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INTERNATIONAL JOURNAL OF ADVANCED RESEARCH (IJAR)

Article DOI: 10.21474/IJAR01/14629
DOI URL: http://dx.doi.org/10.21474/IJAR01/14629

RESEARCH ARTICLE

PRIMARYOVARIANCARCINOID : A CASE REPORT AND REVIEW OF LITERATURE

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Manuscript Info

Manuscript History
Received: 28 February 2022
Final Accepted: 30 March 2022
Published: April 2022

Abstract

Carcinoidtumors are rare, slow-growingneoplasmsthat arise from the neuroendocrine cells. However, carcinoidtumors of the ovaries are uncommon ,especiallyprimaryovariancarcinoidtumors, whichformapproximately 0.3% of all carcinoidtumors . Wepresenthere a rare case of a 51 yearsoldpatientwhoinitiallywaspresented for an ovarianteratoma and whoseanatomo-pathologicalstudyshowedthatwasa rare case of primary mixed insular and trabecularcarcinoidtumor of the ovary. The complement of the therapeutic management in this case wasdoneafteramultidisciplinary consultation meeting.

Introduction:-

Carcinoidtumors are rare, slow-growingneoplasmsthat arise from the neuroendocrine cells [1]. The appendixis the mostcommon site for carcinoidtumors, followed by the rectum, ileum, lungs, bronchi, and stomach [1]. However, carcinoidtumors of the ovaries are uncommon [2], especiallyprimaryovariancarcinoidtumors [3], whichformapproximately 0.3% of all carcinoidtumors [2]. Wepresenthere a rare case of primary mixed insular and trabecularcarcinoidtumor of the ovary

Case Report:

51 yearoldfemale patient withmedicalhistory of type 2 diabetes on oral anti diabetic and hypertension underamlodipine, married, nulligest, and menopause for 1 year, whopresentedinitiallywithchronicpelvic pain evolving for 4 months,,withoutanyotherassociatedsigns, namely no diarrhea or vomiting.

The patient consulted a gynecologist in July 2020 the pelvicultrasoundshowedlimited right laterouterine formation of 45 * 48 mm, withfinelyechogenic content, site of a macro calcification, of ovarianappearance. The patientwassubsequentlylostsight, to consultagain in December 2020 for the persistence of the samesymptoms. A pelvicultrasoundwasredonewhichshowed a right laterouterine mass withafatty, tissue and calcium component infavor of a dermoidcyst of 55 * 47 mm. The result of the CA125 assay came back negative at 4.16UI/mL. The patient thereforeunderwentsurgery in December 2020 for a laparoscopiccystectomy . An ovalcystic formation weighing 60g and measuring 6cm in large diameter. Its surface wasregularsmoothwithoutexocysticvegetationswasremoved and sent to pathologywhichfound the microscopicappearance of a malignanttumorproliferation on mature multi-tissue teratoma. An immunohistochemicalstudywasdone, and showed a mature teratomaassociatedwith a carcinoid, based on the dosage of anti-Pan-cytokeratinantibodies, anti-chromograninantibodies, anti-synatophysinantibodiesrevealing a positivity of the tumorcells, but also anti-Ki67 antibodieswhoselabeling has been estimated at <1% of tumorcells.

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The slides wereread by twodifferentpathologists, bothfinding the sameresults. a thoraco-abdomino-pelvic scanwasdone as part of the extension assessment, returnedwithout anomalies.

The patient's case wasthendiscussed in the multidisciplinary consultation meeting, and the decisionwas to do enlargedadenoidscolpohysterectomy, appendectomy and omentectomy.

The post op Pathology report didn’tshowanyabnormalities.

The patient did not receive adjuvant chemotherapywitha good clinical course

Discussion:-

Primaryovariancarcinoidtumors are rare; and representapproximately 0.3% of all carcinoidtumors [4]. The age of presentation of ovariancarcinoidtumors has a large variation, rangingfrom 14 to 83 yearswith a peak in the perimenopausal or post-menopausalperiod[5,6]. In our case, the patient was 55 Yearsold.

This is due to the factthatvenous drainage from the ovarybypasses the liver and reachesdirectlyinto the systemic circulation. However, the other types of carcinoidtumor of the ovary are generally not associatedwithcarcinoid syndrome [2, 6].

In our case, the patient did not have anysignsand symptoms of carcinoid syndrome/carcinoidheartdisease. Sincediagnosis of carcinoidtumorwas not suspectedclinically, the patient was not investigatedspecifically for it.

The Clinicalsymptoms of POC are usually not specific, abdominal pain, vaginal bleeding, and dysmenorrhea are occasionallyreported.[7]Besides, rare symptomslikeheartdisease, diarrhea, constipation, hypoglycemia, and hirsutism have been reported in some cases.[8]

In ourcase,our patient presented none of the symptomsmentionedaboveapartfrom an isolatedchronicpelvic pain.

Primarycarcinoidtumorishistologicallyclassifiedintoinsular, trabecular, mucinous and mixed types [5, 6]. The majority of the primaryovariancarcinoidoccurs in association withsolidteratoma or mucinoustumors or sertoli-leydigcelltumors; however, pure forms have also been documented [5, 6]. The pure forms are thought to developeither as one-sideddevelopment of a teratoma or from the enterochromaYncellspresent in the ovary [6]. The mostcommonhistological type isinsular, followed by trabecular and mucinous [6]. Grossly, pure primarycarcinoidtumors are almostalwaysunilateral [6, 9]with tan yellow and solidcut surface with few cystic areas and mayvary in size frommicroscopicfocito almost 30 cm in the longest dimension.

In our case, itwas a unilateraltumor, whichwas a yellowishsolid on a cut section with few cystic areas.

Well-differentiated neuroendocrine tumors in the ovary are histologicallysimilar to carcinoid/atypicalcarcinoidtumors in the otherorgans. However, in the ovaries, carcinoids and atypicalcarcinoids are oftenpresentas a part of the components of a teratoma or ‘‘specializedteratoma.’’ In some cases, the neuroendocrine elementmaybe the onlytumor component in the ovary. somestudies show thatapproximately 14 % of ovariancarcinoidtumorswereincidentallyidentifiedfrompathologicexamination of excisedovaries for otherreasons [3]. However, Primaryovariancarcinoidtumorsmaybesubdividedinto four categories: insular, trabecular, mucinous, and strumal (trabecularcarcinoid mixed withthyroid tissue) [5]

In the literature, the insular pattern isreported to be the mostcommonsubtype, and one-third of patients withinsularcarcinoid have presentedwithcarcinoid syndrome beforetreatment [10]. It wassuggestedthat the clinicalsymptoms of carcinoidwererelated to the size of the tumor [11]. The prognoses of insular, trabecular, and strumalcarcinoid are favorable becausethesetumors in the ovary are rarelyassociatedwithmetastasis [12], althoughadhesion to the omentum and to the intestine mayoccurwhenthesetumorsextend to the serosa.

However, there are some rare case reports thatdescribeametastaticcarcinoidfromovarianprimarytumor [14]. In otherorgans, atypicalcarcinoidisdifferentiatedfromcarcinoidwithaslightlyincrease in mitosis but a lack of significantnuclearpleomorphism and tumornecrosis.

IHC analysis,[14,15] such as Syn, CgA, CD56, PYY, and thyroglobulin, couldalsopromote the diagnosis of POC, while in our case, the tumor tissues were positive forSyn, CgA, and anti Pan-cytokeratin. The specificity of CgA and 5-HIAA was 86% and 35%, respectively.[15] Besides, therewasstudyreportingthat (thereis a direct correlationbetweenCgA and the diseaseseverity) the sensitivity of CgAwasassociatedwith the diseaseseverity.[16] Recently, Zhang et al[17] reportedthattherewas a close correlationbetween Ki67 index and patient survival time, and a higher Ki67 index in metastaticcarcinoidindicated a worseprognosiswhencomparedwith POC. In our case, the Ki67 index was positive and estimated<1%, and the metastaticassessmentwasnegative.

The treatmentstrategy of POC shouldtakeintoconsiderationhistology type, patient age, and fertilityneeds.[18] For youngfemales in early stage (stage I) whohave pregnancydesires, fertilitysparingsurgeryshouldbepreferred. For patients in late stage (stage II to IV), comprehensivestaging and cytoreductivesurgeryisrecommended. For patients withmucinous type of POC, omentectomy and para-aorticlymphadenectomymaybenecessary. For insular and trabecular types, TAH+BSO shouldbeselected. In addition, the recurrent and metastaticdiseases are usuallymanaged by a secondarysurgicalresection, chemotherapy, and moleculartherapy. Chemotherapycouldbeused in the later stages OC.[17] our case studywasclassified as stage I, withoutchemotherapyconductedpostoperatively . Satisfactoryresultswereachieved in our patient duringfollow-up visit. Whether radiation therapy, hormonal therapy, and moleculartherapy are useful or not has not been validated. Recently, somemolecularmedicationswerereported to help survival in gastrointestinal and pancreaticcarcinoids, and isalsoproved to beuseful in Ovarien carcinoid.[17,19]

Authorsreportedthat the prognosiswas good in insular, trabecular, and stromalcarcinoids of POC whencomparedwithmucinous or undifferentiated type.[11,20] In our case, our patient wasinsular type and exhibited good prognosis, up to now, sheachievedsatisfactorytherapeuticeffectswithoutdeterioration of general condition.

Conclusion:-

Primaryovariancarcinoidis a rare tumor of the ovaries [2] and isassociatedwith good clinicaloutcome [6]. Thesetumorsmetastasizeoccasionally [5, 6] and shouldbetreated as ovariantumors of lowmalignantpotential [5, 21].

The diagnosis of thistumoris important boththerapeutically and prognostically a simple surgical excision isusuallyassociatedwith a good prognosis in these patients [21] and itspares the patient from the morbidity and cost of pre and postoperativechemotherapy, whichisusually the treatmentprotocol for mostmalignantovariantumors.

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