Case Report

Benign Cervical Teratoma in an Adult: A Rare Neck Swelling

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Received: 11/11/2016 Revised: 28/11/2016 Accepted: 30/11/2016

ABSTRACT

Cervical teratomas are extremely rare germ cell tumors that occur in the neck. Most of these tumors occur in infants and neonates and are almost always benign. In extremely rare cases, cervical teratomas occur in adults and are usually malignant. Cervical teratomas are differentiated into primary thyroid or extrathyroid based on the relationship of the tumor with the thyroid gland. Surgery is the primary modality of treatment as malignant transformation occurs during adulthood. The diagnosis of malignancy is based on histopathologic examination. Adjuvant chemotherapy is indicated when malignancy is confirmed. We report a case of benign cervical extrathyroid teratoma in an adult.

Keywords: Teratoma, Neck, Adult.

INTRODUCTION

Cervical teratomas are uncommon pathological entities that are usually diagnosed at birth and rarely reported in older children and adults. We report a mature cervical teratoma in an adult and offer a brief review of the literature.

CASE REPORT

History: A 24-year-old male patient presented with swelling over anterior aspect of neck since 4 months, which progressively increased in size. There was no history of fever, cold, cough, pain, and trauma, hoarseness of voice, dysphagia, dyspnea, stridor, tremors, hot/cold intolerance, sweating, and palpitations, weight gain/loss.

On Local examination: a well-defined swelling of size 3x4cm was noted on left side of anterior aspect of neck near suprasternal region. It was soft to firm, non-tender and moved with deglutition. Skin over the swelling was normal. There was no e/o sinus, ulceration, pus or bloody discharge. There were no palpable cervical lymph nodes. Systemic examination was unremarkable.

A clinical diagnosis of benign cystic swelling of neck suggestive of branchial cyst was given.

Radiological evaluation: Ultrasonography of neck (USG) revealed walled, anechoic midline cystic lesion of size 45 x 36.9 mm near suprasternal region.

Bilateral thyroid gland appeared to be normal in size shape and echogenicity. Bilateral submandibular glands, parotid glands, carotid arteries appeared normal. (Fig.1)

USG was suggestive of
? thyroglossal cyst (infrahyoid)
? Suprasternal extension of left sided colloid cyst.
Bilateral thyroid gland appeared to be normal in size shape and echogenicity. Bilateral submandibular glands, parotid glands, carotid arteries appeared normal. Routine laboratory tests including CBC, liver function test, kidney function test, lipid profile, and fasting and postprandial blood sugar were within normal limit. Serological examination for HIV and HBsAg was negative and Thyroid function test was within normal limits.

Fine needle aspiration cytology (FNAC) of the swelling was performed. Cytological examination showed plenty of anucleated and nucleated squamous epithelial cells, plenty of polymorphs and cyst macrophages on a background with granular eosinophilic material. Thyroid follicular epithelial cells were not seen in the smear.

A diagnosis suggestive of brachial cyst was given on FNAC.

Fig.1: Ultrasonography of neck (USG) revealed walled, anechoic midline cystic lesion of size 45 x 36.9 mm near suprasternal region.

Fig.2: On gross examination, the excised mass was yellowish white, 3x4cm in size soft and cystic. At places, it was firm and congested. On cut section serous pale brownish fluid oozed out. At one place the cyst wall was thickened and firm in consistency.
**Treatment:** A neck collar skin crease incision with elevation of subplatysmal flaps followed by retraction of the strap muscles laterally, revealed a 3x4cm, yellowish white cystic mass, located just below the thyroid gland in the suprasternal region, separated from the thyroid gland and surrounding tissue. Mass was removed in too. Hemostasis was achieved and incision wound was sutured in layers. Post-operatively, the patient was stable with no hematoma or wound infection. He was discharged, the next day, without any complications and uneventful recovery was followed.  

On gross examination, the excised mass was yellowish white, 3x4cm in size, soft and cystic. At places, it was firm and congested. On cut section serous pale brownish fluid oozed out. At one place the cyst wall was thickened and firm in consistency. (Fig.2)  

![Fig.2: Histologic findings of the benign cervical teratoma](image1)

Histological examination revealed stratified squamous epithelium and pilosebaceous structures, underneath fibrocollagenous and mature adipose tissue, ciliated pseudostratified columnar respiratory epithelium, intestinal columnar epithelium with goblet cells, mature cartilage, lymphoid tissue with prominent lymphoid follicles (Fig.3, 4, 5) 

Since there was no infiltration into surrounding tissues, no primitive neuroectodermal, no thyroid tissue and fetal
tissue was seen microscopically, a diagnosis of benign cervical teratoma was given.

Fig.4-Histologic findings of the benign cervical teratoma- [A] & [B] mature cartilage & mature adipose, [C]&[D] intestinal columnar epithelium with goblet cells,
DISCUSSION

Teratomas are embryonal tumours arising from the abnormal development of pluripotent germ cells, comprising of tissues derived from all the three embryonic germ layers i.e. ectoderm, endoderm and mesoderm. Teratomas are usually diagnosed in unborn babies and infants, but rarely occur in adults. [1] This tumor normally arises inside the gonad, but it can also occur outside the gonad. About 40% of extragonadal teratomas originate from the sacrococcygeal region, followed by (not in order of incidence) the brain, spinal cord, globe, pharynx, mediastinum, and retroperitoneum and liver. [1,2] Head and neck teratomas account for 3% to 6% of all teratomas. [1]

Cervical teratoma incorporates lesions arising in the anterior and posterior triangles of the neck. [2] It comprises only about 3% of all teratomas, [2] with the majority occurring in neonates or infants. They predominate in females (3/4 of the cases). [3]

The reported incidence of cervical teratoma in newborn, children aged 1 month to 18 years and adults were 75.1%, 14.3% and 10.6% respectively. [2] The clinicopathologic behavior of the cervical teratoma in adults is distinctly different from those of the neonate and infant. In contrast to congenital cervical teratomas, which are almost always benign, those in adults are often highly malignant with a tendency to metastasize and have poor prognosis. [2]

The first proved case of teratoma of the neck was published by Hess in 1854, and was restudied microscopically by Wetzel in 1895. [4]

Some researchers have made an attempt to classify these tumors on the basis of their relationship to the thyroid gland. It is still debated and stirs controversy among researchers whether teratomas of the thyroid gland and teratoma of a neck origin are distinctively different entities. [2] However, there is no clinical significance, as these tumors have no difference in terms of prognosis or treatment. [2] Therefore, many researchers have abandoned separating these tumors and classify all neck teratomas as cervical teratomas.

Cervical teratomas are divided into thyroid teratomas and extrathyroid teratomas depending on where they originated. [1] Review of the literature shows that, in general, tumors of the cervical region have been regarded as thyroid teratomas if one or more of these features were present: 1) the tumor occupies a portion of the thyroid gland; 2) there is direct continuity between the tumor and the thyroid gland; and 3) a cervical teratoma is accompanied by total absence of the thyroid gland. [5] The latter cases have been explained by hypothesizing that there might
have been total replacement of the gland by the tumor or, alternatively, that the teratoma had arisen from a thyroid anlage that failed to develop into a mature thyroid gland. [5]

The symptoms associated with cervical teratomas age and size of the tumour. In children, Small tumors may not cause any symptoms (asymptomatic). However, a large teratoma may cause disfigurement and compress nearby structures such as esophagus and trachea and may cause dysphagia, wheezing or stridor, dyspnoea or shortness of breath or life threatening respiratory distress. Hydramnios is commonly found associated with this abnormality.

Cervical teratomas in adults are extremely rare. In most cases, they are malignant and may metastasize to nearby lymph nodes and other organs of the body, especially the lungs. As in children, cervical teratomas in adults can compress nearby structures like trachea and esophagus resulting in respiratory distress and dysphagia.

The diagnosis of cervical teratoma is based on clinical evaluation, and a variety of specialized tests. Such testing includes fine needle aspiration cytology and histopathologic examination. Radiologically; USG, CT scan and MRI may support the diagnosis. Ultrasound neck is the first line modality for evaluating cervical neck abnormalities, especially for assessment of concomitant thyroid pathology. However, it did not identify the tumor as in our case as the echogenic profile mimicked the thyroid gland. Computed tomography (CT) of the neck would have been more accurate but we were misled to believe we were dealing with a simple thyroglossal cyst or suprasternal extension of a colloid cyst. CT appearance will depend on the type of teratomas, with a variable attenuation, consistent of different tissues: fat/ fluid levels with areas of calcification. [2]

On, MRI and fine needle aspiration, mature teratomas are usually cystic in comparison to immature teratomas, which are usually solid. [2]

Teratomas have diverse histopathologic differentiations due to a wide range of cellular differentiation and variable degrees of maturation. [2] However, histologic immaturity should not be equated to malignancy. [2] In general, they can be categorized into three types: mature teratomas, immature teratomas and teratomas with malignant transformation. [2]

Mature teratomas, are composed of fully differentiated tissue elements of all 3 germ layers although the presence of only 2 germinal components does not exclude this diagnosis. [2] Derivatives of 1 or 2 germ layers may overgrow others. Immature teratomas contain the primitive tissues found in fetus (usually immature neuroectodermal tissues arranged in primitive neuroepithelial rosettes and tubules) Teratomas with malignant transformation occur in 30% of cases and are usually adenocarcinomas occurring in mature teratomas; or angiosarcomas or rhabdomyosarcomas occurring in immature teratomas. [2]

Teratomas have an unpredictable biologic potential and often undergo malignant transformation during adulthood. [2] Therefore, complete surgical excision is recommended for cervical teratomas found in adults. If histopathological examination shows the tumour to be a malignant cervical teratoma, then complete resection is followed by adjuvant chemotherapy.

The long dormant period from inception to diagnosis for cervical teratomas in adults is uncommon. Generally in males, anterior neck swellings are only noticed once they have achieved a significant size as the well developed muscles in males mask smaller swellings. The tumour itself is slow growing by nature and has been undetected all this while. Clinical examination grossly underestimated the size of the lesion and most of the tumour was located retrosternally pushed down by the muscles.

In approximately 50 percent of cases, the appearance of clusters of calcium (calcifications) can be detected on x-rays, a finding suggestive of a teratoma. Laboratory
tests and specialized imaging tests may also be conducted to determine possible infiltration of regional lymph nodes and the presence of distant metastases.\[6\]

The differential diagnosis is done with a metastasis from thyroid carcinoma, cystic squamous cell carcinoma of cervical lymph node arising in the oro/nasopharynx, follicular adenomas of the thyroid, lymphangiomas, and bronchial cysts.\[5\]

**CONCLUSION**

Our patient was unique clinically and ultrasonographically that the tumor strongly mimicked a thyroglossal cyst and intraoperatively there was a midline cystic mass just below thyroid gland and separated from it. As there was no infiltration into surrounding tissues, or metastasis to lymph nodes, the possibility of a malignant tumor was ruled out.

In summary, benign cervical teratoma of adult is extremely rare. At the clinical level, entities to be considered in the differential diagnosis, thyroid carcinoma, metastasis of cystic squamous cell carcinoma to cervical lymph node arising in the oro/nasopharynx, follicular adenomas of the thyroid, lymphangiomas, thyroglossal ductal cyst, and branchial cleft cyst, all of which should be easily recognizable at the microscopic level. The diagnosis is based on histopathologic examination. Preoperative investigation with CT neck requires high index of suspicion. Due to concerns of malignant transformation during adulthood, complete surgical resection is recommended. Adjuvant chemotherapy is indicated when malignancy is confirmed histologically.

**REFERENCES**


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International Journal of Health Sciences & Research (www.ijhsr.org) Vol.6; Issue: 12; December 2016