Case Report

Parotid Lymphangioma of adult: A case report

K.C. Sarita, Rajeev Kumar Mahaseth, Manita Raut, Stuti Yadav, Digraj Yadav, Pooja Ghimire

Shree Birendra Hospital, Chaunni, Kathmandu, Nepal
Nepalese Army Institute of Health Sciences, Kathmandu, Nepal
Marajgunj Medical Campus, Kathmandu, Nepal

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ABSTRACT

Introduction: Lymphangioma is a malformation of the lymphatic system. It is a rare occurrence in adults. The exact etiology of the disease is unclear but it is expected to be either congenital or due to obstruction and retention of lymph in developing lymphatic vessels.

Case Description: We report a case of Parotid Lymphangioma in a 35-year-old male who presented to ENT OPD with painless right infraauricular swelling for 1 year which was insidious in onset and progressive in nature.

Investigation: FNAC of the right parotid lump, USG of the parotid and neck, CT scan, and MRI of the neck were done which demonstrated the lesion and helped in the diagnosis.

Treatment: The lesion was surgically excised under general anesthesia.

Outcome and Follow up: After the surgery, the patient was given IV antibiotics for 7 days and then discharged by prescribing Cefixime, Mupirocin, Pantop, Flexon, and eye drops. There was mouth deviation to the left side and incomplete closure of the right eye. However, the follow-up was uneventful with normal mouth and eye closure.

Conclusion: Parotid Lymphangioma must be diagnosed and treated as early as possible as it may cause complications. Regular follow-up even after the treatment is recommended.

1. Introduction

Lymphangioma is a benign, uncommon malformation of the lymphatic system usually seen in the pediatric population and rarely seen in adults [1]. It can be classified as superficial or deep based on the depth and size of the lesion or as congenital or acquired [2]. These lesions are most commonly seen in the cervicofacial region though they can be seen anywhere [3].

Clinical diagnosis is generally made by history and imaging examinations [2]. A confirmed diagnosis is ensured by histopathological findings. Resection of the lesion is the treatment of choice [2]. Here is a report on a case of Parotid Lymphangioma in a 35-year-old male who was treated by surgical excision. The objective of this study is to report and add to the literature about the adult case presentation of Parotid Lymphangioma. We particularly emphasize the investigation and management of this rare condition in absence of standard protocols. This case has been reported in line with SCARE’s guidelines [4].

2. Case description

A 35-year-old male, military by profession, presented to ENT OPD with painless right infraauricular swelling for 1 year which was insidious in onset and progressive in nature. It was not associated with pain, fever, night sweat, loss of appetite, or weight loss. There was no history of difficulty in swallowing, breathing, ear pain, or ear discharge. There was no change in the swelling upon deglutition, and on intake of food, had no pulsation or inflammatory change. The family history was unremarkable.

Clinical examination revealed a single swelling of 10cm × 6cm size was seen extending below the right tragus superiorly, at the level of thyroid notch inferiorly, anterior to mastoid posteriorly and 2 cm from the right tragus anteriorly. It was globular in shape, cystic in consistency, non-tender, fluctuant, and showed a negative slip sign.

3. Investigation

FNAC of the right parotid lump showed features of the benign non-
mucinous cystic lesion with no malignant cell. USG of the parotid and neck demonstrated multicystic and multiseptated lesions nearly replacing the right parotid gland. The CT scan revealed parotid glandular hypertrophy. MRI of the neck showed enlargement of the right parotid gland. It was $29 \times 47 \times 58$mm in size involving the superficial and deep lobes of the right parotid gland. Complete blood test including biochemistry was normal.

Cystic Lymphangioma, Warthin tumor, benign lymphoepithelial cyst, and branchial cyst were considered differential diagnoses in this patient.

4. Treatment

Total conservative parotidectomy was done under general anesthesia. Modified Blair’s incision was given, and a flap was raised anteriorly up to the anterior border of the parotid gland, posteriorly along the posterior margin of SCM muscle and the subplatysmal flap was raised inferiorly. The lymphangioma was removed along with both lobes. Excised mass was sent for histopathological examination. Dilated large lymphatic channels with disorganized smooth muscle in dilated channels were seen. The findings were consistent with Parotid Lymphangioma.

5. Outcome and follow up

Post operatively he had mouth deviation to the left side with incomplete right eye closure. He was given IV antibiotics for 7 days and was discharged 10 days after the admission. He was advised to undergo facial physiotherapy for two weeks. Cefixime 200mg per oral BD for 3 days, Mupirocin locally applied TDS for 2 days, Pantop (Pantoprazole) 40 mg per oral OD for 5 days, Flexon (Ibuprofen and Paracetamol) 1 tab per oral SOS for 10 days, and Refresh tears (Carboxymethylcellulose sodium) drops were prescribed.

6. Discussion

Lymphangioma is a rare congenital anomaly of the lymphatic system mainly seen in children and atypically in adults [5]. The incidence among children is 1.2–2.8 per 100,000 [6]. Both sexes are equally affected. It is categorized into three types: capillary, cavernous, and cystic [7]. The accurate etiology of the disease is unclear but it is supposed to be either congenital or due to obstruction and retention of lymph in developing lymphatic vessels [8,9].

It commonly occurs in the head and neck region but can occur on any site like the axilla, shoulder, chest, mediastinum, and thigh [7]. In our case, the lymphangioma was seen in the right intraauricular area. Typically patients present with painless, soft, cystic swelling that grows with time [7]. Our patient presented with similar symptoms and the swelling grew from the size of a pea to the present size. The literature suggests that the parotid gland will show positive Fluctuation and transillumination test but in case, the transillumination was negative. The swelling in our patient was mobile, fluctuant, compressible but non-reducible, and had a smooth surface with an irregular margin.

Diagnosis is done by history and imaging examinations like USG, CT, and MRI. FNAC is also used for the investigation. USG of Lymphangioma
shows multicystic mass which is hypoechoic within thin walls [10]. A thin-walled, multicystic, homogenous mass with smooth septa can be appreciated on the CT scan of parotid lymphangioma [11]. MRI is considered the choice of investigation. MRI of lymphangioma displays hypointense T1 weighted images in hyperintensity and T2-weighted images with the thin-walled multicystic lesion [11]. We first did FNAC and USG followed by a CT scan and MRI. Definitive diagnosis was made by histopathological examination of excised mass. Along with normal salivary glands separated by connective tissue septa, the section showed, dilated lymphatic channels with focally disorganized smooth muscle in dilated channels. No atypia or malignancy seen.

Surgical management is the treatment of choice that includes superficial or total parotidectomy, and enucleation [11]. In our case, we opted for Total conservative parotidectomy. The lymphangioma was removed along with both lobes. We preserved the temporal, zygomatic, marginal mandibular and cervical branches of the facial nerve. In parotidectomy, the injury to the facial nerve and particularly to the marginal mandibular is quite common [3]. In this case, we sacrificed the buccal branch of the facial nerve and the greater auricular nerve. The wound was closed in two layers.

Infection, bleeding, hematoma, injury to the facial, hypoglossal, glossopharyngeal, recurrent laryngeal and lingual nerves are some of the reported complications of resection [5]. The rate of recurrence of the surgery is seemingly high, ranging from 10% to 38% [12].

Radiation is not used anymore due to its inefficiency [13]. It is avoided due to its tendency to transform the lesion into malignant [14]. Aspiration can be done for decompression temporarily but recurrence is quite obvious in these cases [11]. When surgery is difficult, percutaneous sclerotherapy is considered the choice of treatment. It avoids damage to the facial nerve. Sclerosing agents like bleomycin, OK-432, triamcinolone, alcohol, and fibrin sealant are used [3].

7. Conclusion

Parotid Lymphangioma, though a rare condition in adults, should be suspected based on history and findings of radiological investigations. The definitive diagnosis in our case was done by histopathological examination after surgical resection of the lesion. Since recurrence after surgery has been reported, regular follow-up is of utmost importance.

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