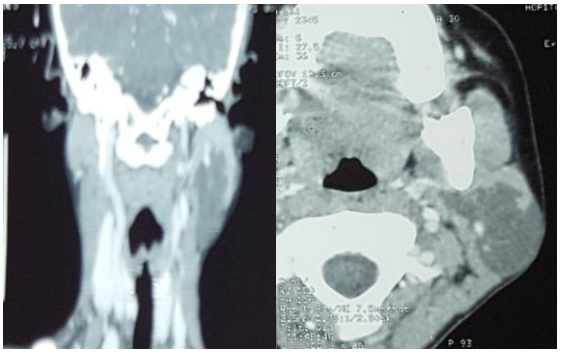
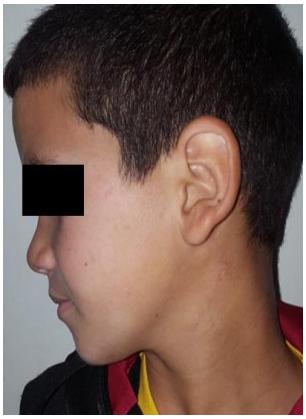


Fig-1b,c : CT showing a cystic structure in the left parotid



Fg-1d :resalt after a 18-month follow-up

Case 2

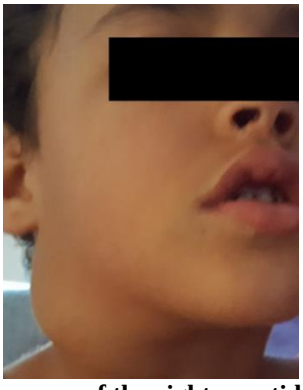


Fig-2a: masse of the right parotid region

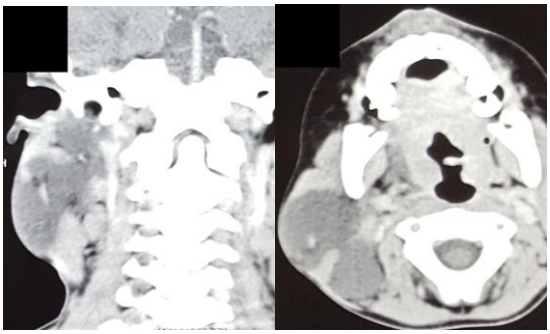


Fig-1b,c : CT showing a cystic structure in the right parotid

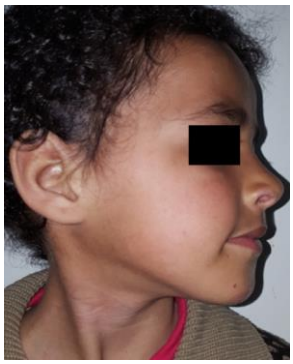


Fig-2d : after a 24-month follow-up

DISCUSSION

The origin of lymphangiomas is not clearly understood. Most authors favor the theory that dysplastic lymphatic tissue is sequestered in a target tissue during the fetal development. 6 lymphatic sacs (2 jugulars, 2 iliac, 1 root of mesentery and 1 abdominal aorta) develop in the 8th week of gestation. Later, communications are established between lymphatics of various regions and with the venous system. It is suggested that the majority of lymphangiomas arise from parts of lymph sacs that fail to establish these connections.1 The disconnected lymphatic structures may dilate and evolve to a cystic formation. Lymphangiomas may also be acquired after surgery or trauma, 2infection, or chronic inflammation [1, 3, 4].

Lymphangiomas are divided into three categories: capillary, cavernous, and cystic type. Cystic types have the potential for extensive infiltration of surrounding tissues and lead to surgical difficulties. Therapeutically, they are classified into macrocystic, microcystic or mixed lymphangiomas [5].

Malignant transformation has never been reported. The mass evolves in three ways: spontaneous regression, slow progression, rapid enlargement. Infection within the cyst is usually caused by staphylococcus or streptococcus species. Nerve paralysis by compression could be secondary to the hemorrhage within the cyst. Lymphangioma can infiltrate and cause osteolysis of adjacent bones. An ulceration or rupture of the cyst has been reported [2, 5, 6].

Physical examination Nevertheless, imaging studies play an important role to confirm the diagnosis and to exclude other possible differentials of congenital lesions namely branchial cyst, thyroglossal cyst, teratoma and lipoma [7-9]. Computed tomography and magnetic resonance imaging play a significant role in surgical management of lymphangiomas. They help to delineate extension and anatomy study before operation to avoid iatrogenic injury to vital structures [7-9].

Because of its development between the facial nerve ramifications, the occurrence of a cystic lymphangioma in the parotid gland renders its surgical management more difficult. The ratio concerning the benefits versus the risks also renders decision making process more complex because one needs to confront radical tumour ablation to the presence of a benign tumour [10, 1].

Two fundamental principles are opposed in the management of intra-parotid lymphangioma: firstly, to have a radical approach when facing such a tumour with a high rate of recurrence; secondly, to preserve the function when preserving facial nerve function [11, 12].

Treatment is indicated in cases where potential complications are anticipated such as primarily airway compromise as well as aesthetic issue or the size of the tumour.

Surgery remains the mainstay of treatment. As lymphangiomas often infiltrate adjacent structures like

great vessels and nerves, complete removal is technically challenging. On the other hand, partial removal leads to recurrence. Postoperative complications occurred in 12–33% of cases and high recurrence rate of 15–53% did occur even in expert hands [13].

Sclerotherapy has been advocated but is less used because of its potential of scar retraction, which raised aesthetic concern and also making subsequent surgery more difficult [14, 15].

CONCLUSION

Even if lymphangiomas can easily be diagnosed from history, physical examination and imaging studies, its management is challenging and must always been done on an individual case basis, taking to account the potential risks on adjacent vital structures.

REFERENCES

1. Bailey CM. Cystic hygroma. Lancet. 1990;335 (8688):511–512.
2. Alqahtani A, Nguyen LT, Flageole H, Shaw K, Laberge JM. 25 years' experience with lymphangiomas in children. Journal of pediatric surgery. 1999 Jul 1;34(7):1164-8.
3. Oak SN, Redkar RG, Kapur VK. Posterior midline cervical fetal cystic hygroma. J Postgrad Med. 1992; 38:93–96.
4. Kumar N, Kohli M, Pandey S, Tulsi SP. Cystic hygroma. Natl J Maxillofac Surg.1 (1) (2010), pp. 81–85.
5. Ngui LX, Gopalan S, Chai CK. A rare case of spontaneous regression of huge neck lymphangioma post primary infection. Egyptian Journal of Ear, Nose, Throat and Allied Sciences. 2016 Jul 1;17(2):99-102.
6. Som PM, Lidov M, Lawson W. Hemorrhaged cystic hygroma and facial nerve paralysis: CT and MR findings. Journal of computer assisted tomography. 1990;14(4):668-71.
7. Perkins JA, Maniglia C, Magit A, Sidhu M, Manning SC, Chen EY. Clinical and radiographic findings in children with spontaneous lymphatic malformation regression. Otolaryngology—Head and Neck Surgery. 2008 Jun;138(6):772-7.
8. Chisin R, Fabian R, Weber AL, Ragozzino M, Brady TJ, Goodman M. MR imaging of a lymphangioma involving the masseter muscle. Journal of computer assisted tomography. 1988;12(4):690-2.
9. Liu DY, Guan J, He HX. Diagnosis and surgical treatment of cervical lymphangioma. Zhonghua Yi Xue Za Zhi., 89 (48) (2009), pp. 3413–3416
10. Berri T, Azizi S. Large cystic lymphangioma of the parotid gland in the adult. Egyptian Journal of Ear, Nose, Throat and Allied Sciences. 2014 Nov 1;15(3):259-61.
11. Oosthuizen JC, Burns P, Russell JD. Lymphatic malformations: a proposed management algorithm. International journal of pediatric otorhinolaryngology. 2010 Apr 1;74(4):398-403.
12. Zhou Q, Zheng JW, Mai HM, Luo QF, Fan XD, Su LX, Wang YA, Qin ZP. Treatment guidelines of lymphatic malformations of the head and neck. Oral oncology. 2011 Dec 1;47(12):1105-9.
13. Colbert SD, Seager L, Haider F, Evans BT, Anand R, Brennan PA. Lymphatic malformations of the head and neck—current concepts in management. British Journal of Oral and Maxillofacial Surgery. 2013 Mar 1;51(2):98-102.
14. Sichel JY, Udassin R, Gozal D, Koplewitz BZ, Dano I, Eliashar R. OK-432 therapy for cervical lymphangioma. The Laryngoscope. 2004 Oct;114(10):1805-9.
15. Ogita DS, Tsuto TO, Tokiwa K, Takahashi T. Intracystic injection of OK-432: a new sclerosing therapy for cystic hygroma in children. British journal of surgery. 1987 Aug;74(8):690-1.