Low Grade Intraductal Carcinoma

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Abstract

Low-grade Intraductal carcinoma (LG-IC) is rare, malignant salivary gland tumor present most commonly in the parotid gland (80%), affecting female more than male (2:1).

Histologically, LG-ICs well-circumscribed, non-encapsulated and cystic lesions ranged from 7 mm to 53 mm, containing serous to hemorrhagic fluid [2].

Here we report a case of LG-IC arise in the left parotid gland of 47 female patient, the lesion was first diagnosed according to general pathologist as benign lymphoepithelial cyst (BLEC) and a superficial parotidectomy is done, due to many concern from the general pathologist regarding the final diagnosis, the lesion again reviewed by two oral pathologists and the diagnosis confirmed as Low-grade Intraductal carcinoma (LG-IC).

Keywords: Low-grade Intraductal carcinoma (LG-IC); Parotid Gland

Introduction

Low-grade Intraductal carcinoma (LG-IC) is rare, malignant salivary gland tumor present most commonly in the parotid gland (80%), affecting female more than male (2:1), the previous name of LG-IC was (Low-grade cribriform cystadenocarcinoma), and it was reclassified according to the 4th WHO Classification of Head and Neck Tumors in 2017 [1]. In 1996, Delgado., et al. defined Low-grade Intraductal carcinoma (LG-IC) as a variant of salivary duct carcinoma. (LG-IC) generally affecting female 2:1 predominance in elderly individuals. Parotid gland is the most prevalent involvement location that may happen but rarely in the palate, submandibular gland, intraparotid lymph node, and accessory parotid gland. (LG-IC) is distinguished by a papillary-cystic or cribriform pattern of proliferation and is similar to low-grade in situ or atypical breast ductal hyperplasia in histology and biological characteristics. Initially, (LG-IC) was referred to as low-grade salivary duct carcinoma (LGSDC) to differentiate it from standard salivary duct carcinoma (SDC) [2].

Histologically, LG-ICs well-circumscribed, non-encapsulated and cystic lesions ranged from 7 mm to 53 mm, containing serous to hemorrhagic fluid, exhibited multiple cysts (92%) and a typical Intraductal growth with three possible architectural patterns [3]:

1. Cystically dilated ducts with tufted, micropapillary anastomosing proliferations.
2. Distended ducts with solid “pseudocribriform” fenestrations or solid papillary proliferations.
3. Intraductal proliferations with Roman Bridges akin to the cribriform architectural pattern typical of low-grade intraductal breast carcinoma.

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Case Description

A 47-year old Filipino women with past medical history of hypertension presented to Maxillofacial surgery department complaining of painless swelling on left pre-auricular region since 2 years. on examination, there was 4x4 mass on left pre-auricular region well-palpable under the face skin, the neck examination revealed no any lymphadenopathy on the neck from level I-VI, MRI were requested and show CT scan and MRI for more details, CT reported, there is a well circumscribed encapsulated cystic lesion noted involving the posterior aspect of the superficial lobe of the parotid gland as well as the deep lobe and has thin wall enhancement with thin septa, it measures about 2.5 x 2.7 cm with no evidence of mural soft tissues nodule, differential diagnosis would include warthin’s tumor versus first branchial cleft cyst. Homogenous enhancement of the right parotid gland with no similar lesion seen. Symmetrical nasopharynx, mild inflammatory changes of paranasal sinuses. Normal thyroid gland with no evidence of significant cervical lymphadenopathy.

MRI reported, there is large, well-defined, sharply demarcated, lobulated cystic mass lesion seen occupying most of the posterior part of the superficial lobe as well as the deep lobe of the left parotid gland, measuring about 4 x 4.6 x 3.8 cm in maximum AP, transverse and CCE dimension respectively, showing a hyperintense in both T1 and T2-weighted images with small posterior fluid-fluid level, suggesting high proteinaceous content versus intracystic hemorrhage. The postcontrast subtraction images show a thin peripheral enhancement with no irregularity is noted. No size significant enlarge cervical lymph nodes are noted. Left parotid airspace and parotid gland, bilateral submandibular and sublingual salivary glands are unremarkable. There are changes of paranasal sinusitis, mainly in bilateral ethmoidal air cells.

Surgical procedure

The patient was brought to the holding area where the area was greeted by the surgical, anesthesia and nursing teams. The patient's history was updated. The site was marked and patient was then brought to the Operating Room. She was transferred to the operating table and all appropriate leads and monitors were placed by the anesthesia team. All pressure points were padded. The patient was then induced by the anesthesia team. both eyes were protected with half cut tegaderms. The patient was then intubated orally and the tube was secured. Patient was then prepped and draped in standard fashion. A timeout was performed verifying patient name, medical record number, preoperative diagnosis, planned procedure, sidedness, preop medications, allergies.

Started with marking for modified Blair incision in left parotid area with posterior extension and inferomedial extension in neck skin crease below the inferior border of the mandible by 2cm. Next approximately a total of 3 mL of 2% Xylocaine with 1: 100,000 of epinephrine were infiltrated into dermal layer of the surgical site. A 15 blade was used to make an incision to the level of subcutaneous tissue. Then skin flap was elevated in just superior to periparotid fascia and the flap was elevated anteriorly until the leading edge was anterior to the parotid mass. Then 2-0 silk "stay sutures" was placed into ear lobe for posterior retraction and double stay sutures into the cheek flap for anterior retraction.

Attention was then turned on identifying the main trunk of facial nerve where was identified the cervicofacial and temporofacial branches. Then the tumor was dissected using blunt safe dissection posterior to anterior fashion in a plane just superior to the course of facial nerve branches. Specimen was totally out, The mass was then oriented and sent to pathology. The facial nerve was stimulated and caused contraction of all facial muscles supplied by the nerve. Bleeding was stopped with bipolar cautery and the wound was irrigated with sterile saline. flap was raised from SCM muscle and secured on top of facial nerve branches for isolation and protection. JP drain
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was used and secured to skin with silk suture. The wound closed in layers using 3-0 Vicryl sutures to reapproximate the capsule, fascia, and subcutaneous tissue. 5-0 praline stitches to reapproximate the skin. The patient tolerated the procedure well, was extubated in the operating room and transferred uneventfully to the recovery unit.

Surgical tissue block consists of; Lymph node parotid: It consists of one lymph node measuring 0.7 x 0.4 x 0.2 cm, soft and grayish tan in color. ALL/1 block, left superficial parotid: It consists of part of parotid gland measuring 6.5 x 5.5 x 1 cm. The outer surface is rough. Cut sections show a cyst occupying most of the lobe with thick, firm, grayish white wall and smooth lining. RSS/6, deep tissue from left parotid gland: It consists of one piece of fibrofatty tissue measuring 1 x 1 x 0.2 cm, soft and grayish tan in color. ALL/1 block.

Figure 2
Macroscopic and histopathological findings

Histopathological examination showed well-circumscribed cystic neoplasm of parotid gland. The cyst is lined by epithelial neoplastic cells with apocrine morphology. There is no true invasive growth, nor in the gland neither in fibro-fatty tissue. No Lymphovascular invasion. The intraluminal pseudopapillae are apocrine but negative for AR.

Luminal epithelial cells are slightly polymorphic/apparently malignant, decorated with S100 protein, GATA3, SOX10 and mammaglobin (the immunoprofile shared by secretory carcinoma (MASC) and intraductal carcinoma (IC); they are negative for p63 and p40.

Figure 3A and 3B: Case 1 demarcated the tumor with a relative border from the adjacent mildly lipomatous parotid glands.

Figure 3C and 3D: C: Without important cytological and nuclear types, the tumor cells were uniform. They exhibited fine chromatin and pale to amphophilic cytoplasm round to oval nuclei. D: Focally observed apocrine differentiation of tumor cells.

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Discussion

Like other rare tumors in parotid glands, (LG-IC) is a low-grade malignant tumor with indolent clinical conduct. It is mentioned in a distinct heading in the third WHO classification, although as a variant of cystadenocarcinoma, 35 instances of (LG-IC) have been recorded to date in nine articles. Therefore, all of the reported (LG-IC) happened in big salivary glands except for one contentious hard palate report [5,6]. The size of the tumor ranged from 0.9 to 4 cm. No one had recurrence or died due to the tumor from the instances with accessible follow-up. Most instances were handled with parotidectomy without radiotherapy, according to the clinically indolent conduct.

Grossly, according to the morphological characteristic, that is, a slowly increasing cystic mass, the clinical proposal of (LG-IC) is used to be Warthin tumor [7]. Histologically, the tumor is unencapsulated, consisting of single or multiple cysts with proliferation intraductal. The cytologically shallow ductal cells are consistent with the cystic cavity [8]. The intraductal proliferation has a cribriform pattern comparable to breast proliferations with "sieve-like spaces." Most tumors are intraductal however, there may be tiny regions of invasion.

(LG-IC) differential diagnosis involves PCV-ACC, standard SDC, cystadenocarcinoma, PLGA, ex pleomorphic adenoma carcinoma, and MASC. PCV-ACC is similar to (LG-IC) by PAS-positive/diastase-resistant granules (zymogen) and hemosiderin intracytoplasmic. Unlike (LG-IC), PCV-ACC generally shows the more prevalent microcystic development pattern adjacent to cystopapillary fields and displays basophilic granular cytoplasm [9]. PCV-ACC has no intraductal component predominance in histology and the intracytoplasmic vacuoles are consistent in size PCV-ACC is predominantly negative (about 90 percent) for S100 compared to (LG-IC) and focally expressed when present. Furthermore, PCV-ACC happens primarily in youth, whereas (LG-IC) generally impacts elderly individuals [10].

In the present WHO classification, according to the histological characteristics, (LG-IC) is considered as a variant of cystadenocarcinoma. Conventional cystadenocarcinoma, however, appears to be an invasive tumor and lacks general similarity to proliferation of the intraductal breast [11]. Also, consideration should be given to PLGA and carcinoma ex pleomorphic adenoma when diagnosing (LG-IC). The unique neurotropism and lobular, trabecular and tubular patterns of PLGA can be differentiated from (LG-IC). The shift from myoepithelium to stromal spindle cell and myxoid or cartilaginous stroma are the typical characteristics of ex pleomorphic adenoma carcinoma that are not observed in (LG-IC) [12].

The instance showed the typical characteristics of (LG-IC), i.e. intraductal proliferation and bland ductal cells organized in a cribriform pattern resembling low-grade breast lesions. In these instances, the majority of tumor nests or cribriform structures were rimming with the constant myoepithelium confirmed by p63, thus clarifying the neoplasm's in situ nature. The tumor cells were diffusely positive for S100 in both cases. Furthermore, in our instances, the Ki67 index was below 5%. All of these results endorsed (LG-IC) diagnosis [13,14].

Conclusion

In conclusion, our study demonstrated a rare salivary tumor instance of (LG-IC). In the differential diagnosis of salivary tumors, (LG-IC) should be regarded to prevent making a misdiagnosis of other extremely aggressive salivary tumor.

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Bibliography


