IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 16, Issue 5 Ver. IX (May. 2017), PP 90-93
www.iosrjournals.org

Primary Anal Canal Mucinous Adenocarcinoma: About Two Cases And A Review of The Literature

Ghalleb M ; Jaidane O; Boujelbene N[2];Rezguani M ; Chemlali M[1] Benzarti Z[1];Dhieb T[1]; Mrad K[2];Rahal K[1]

[1]*Surgical Oncology Department, Institute Salah Azeiez Of Oncology Tunis*
[2]*Anatomopathology Department, Institute Salah Azeiez Of Oncology Tunis*

Summary:
Introduction: *Anal tumors are rare neoplasms of the digestive tube that represent 1,5% of gastrointestinal tumors. They present a more aggressive natural history, with shorter survival times and higher rates of local as well as distant relapse. It is difficult to distinguish from adenocarcinoma of the lower rectum unvading the anal canal.*
Case Report: *The two patients were female with a long medical history of proctologic disease such as anal fistula, anal prolapse, hemorrhoids and bleeding.*
Patient one: a 71 year old woman. She presented with, bulging mass in the anal canal. The biopsies concluded to a medially differentiated adenocarcinoma. The patient did not show up for a year due to personal reasons. The patient got a flash radiation followed by Abdomino perineal resection (APR).
The anatomopathology concluded to a mucinous adenocarcinoma. The patient died within a month after surgery due to a deteriorated heart condition.
Patient2: A 62 year old female patient. She presented in april 2007 with a 4 cm mobile, bulging mass mass in the anal canal with 3 anal fistula. The patient had neoadjuvant chemoradiation followed by an APR. The hitology concluded to a mucinous adenocarcinoma of the anal glands.
The follow up of the patient until 08/09/2015 showed no sign of recurrence.
Conclusion: *The diagnosis is often late, in an advanced stage of the sickness. Recurrent or non recurrent fistula-in-ano requires multiple biopsies for pathology analysis in order to screen a related cancer. Early suspicion is crucial to avoid delayed diagnosis and treatment .*
Keywords: *Adenocarcinoma Anal canal, Mucinous adenocarcinoma, Abdominoperineal resection, Radiation therapy, Colorectal cancer*

I. Introduction

Anal tumors are rare neoplasms of the digestive tube that represent 5% of all anorectal neoplasms and 1,5% of gastrointestinal tumors (1). The most frequent lesion in both anal canal and perianal skin is squamous cell carcinoma (2). Around 10% of all malignant anal lesions are anal adenocarcinoma (ADC) that present a more aggressive natural history than squamous carcinoma, with shorter survival time and higher rates of local as well as distant relapse (2–4). In our study, we present two cases of mucinous adenocarcinoma of the anal gland treated in the Salah Azaiez institute of oncology, both with long proctologic medical history but with two different outcomes.

II. Case report

Case one:

A 73-year-old woman, presented to the outpatient clinic in April 2005, with anal swelling in the last 6 months. The patient had a long medical history of anal prolapse , hemorrhoids and bleeding. In the physical examination, there was a 3 cm mobile, bulging mass developed 1 cm from the anal verge in the right wall of the anal canal. The colonoscopy found a suspicious tumor of the anal canal measuring 3 cm. The biopsies of the tumor concluded to a moderately differentiated adenocarcinoma. The carcinoembroyenic antigen (CEA) was 8,2 ng/ml. The chest X ray and the abdominopelvic sonography found no abnormalities. The patient was lost of view during one year. The patient reconsulted and the physical examination did found the same bulging mass A new colonoscopy, chest radiograph and computed tomography didn't reveal any other localization. Patient was managed with Flash radiotherapy (gy in 5 times from the 7/8/2006 to 11/8/2006) followed by radical surgery consisting on an abdomino-perineal amputation (APA). Histopathologically, the anal tumor was composed of disorganized and infiltrative tubular glands infiltrating the anal wall and the perianal musculature (Fig 1). On immunohistochemical study, tumour cells expressed cytokeratin 7 and 20.One month after surgery, the patient died from a heart failure.

Case two:

A 62-year-old woman, with a long medical history of anal fistula, was addressed to our institute of oncology after the discovery of a bulging mass of the anal canal. The patient complained of acute anal pain since two months. On examination, there was a bulging mass in the anal canal with three anal fistulae. Colonoscopy showed a 4 cm mass starting from the anal verge and going up to 5 mm from the pectineal line. -iopsies concluded to an infiltrating adenocarcinoma. On immunohistochemical study, tumor cells expressed cytokeratin 7 and 20 focally (Fig 2, 3). The abdomino-perineal computerized tomography, showed no metastasis. The patient had neoadjuvant concomitant chemoradiation (45 gray of radiation associated to 5 fluouracile and folinique acide) followed by an APA . The histopathological examination of specimen concluded to a mucinous adenocarcinoma of the anal glands stage pT2 N0. The follow up of the patient was up to 8 years without recurrence.

III. Discussion

Although the anal canal is short in length, it can present a great variety of tumors, which reflects the anatomic, embryologic and histologic complexities of this structure. Tumor localization and the interpretation of morphologic findings are both controversial and, occasionally, very difficult (3). ADC of the anal canal is a rare entity. According to the World Health Organization, three types of ADC can be distinguished, mainly determined by their origin. The most common are tumors that originate in the mucosa of the superior portion of the anal canal which present with a colorectal phenotype. The age of presentation is around the sixth decade, with equal distribution between sexes (5,6). Multiple risk factors have been proposed, such as HPV and HIV, smoking and immunosuppression (2,7). The presence of chronic perianal fistulae, associated or not to Crohn's disease, represents an important risk factor, especially if lasting 10 years or more (7,8). The clinical manifestations are usually non specific. Patients may present pain, indurations, abscesses, fistulae or palpable masses. Other symptoms include bleeding, pruritus, spotting, prolapse and weight loss (4,5). Early diagnostic suspicion is crucial in order to avoid any delays in treatment. Although the clinical characteristics can lead us to suspect this type of tumor, the definitive diagnosis can only be established with biopsy and histological studies.(1)

Distinguishing between true anal canal adenocarcinoma and lower rectal adenocarcinoma with extension to the anal canal can be extremely difficult. The carcinoma of anal gland type is an anal canal tumor composed of haphazardly dispersed, small glands with scant mucine production invading the wall of the anorectal area without an intraluminal component. The glands are positive for CK 7. This definition will exclude mucinous adenocarcinomas and most rectal-type carcinomas. It does not require continuity between invasive and benign anal glands/ducts, because this criterion is very rarely fulfilled and does not prove origin of carcinoma from within anal glands. Even without examination of cytokeratin expression, a primary intramural anal canal tumor composed of small dispersed glands could be considered an anal gland carcinoma.(9) The limited number of anal canal adenocarcinoma cases published does not allow a thoroughly proven therapeutic approach (4).

Papagikos and al (10) recommend preoperative concomitant chemoradiation (CRT) followed by APA to maximize the pelvic control of the disease. li and al (11) study that included 49 patients with anal canal ADC concluded that APA together with CRT is the recommended treatment. Five-year overall survival in patients with APA alone, CRT, APA plus CRT, and with no treatment was 34.4%, 0%, 37.5% and 0%, respectively.

Chang and al (4) studied potentially curable patients, treated with local surgery followed by RT or CRT, or radical surgical treatment (APA) plus pre- or postoperative CRT. The disease-free survival was 13 months after local resection and 32 months after radical surgery, and overall 5-year survival was 43% for local treatment and 63% for patients treated with radical surgery. Currently, most authors advocate maximizing the local control of the disease, avoiding transanal resection which has had a negative impact on survival in some series(4,12). In patients with no inguinal node invasion who receive treatment with CRT either with or without associated surgery, the recommended prophylactic dose is 45 Gy (13). Due to the poor prognosis of patients who present with inguinal lymph nodes, Papagikos et al (10) prioritize the use of initial CRT (with doses above 55 Gy) and additional systemic chemotherapy, with surgical resection used selectively or for isolated local relapse. The recommended chemotherapy regimens used are classic rectal AC regimens based on 5-fluorouracil, either with or without associated oxaliplatin(4,10,14).

A greater percentage of patients with anal canal Adenocarcinoma presents with advanced disease, distant metastasis and, consequently, shorter overall survival compared with squamous cell carcinoma. Data from the Spanish National Cancer Data Base reveal that, at the time of presentation, 9.8% of patients with anal ADC are stage IV, compared with 5% of squamous cell carcinomas(1). Likewise, distant lesions occur in 28.1% of patients with adenocarcinoma compared with 11.8% of squamous cell cancers. The 5-year survival rate in patients with Adenocarcinoma is poorer in all stages than in patients with squamous cell carcinoma, with the

greatest difference observed in stage IV (13% for patients with ADC and 29% for those with squamous cell cancer) (1). There are independent prognostic factors such as the tumor size, lymph node invasion and the therapeutic management of the patient that interfere with the overall survival (15). The overall five years survival is estimated to 39% go to from 79% for T1 tumors to 9% for T4 (6).

IV. Conclusion

Anal canal adenocarcinoma is a rare entity that is occasionally difficult to distinguish from adenocarcinoma of the lower rectum with extension to the anal canal. Early suspicion is crucial to avoid delayed diagnosis and treatment. Although there is no proven therapeutic approach for the treatment of anal canal ADC, the current recommended approach is preoperative CRT followed by radical surgery (APA), with subsequent adjuvant therapy for the prevention of micrometastasis. CRT used alone should be reserved for those patients who would not tolerate radical surgery and, according to some authors, when there are proven inguinal lymph node metastases.

V. Declaration

Ethics approval and consent to participate I declare no conflicts of interest between the author And that this work was made with all the due respect to the code of ethics under the supervision of the medical and ethic comitee of the Tahar Mammouri Hospital . Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal."

Data and supporting materials section :
Google scholar have been used searching for the articles cited in the refrence list
Zotero was used fo the refrencing.
Competing interests :
I declare no compteing interests
Funding :
No source of funding
Figure legends :

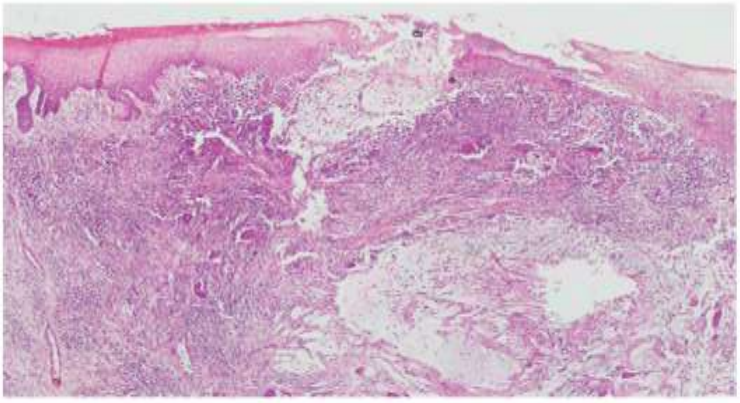

Fig.1 : tumoral glands infiltrating and ulcerating squamous anal mucosa

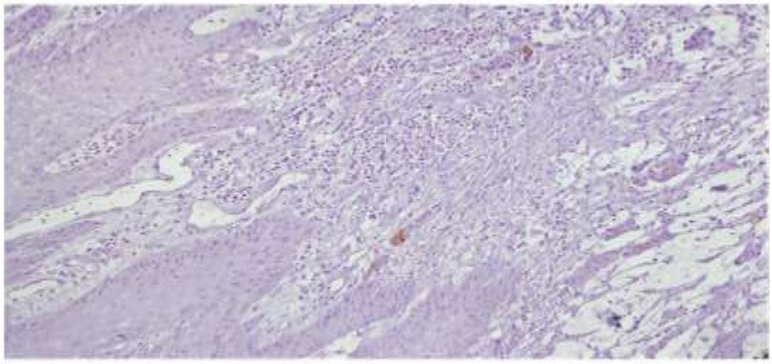

Fig 2 : rare cells express CK 20

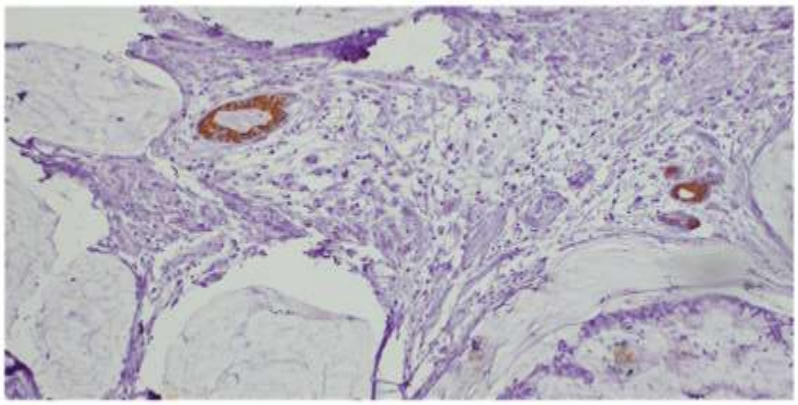

Fig 3 : tumoral glands express CK7

Authors' contribution :
Mg,Oj,Nb: data collection, review of the littérature and drafted the manuscript
Zb,Mc,Td: review of the litterature and drafted the manuscript
Km,Kr: drafted the manuscript

Acknowledegment

we want to acknowlledge the medical and non medical staff in the institute salah aziez of oncology who helped taking care of those patients and thank them for they dedication to their work.

Refrences

[1]. Márquez MF, Albendea FJV, Lozano RB, Ibáñez M del MB, Duarte ÁR. Adenocarcinoma of the Anal Canal. Narrative Review. Cir Esp Engl Ed. 2013;91(5):281–286.
[2]. Leonard D, Beddy D, Dozonis EJ. Neoplasms of anal canal and perianal skin. Clin Colon Rectal Surg. 2011;24:54–63.
[3]. Shia J. An update on tumors of the anal canal. Arch Pathol Lab Med. 2010;134:1601–11.
[4]. Chang GJ, Gonzalez RJ, Skibber JM, Eng C, Das P, Rodriguez- Bigas MA. A twenty-year experience with adenocarcinoma of the anal canal. Dis Colon Rectum. 2009;52:1375–80.
[5]. Basik M, Rodriguez-Bigas MA, Penetrante R, Petrelli NJ. Prognosis and recurrence patterns of anal adenocarcinoma. Am J Surg. 1995;169:233–7.
[6]. Belkacemi Y, Berger C, Poortmans P, Piel G, Zouhair A, Me´ ric JB, et al. Management of primary anal canal adenocarcinoma: a large retrospective study from Rare Cancer Network. Int J Radiat Oncol Biol Phys. 2003;56: 1274–83.
[7]. Lesalnieks I, Gaertner WB, Glass H, Strauch U, Hipp M, Agha A, et al. Fistula-associated anal adenocarcinoma in Crohn's disease. Inflamm Bowel Dis. 2010;16:1643–8.
[8]. Gaertner WB, Hagerman GF, Finne CO, Alavi K, Jessurun J, Rothenberger DA. Fistula-associated anal adenocarcinoma: good results with aggressive therapy. Dis Colon Rectum. 2008;51:1061–7.
[9]. Hobbs CM, Lowry MA, Owen D, Sobin LH. Anal gland carcinoma. Cancer. 2001;92(8):2045–2049.
[10]. Papagikos M, Crane CH, Skibber J, Janjan NA, Feig B, Rodriguez-Bigas MA, et al. Chemoradiation for adenocarcinoma of the anus. Int J Radiat Oncol Biol Phys. 2003;55:669–78.
[11]. Li LR, Wan DS, Pan ZZ, Zhou ZW, Chen G, Wu XJ, et al. Clinical features and treatment of 49 patients with anal canal adenocarcinoma. Chin J Gastrointest Surg. 2006;9:402–4.
[12]. Lee J, Corman M. Recurrence of anal adenocarcinoma after local excision and adjuvant chemoradiation therapy: report of a case and review of the literature. J Gastrointest Surg. 2009;13:150–4.
[13]. Adenocarcinoma of the Anal Canal. Narrative Review§ Manuel Ferrer Ma´ rquez,a,* Francisco Javier Velasco Albendea,b Ricardo Belda Lozano,a Marı´a del Mar Berenguel Iba´n˜ ez,b A´ ngel Reina Duarte a.
[14]. Warsch S, Bayraktar UD, Wen BC, Zeitouni J, Marchetti F, Rocha-Lima CM, et al. Successful treatment of anal gland adenocarcinoma with combined modality therapy. Gastrointest Cancer Res. 2012;5:64–6.
[15]. H.-N. Rakoto-Ratsimba *,a, A.-F. Rakototiana a, J. Rakotosamimanana b, A. Ranaivozanany Anal adenocarcinoma revealed by a fistula-in-ano. Report of a case.