Case report/Kazuistyka

Atypical lipomatous tumor of the cheek – a case report
Nietypowy tłuszczakomięsak policzka – opis przypadku

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A R T I C L E   I N F O

Article history:
Received: 19.05.2012
Accepted: 26.06.2012
Available online: 02.07.2012

Keywords:
• Atypical lipomatous tumor
• Head and neck
• Immunohistochemistry

Słowa kluczowe:
• nietypowy tłuszczakomięsak
• głowa i szyja
• immunohistochemia

A B S T R A C T

Liposarcoma is the most common soft tissue malignant tumor. It mostly arises in the subcutaneous tissue of shoulders, limbs and neck, and retroperitoneal space, but head occurrence is very rare. Atypical lipomatous tumor (ALT) is a well-differentiated liposarcoma (WDLPS) and constitutes 40–45% of all liposarcoma cases. A case of 57-year-old woman with a tumor of the left cheek, causing a discreet face asymmetry is presented. The tumor was soft and caused no tenderness. The patient reported no previous injury of the region. Fine-needle aspiration biopsy (FNAB) revealed atypical cells suspected of liposarcoma. MR examination showed fascicles of adipose tissue, which made the left cheek prominent. The patient was operated under general anaesthesia. Adipose tissue of the left cheek was removed. Postoperative course was uneventful. The final histopathological diagnosis – was atypical lipomatous tumor/well-differentiated liposarcoma (ALT/WDLPS). The patient remains under laryngological care. No recurrence of the disease has been observed during the 5 years follow-up.

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I N T R O D U C T I O N

White adipose tissue (WAT) makes up about 22% of body mass in women and about 15% in men. Brown adipose tissue, which occurs less frequently in postnatal life, is placed in the retroperitoneal and neck area. Mesenchymal malignant tumors constitute about 1% of all neoplasms, 20% of which are liposarcomas.

Atypical lipomatous tumor/Well-differentiated liposarcoma (ALT/WDLPS) accounts for 40–45% of all liposarcoma cases, being the most common adipose tissue malignant neoplasm. ALT/WDLPS can develop at any age. The greatest incidence is the 6th decade of life, with higher predilection for men [1]. A link between ALT/WDLPS and q13-15/q14-15 mutation in chromosome 12 has been proven [2–6].

ALT/WDLPS is most often found in retroperitoneal area. Subcutaneous tissue and neck area are also common locations [7]. Less frequently, it appears in craniofacial region; tongue [8, 9] and parapharyngeal space [10] cases can be found in the literature. Kuhnen has reported a rare case of ALT/WDLPS in stomach [11].

ALT/WDLPS is usually built of adipocytes or adipocytes and stroma cells. Dedifferentiation risk depends on tumor location.
and ranges from more than 20% (retroperitoneal tumors) to less than 2% (extremities tumors). Mortality rate is 0% for extremities and 80% for retroperitoneum, respectively. A case of ALT/WDLPS with leiomyoma can be found in literature [12]. The lesion is curatively treated by surgical excision. Localization ALT within the oral cavity, especially within the cheek is very rare.

Case report

Patient MM, a female aged 57 (case number 289 021), suffering from left buccal tumor, was admitted to Head and Neck Neoplasms Surgery Clinic of Medical University in Lodz. The tumor appeared 4 months before admission and caused no pain. Patient reported no injury in this particular area. Based on fine needle aspiration biopsy (FNAB), liposarcoma was suspected.

Medical history included heart valve surgery and cholecystectomy, 3 and 2 years prior to this case, respectively. Patient took Syncumar on regular basis. She was a non-smoker and drank little amounts of alcohol. She was an administration clerk.

On admission, the patient had no otolaryngological complaints apart from tumor of the left cheek. In the course of examination, a small prominence of left cheek with no tenderness on palpation was reported. No further abnormalities were found. Blood morphology: lowered level of White Blood Cells (WBC) to 4.12 (laboratory values: 4.4–11.3), WBC% values remained normal. Remaining blood tests showed no irregularities.

Chest X-ray revealed postinflammatory congestion in supradiaphragmatic region, slight cardiac enlargement, metal stitches behind the sternum and heart valves. Neck USG showed no enlarged lymph nodes. MR investigation was performed after intravenous administration of contrast solution and disclosed minor prominence of the left cheek (Fig. 1). The outgrowth was a result of extended amount of adipose tissue on anteriodistal surface of masseter muscle. Other buccal structures manifested no abnormalities.

Patient was qualified for surgical excision of the tumor. She received no Syncumar pre- and postoperatively, with Clexane being given as replacement. Under endotracheal general anaesthesia, left buccal adipose tissue was completely removed from preauricular incision. The tumor size was 3 cm × 2 cm. Minute bleeding occurred during procedure and wound was closed using layered and continuous sutures. Postoperative healing was uneventful.

Postoperative samples were routinely stained with H + E (Fig. 2a–d) and by immunohistochemistry with S-100 (positive outcome) (Fig. 2e), P53 (negative), Ki67 (isolated cells positive). MDM2 (negative) and p16 (positive) (Fig. 2f).

Histopathological diagnosis: was consistent with atypical lipomatosus tumor/well differentiated liposarcoma (ALT/WDLPS).

Patient was discharged 4 days after surgical procedure in good local and general condition. She remains under ambulatory care – no local recurrence has been reported during 5-years observation.

Discussion

Retroperitoneal, subcutaneous, intramuscular, trunk, extremities and neck ALT/WDLPS are the most frequent locations reported in the literature. Craniofacial cases are particularly rare.

ALT/WDLPS are slowly growing tumors, occasionally characterised as locally malignant. Even when growing large in size they cause no pain. Most often, patients seek medical advice due to noticeable asymmetry or development of small, intender tumor (as in the presented case). ALT/WDLPS can also be diagnosed in the course of diagnostic imaging performed for different purpose.

The largest ALT/WDLPS are usually found in retroperitoneal space. This could be caused by specific location where tumor growth is not restrained as compared to intramuscular area. When localized in subcutaneous tissue of extremities, trunk or neck, the tumor is noticed much easier, which determines early diagnosis and treatment.

MR examination enables differential diagnosis of tumor originating from adipose tissue from those of other soft tissue origin, and is a recognized form of diagnostic imaging in cases of ALT/WDLPS. Computer tomography is also commonly performed. Einarsdottir et al. stated that when adipose tissue constitutes 75% of a tumor, liposarcoma can be suspected on MR [13, 14].

FNAB is not sufficient diagnostic method in patients with ALT/WDLPS as the high ratio of well differentiated cells and only suggested the diagnosis of liposarcoma. Histopathologic examination confirmed the diagnosis. Immunohistochemistry provides better diagnostic value and requires greater amounts of material than thin needle biopsy.

Immunohistochemical expression of MDM2 and CDK4 [5, 6, 15, 16] had a diagnostic value in confirmation of diagnosis. He et al. [17, 18] recommended p16 protein as a highly specific marker for ALT/WDLPS, alternative to MDM2, that allows to distinguish it from deep-seated lipoma.
Fig. 2 – Histopathological pictures of ALT: (a) the tumor was compound of the mixture of adipose and fibroblastic septae with large atypical cells (HE × 100), (b) the infiltration of the salivary gland (upper right) by the neoplastic tissue (HE × 200), (c, d) the major magnification of the fibroblastic septae revealed the atypical adipocytic cells (diagnostic clue (HE × 400), (e) nuclear staining (brown) of the large atypical cell with anti-S-100 p confirms the diagnosis (S-100 DAKO, Hematoxylin counterstaining × 400), (f) some of the atypical cells were also positive with anti-p16 (p16 DAKO, Hematoxylin counterstaining × 400)
Incomplete excision may result in recurrence [19]. Radio- and chemotherapy are ineffective in ALT/WDLPS treatment. Patients have to remain under ambulatory care, as there is a risk of a relapse. CT and MR examinations are commonly used in early diagnosis of recurrent conditions.

Conclusions

1. The best recognizing of ALT from deep-seated lipoma is very important through proper histopathological diagnosis at ancillary immunohistochemical study (p16 and/or MDM2 expression).
2. Optimal treatment for ALT/WDLPS is a surgical removal.
3. All patient with ALT must be monitoring after therapy.

Authors’ contributions/Wkład autorów

According to order.

Conflict of interest/Konflikt interesu

None declared.

Financial support/Finansowanie

None declared.

Ethics/Etyka

The work described in this article have been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans; EU Directive 2010/63/EU for animal experiments; Uniform Requirements for manuscripts submitted to biomedical journals.

The own research were conducted according to the Good Clinical Practice guidelines and accepted by local Bioethics Committee, all patients agreed in writing to participation and these researches.

References/Piśmiennictwo


