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ПРИКАЗ БОЛЕСНИКА / CASE REPORT
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Buschke–Löwenstein Tumor: Squamous Cell Carcinoma of the Anogenital Region

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SUMMARY

Introduction Buschke–Löwenstein tumor (BLT), as a rare form of condylomata acuminatum, was firstly described by Buschke in 1886 as a "carcinoma-like condyloma acuminatum of the penis". BLT is generally considered to be a low-grade variant of squamous cell carcinoma of the anogenital region.

Case Outline We describe a case of BLT in a 56-year-old male patient who was referred to our institute due to a large defect in the gluteal region. The biopsy of the lesion was performed and the diagnosis of BLT was made on histopathological examination. Magnetic resonance imaging of the pelvis showed the extensive vegetant lesion that significantly infiltrated pelvic organs accompanied with an enlargement of para-iliac lymph nodes. Sygmoidostomy for fecal diversion was done and chemotherapy with 5-fluorouracil and cisplatin was initiated. Unfortunately, the patient's severe condition caused fatal outcome.

Conclusion Our case points out that BLT should be treated at the initial stage in order to prevent untreatable condition which happened in our patient. Therefore, early diagnostics and staging of the disease using modern technologies are crucial in order to treat patients effectively.

Keywords: MRI scan; rectum; carcinoma

INTRODUCTION

Condylomata acuminata (gential warts) caused by human papillomavirus (HPV) is a common sexually transmitted disease with an estimated prevalence of 1% of sexually active individuals [1]. There are numerous types of HPV and almost 30 are able to infect anogenital region. Considering their malignant potential, in more than 90% condylomata acuminata are benign lesions caused by low risk HPV genotypes – mostly HPV 6 and 11. Unfortunately, patients can be infected by various types of HPV, including oncogenic high risk types - HPV 16, 18, 31, 33, 35, 45 etc. High risk HPV genotypes are responsible for approximately 50% of sqamous cell carcinomas of the anal and outer genital region [2].

Giant condyloma acuminatum or Buschke–Löwenstein tumor (BLT), as a rare form of condylomata acuminata, was firstly described by Buschke in 1886, and later by Buschke and Löwenstein in 1925 and 1933 as a "carcinoma-like condyloma acuminatum of the penis" [3]. However, until today there have been a lot of diagnostic and therapeutic controversies about this disease which will be discussed.

In this report, we describe a case of BLT that unfortunately led to fatal outcome.

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CASE REPORT

A 56-year-old male patient was referred to our institute due to a large defect in the gluteal region (Figure 1). On admission, the patient was pale, slightly intoxicated and unable to sit. He denied any risk factors for human immunodeficiency virus (HIV) infection. Except anemia, other laboratory findings including cancer antigen (CA) 19-9 and carcinoembryonic antigen (CEA) were unremarkable. Abdominal ultrasound and chest X-ray examinations were also unremarkable. The biopsy of the lesion was performed and the diagnosis of BLT was made by histopathological examination (Figure 2).

Magnetic resonance (MR) imaging of the pelvis showed the extensive vegetant lesion that significantly infiltrated pelvic organs accompanied with an enlargement of parailiac lymph nodes (Figure 3).

Since the disease was unresectable, sygmoidostomy for fecal diversion was done and chemotherapy with 5-fluorouracil (5-FU) and cisplatin (CDDP) was initiated. Nonetheless, the treatment with 5FU/CDDP had to be ceased shortly after the application due to the aggravation of the patient's anemia. The patient was discharged in satisfactory condition with prescribed symptomatic therapy and frequent wound toilet. However, the patient was in extremely poor general condition and soon died.

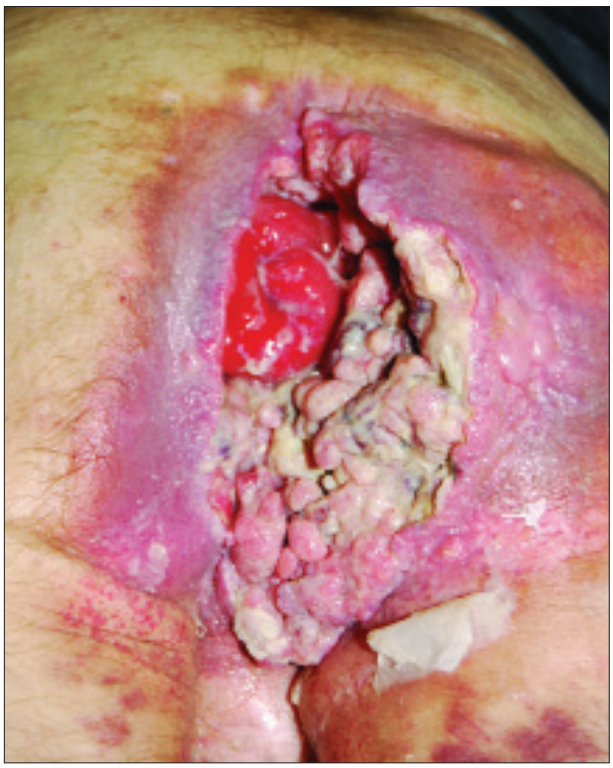

Figure 1. A massive vegetant lesion of the gluteal region with a central defect exposing the rectum is obvious

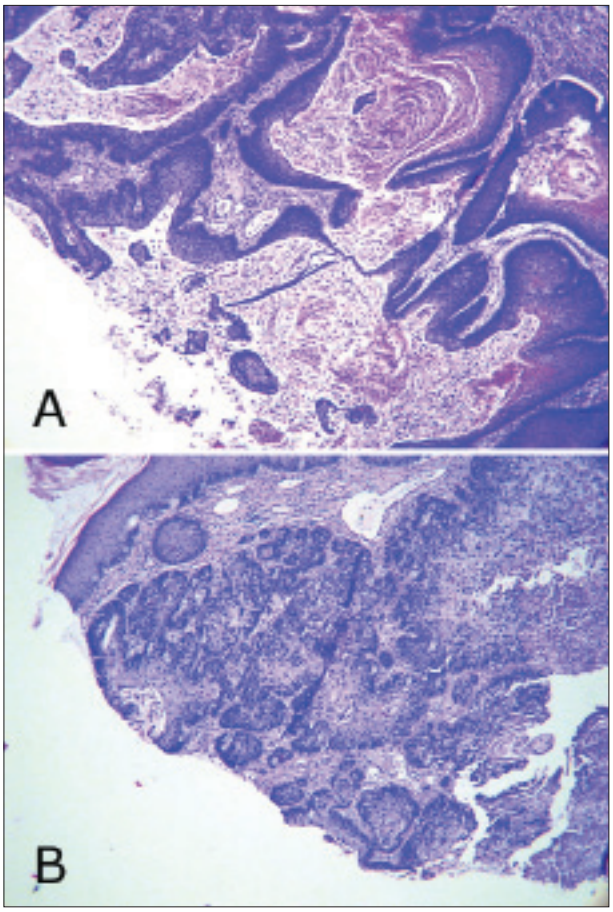

Figure 2. Papillary lesion characterized by a proliferation of squamous epithelium showing orderly maturation (A); well-differentiated invasive squamous carcinoma – infiltrative growth pattern and significant cytologic atypia are evident (B).

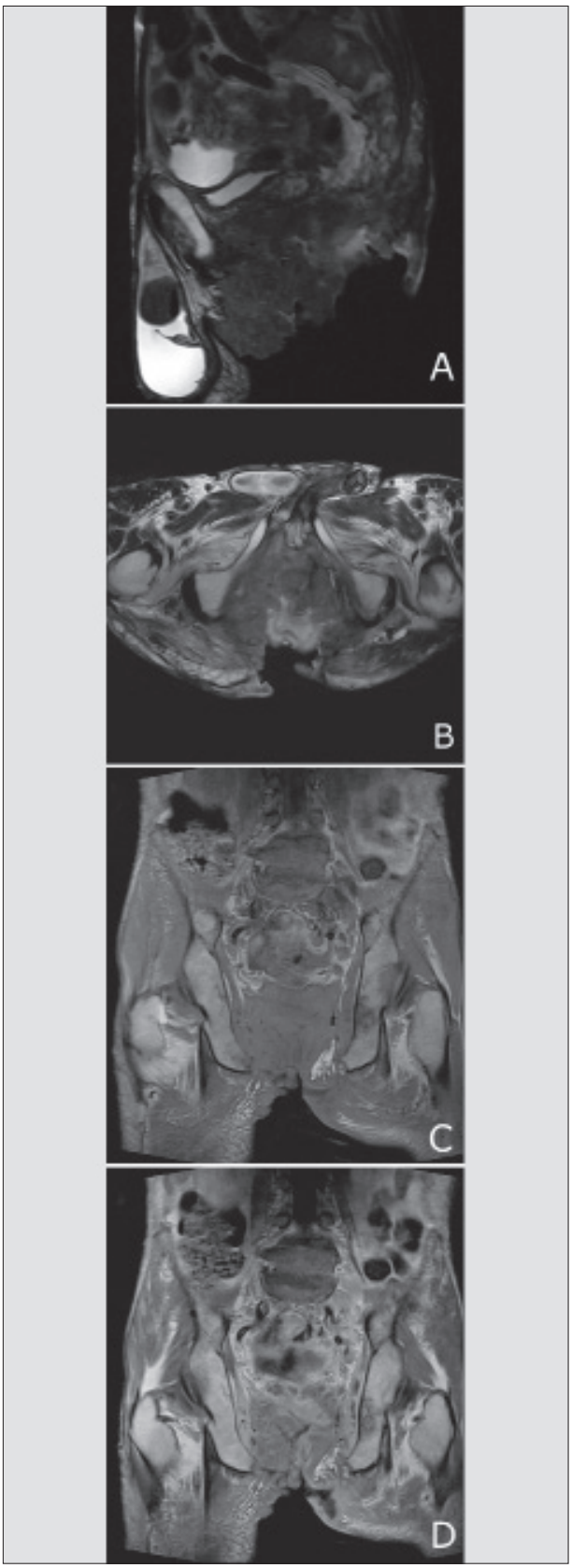

Figure 3. Magnetic resonance imaging of the pelvis, performed with a patient in prone position, shows the extensive vegetant lesion infiltrating the pelvic floor (A – T2W sagittal image), inner gluteal region bilaterally (B – T2W axial image) with invasion of the anus, urinary bladder, scrotum, penis, sacrococcygeal and pubic bone (C – T1W coronal image). Intensive postcontrast enhancement of infiltrating tissue is present (D – contrast enhanced T1W coronal image). Enlarged parailiac lymph nodes are evident (C, D).

DISCUSSION

Despite disagrements, BLT is generally considered to be a low-grade variant of squamous cell carcinoma (SCC) of the anogenital region. BLT, along with the oral florid papillomatosis of Ackerman, palmoplantar epithelioma cuniculatum and cutaneos Gottron's papillomatosis, belongs to the subgroup of SCC named verrucous carcinomas (VC) [4, 5]. On the other hand, some authors assume BLT represents a spectrum between simple genital warts and SCC [6, 7]. The size of the lesion is not a valid criterion in making a distinction between BLT and genital warts. Although genital warts can also gain a large size, the growth always remains superficial and no destruction of the underlying structures takes place. The histopathological picture of BLT is characterized by prominent papillomatosis, acanthosis, thickened rete ridges and increased mitotic activity with the lack of vascular and neural invasion. These altered rete ridges "push", rather than infiltrate, the dermis and subcutis compres the adjacent structures. Therefore, tissue destruction in BLT could be reflected through compression and displacement, instead of direct infiltration. Conventional SCC of comparable dimensions, in contrast to BLT, often results in metastasis [8].

Clinically, BLT presents as a slowly but relentlessly growing exophytic, cauliflower-like or polypoid malodorus erythematous mass with the tendency to ulcerate and hemorrhage. Secondary bacterial infection with formation of abscesses and fistulas are uncommon. The average age of affected persons with BLT is 42.9 years for males and 46.6 years for females. There is a male predominance that increases from 2.7:1 for those under 50 years to 3.5:1 for patients over 50 [9]. Except HPV infection, other etiologic contributors have been proposed, such as poor hygiene, lack of circumcision, chronic irritation and immunosuppression including HIV infection [6].

Takezawa et al. [10] were the first who described MR appearance of BLT and underlined the importance of diagnostic imaging in the staging of the disease. Even if computerized tomography is able to show local extent into the periorgan fat tissue, MR is superior in depicting the invasion of the pelvic wall muscles, rectum and prostate [6].

The treatment option for BLT is still the subject of debate, since there are no evidence based facts. However, a wide surgical excision is recommended unless infiltration of the anal sphincter or rectum exists when abdominoperineal resection is advisable. Some authors suggest a radical excision along with careful follow-up examinations as the definite treatment if the resected margins are tumor-free cells [11]. Multimodal treatment approach is reported to be a substitute for extensive surgical excisions. Local control can be achieved by chemoradiotherapy [9]. Previously used topical agents for treating perianal BLT are nowadays regarded as ineffective [12]. Radiation therapy is a controversial issue since it is connected to dedifferentiation of VC [13]. Some authors emphasize the benefit of radiation therapy as neoadjuvant or palliative [14]. Butler et al. [15] reported that otherwise unresectable BLT may be rendered operable with neoadjuvant chemotherapy and radiation. Certainly, the extensiveness of the disease in our patient did not promise such a successful outcome.

In conclusion, our case points out that BLT should be treated at the initial stage in order to prevent untreatable condition which happened in our patient. Therefore, early diagnostics and staging of the disease using modern technologies such as magnetic resonance imaging is crucial in order to treat such patients effectively.

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Бушке–Левенштајнов тумор – сквамоцелуларни карцином аногениталне регије

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КРАТАК САДРЖАЈ

Увод Бушке–Левенштајнов (*Buschke–Löwenstein*) тумор (БЛТ), редак облик шиљастог кондилома, први је описао немачки дерматолог Бушке 1886. године као „шиљаст кондилом пениса налик карциному". БЛТ се сматра нискоградусним обликом сквамоцелуларног карцинома аногениталне регије.

Приказ болесника Педесетшестогодишњи мушкарац са БЛТ упућен је у наш институт због великог оштећења глутеусне регије. Начињена је биопсија лезије и патохистолошки је постављена дијагноза БЛТ. Магнетном резонанцијом карлице уочена је екстензивна вегетантна лезија која је значајно инфилтрирала карличне органе уз увећање параилијачних лимфних чворова. Начињена је сигмоидостомија ради фекалне деривације и започета хемиотерапија 5-флуороурацилом и цисплатином. Нажалост, болесник је услед тешког стања преминуо.

Закључак Овај приказ треба да подигне клиничку свест о могућем потенцијалу БЛТ ка локалној инвазивности, што може драстично да ограничи терапијске могућности. Стога су рана дијагностика и утврђивање стадијума болести коришћењем савремених технологија веома важни у лечењу болесника.

Кључне речи: магнетнорезонантно снимање; ректум; карцином

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