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Case Report

A Rare and Giant Dedifferentiated Liposarcoma of Buccal Fat Pad

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Abstract

Although liposarcoma is one of the most common soft-tissue sarcomas, facial localization is extremely rare. The buccal fat pad is an important anatomic structure located in the face that recently gained interest as a result of increasing research on facial anatomy. In this paper, we report a case of giant liposarcoma originating from the buccal fat pad. The precise localization of the tumor was determined preoperatively with computed tomography examination. The liposarcoma that invaded the body· and the extensions of the buccal fat pad was resected completely. The pathological examination revealed a sclerosing, dedifferenciated liposarcoma, which is known to be very rare in the head and neck region. Only 51 cases of dedifferentiated liposarcoma of the head and neck have been reported, of theses only 5 in the cheek. In this report we present the only case of a dedifferentiated liposarcoma originating in the left buccal fat pad of the cheek.

Keywords: *dedifferentiated liposarcoma, buccal fat pad, cheek, sarcoma, facial.*

INTRODUCTION

Liposarcoma is one of the most common malignant soft-tissue sarcomas in adults; however, its occurrence in the head and neck region is rare, accounting for approximately 1% of all head and neck sarcomas [1,3,5]. Among the histological subtypes, dedifferentiated liposarcoma (DDLPS) represents a high-grade variant characterized by the coexistence of a well-differentiated liposarcoma component and a non-lipogenic sarcomatous component [2,3,7]. DDLPS is typically associated with aggressive behavior, including a high rate of local recurrence and a potential for distant metastasis [3,4].

DDLPS most frequently arises in the retroperitoneum and deep soft tissues of the extremities [3,4]. In contrast, involvement of the head and neck region is uncommon, and facial localization is exceedingly rare [6-8,10]. When present in this region, DDLPS poses significant diagnostic and therapeutic challenges due to its nonspecific clinical presentation, complex anatomy, and the difficulty in achieving wide surgical margins without functional or aesthetic compromise [6,8].

The diagnosis of DDLPS can be challenging, particularly in young patients and in anatomically unusual locations. Imaging findings are often heterogeneous, reflecting both adipocytic and solid non-lipogenic components, while preoperative biopsy may fail to identify areas of dedifferentiation [6,8]. Histopathological examination supported by immunohistochemistry or molecular studies demonstrating amplification of MDM2 and CDK4 genes is crucial for establishing an accurate diagnosis [3,4].

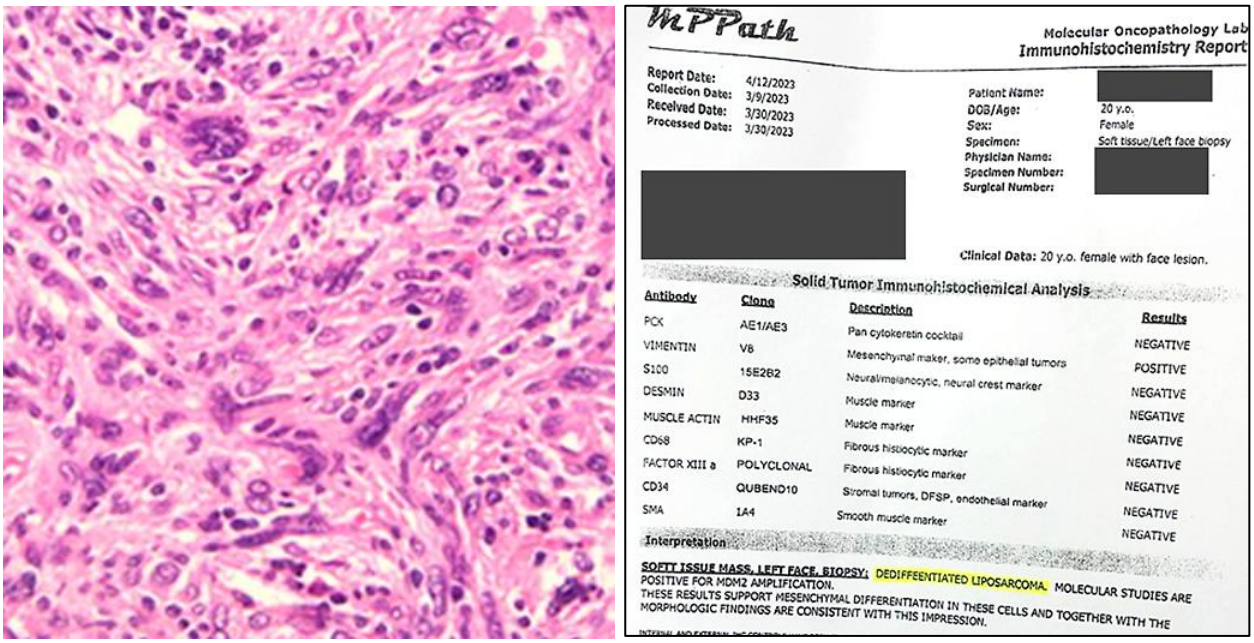
DDLPS predominantly affects patients in the fifth to seventh decades of life, with a male predominance [3,4]. Occurrence in young adults, particularly females, is exceptionally rare, and only isolated cases involving the facial region have been described in the literature [6-8].

In this report, a case of giant dedifferentiated liposarcoma originating from the left buccal fat pad is presented.

Report of a Case

A 21-year-old white man was referred to the maxillofacial surgery service of National Cancer Institute, Dominican Republic, for treatment of mass located in the left cheek and temporal region.

The patient was initially treated with radiotherapy and chemotherapy at another health center 9 months prior to our consultation.He had noticed a progressive but painless swelling for the past 6 months.



MPPath

Molecular Oncopathology Lab
Immunohistochemistry Report

Report Date: 4/12/2023
Collection Date: 3/9/2023
Received Date: 3/30/2023
Processed Date: 3/30/2023

Patient Name: [redacted]
DOB/Age: 20 y.o.
Sex: Female
Specimen: Soft tissue/Left face biopsy
Physician Name:
Specimen Number:
Surgical Number: [redacted]

[redacted]

Clinical Data: 20 y.o. female with face lesion.

Solid Tumor Immunohistochemical Analysis

Antibody	Clone	Description	Results
PCK	AE1/AE3	Pan cytokeratin cocktail	NEGATIVE
VIMENTIN	V8	Mesenchymal maker, some epithelial tumors	POSITIVE
S100	15E2B2	Neuralmelanocytic, neural crest marker	NEGATIVE
DESMIN	D33	Muscle marker	NEGATIVE
MUSCLE ACTIN	HHF35	Muscle marker	NEGATIVE
CD68	KP-1	Fibrous histiocytic marker	NEGATIVE
FACTOR XIII a	POLYCLONAL	Fibrous histiocytic marker	NEGATIVE
CD34	QUBEND10	Stromal tumors, DFSP, endothelial marker	NEGATIVE
SMA	1A4	Smooth muscle marker	NEGATIVE

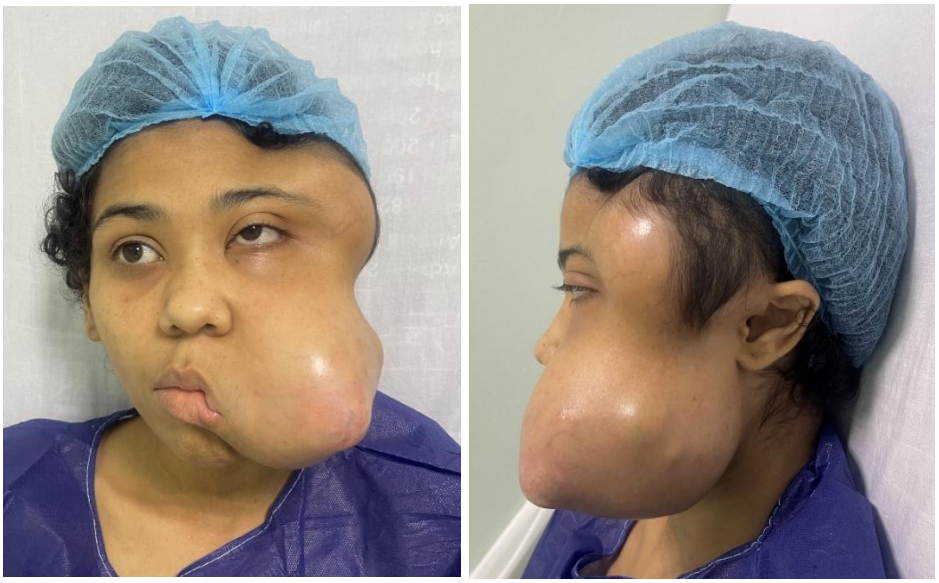
Interpretation

SOFTT ISSUE MASS, LEFT FACE, BIOPSY: DEDIFFEENTIATED LIPOSARCOMA. MOLECULAR STUDIES ARE POSITIVE FOR MDM2 AMPLIFICATION.
THESE RESULTS SUPPORT MESENCHYMAL DIFFERENTIATION IN THESE CELLS AND TOGETHER WITH THE MORPHOLOGIC FINDINGS ARE CONSISTENT WITH THIS IMPRESSION.

Figure 1. Magnification, x200. DDL shows a wide spectrum of morphological appearances, although most show the features of undifferentiated pleomorphic sarcoma (UPS) or spindle cell sarcoma (not otherwise specified) In the dedifferentiated area, bizarre multinucleate giant cells were occasionally observed. Inmunohistochemistry report is a molecular confirmation using amplification of MDM2 and CDK4.

On clinical examination, his face was asymmetric and exhibited a well-circumscribed enlargement of the left cheek. Physical examination revealed two well-circumscribed and fixed masses in the cheek and temporal (fig. 2).

The temporal mass was round and the cheek mass was lobulated. Further intraoral examination presents with moderate trismus and infiltration of the oral mucosa. The cervical lymph nodes were not enlarged. General physical examination showed the patient to be within normal limits. He comes to us with a diagnosis of dedifferentiated liposarcoma. During the preoperative period, the tumor increased in size rapidly.



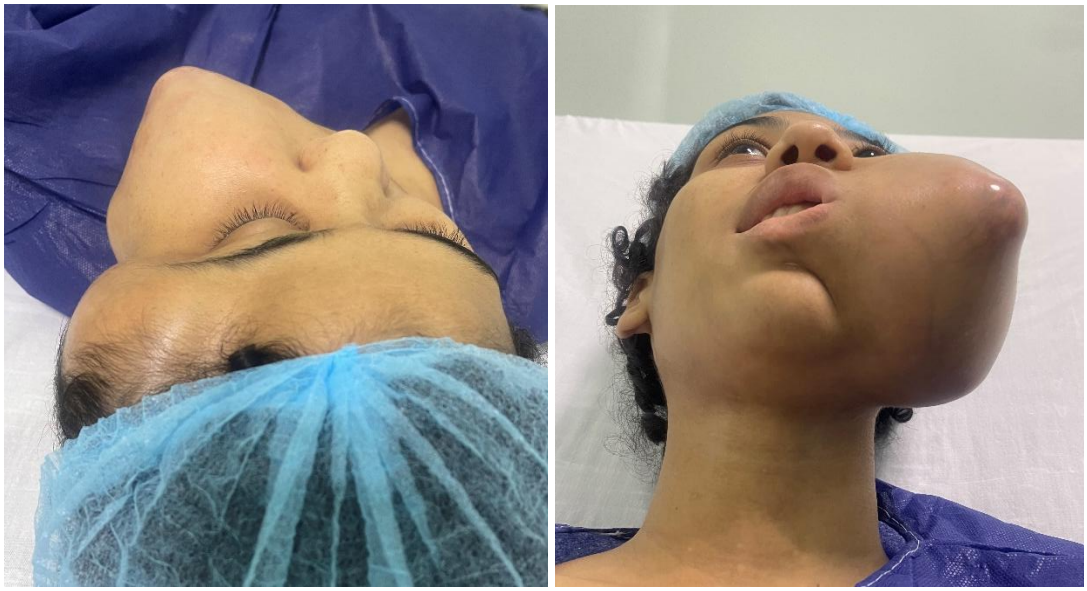


Fig. 2: Preoperative views of the patient with dedifferenciated liposarcoma: A) frontal view and B) lateral view. Postoperative photographs: frontal view (C) and lateral view (D).

MIR examination revealed a tri-lobulated soft- tissue mass with a medial extension to the infra- temporal fossa extending from the mandibular angle to the parietal bone. The mass area measuring 17 x 10 x 11 cm, respectively in the last MRI. The margins of the lesion were well defined. The mass was located under the superficial temporal fascia and in front of the masseter muscle. Tissue density measurement showed various soft-tissue and fat densities. Bony destruction was detected in the maxilla and cigomatic bones (Fig. 3). The possible imagenological diagnosis was liposarcoma.

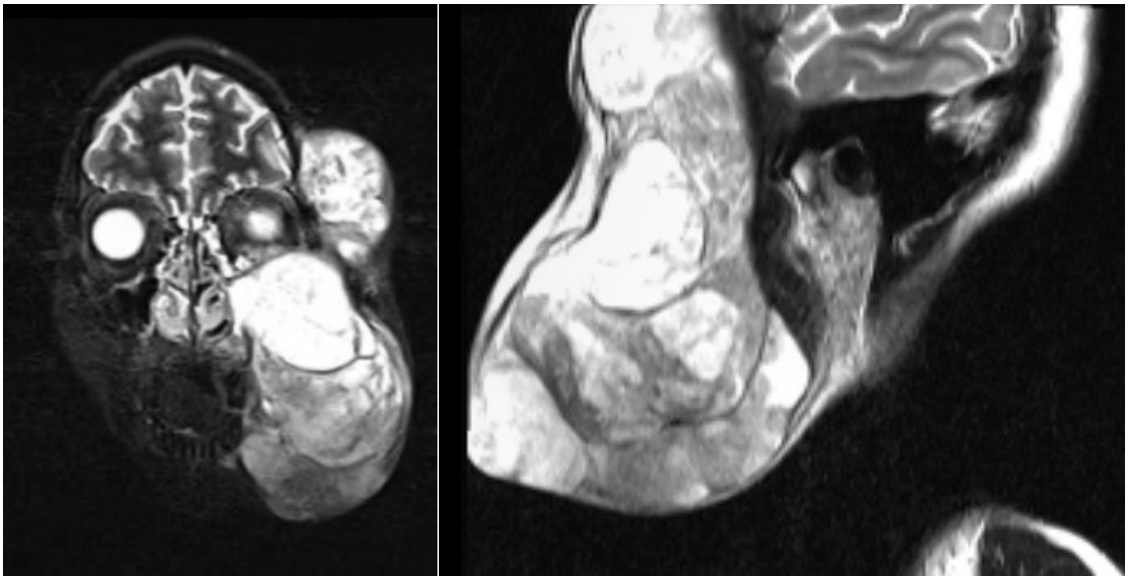


Fig. 3: MRI gives us detailed information about the location, size, boundaries, and contents of the tumor. Low signal intensity on T1 weighted images, high signal intensity on T2 weighted images and marked enhancement after IV gadolinium.

A tracheostomy was performed to administer general anesthesia. An *ad-hoc* splindel-shaped incision was made at the anterior facial level to excided the inflitrated facial skin. The posterior facial skin flap was raised at "face-lift" level, and a submandibular incision was do it.

The incision was continued with blunt and sharp dissection in some points until reaching the zygomatic arch. The superficial musculoaponeurotic system (SMAS) was also elevated and the tumor was dissected over the masseter muscle while the frontal facial nerve branches were preserved. the entire left vestibular mucosa was included in the specimen.

The tumor was sectioned upon reaching the zygomatic arch, and the zygomatic bone, which was extensively resorbed due to tumor compression, was also sectioned. A masseter muscle flp was ussed to reconstruction of the mucosal cheek.

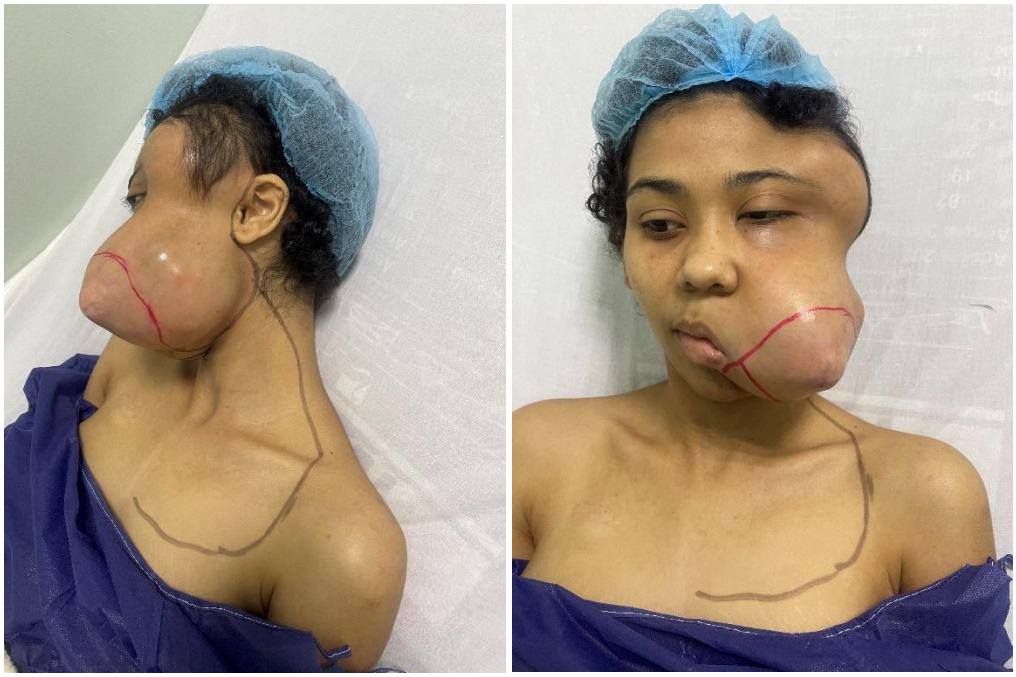
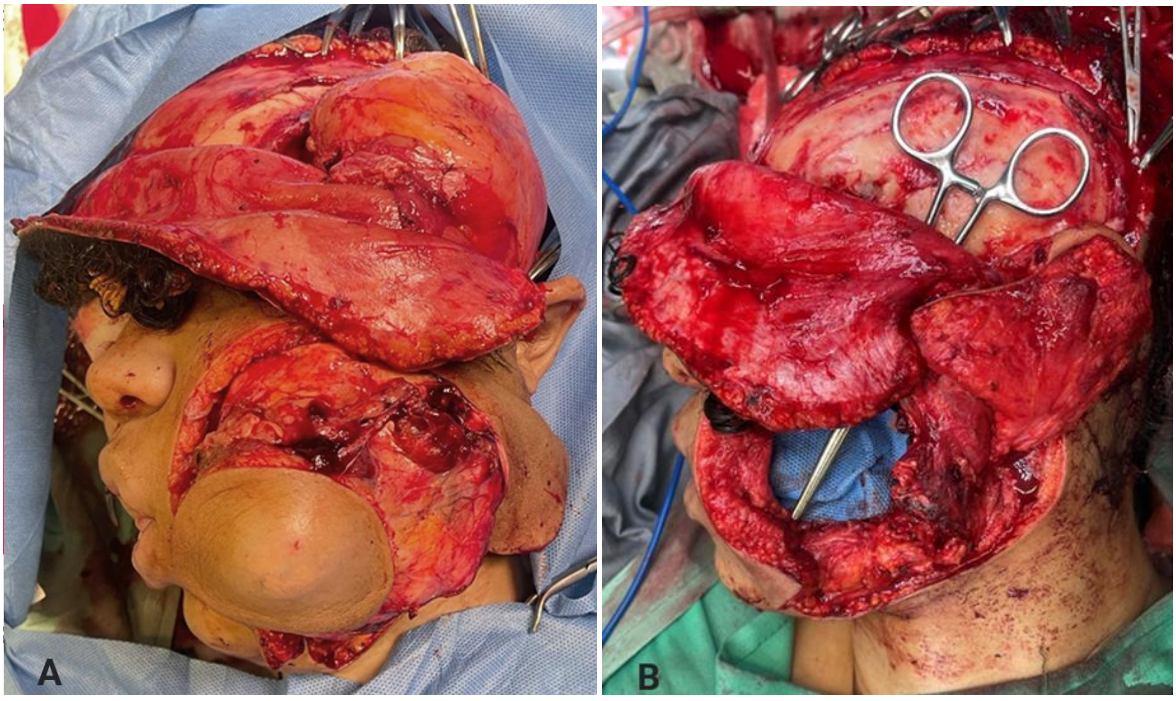


Fig. 4: A cervicopectoral flap was planned in the inicial incision design due to the possibility of cutaneous infiltration that would occupy the entire cheek. However, it was not necessary to create this flap.

The mass was located under the superficial temporal fascia and in front of the masseter muscle. The incision began in the temporal scalp approximately 5 cm above the ear and 5 cm behind the hair line and curved toward the supe- rior route of the helix. It continued inferiorly in the preauricular skin crease, followed the junction between the ear and adjoining skin around the earlobe, and from there curved down and forward to the neck like a parotidectomy incision. The temporal fascia was incised. The temporal extension of the tumor was dissected over the temporal muscle and extirpated from the infratemporal fossa.



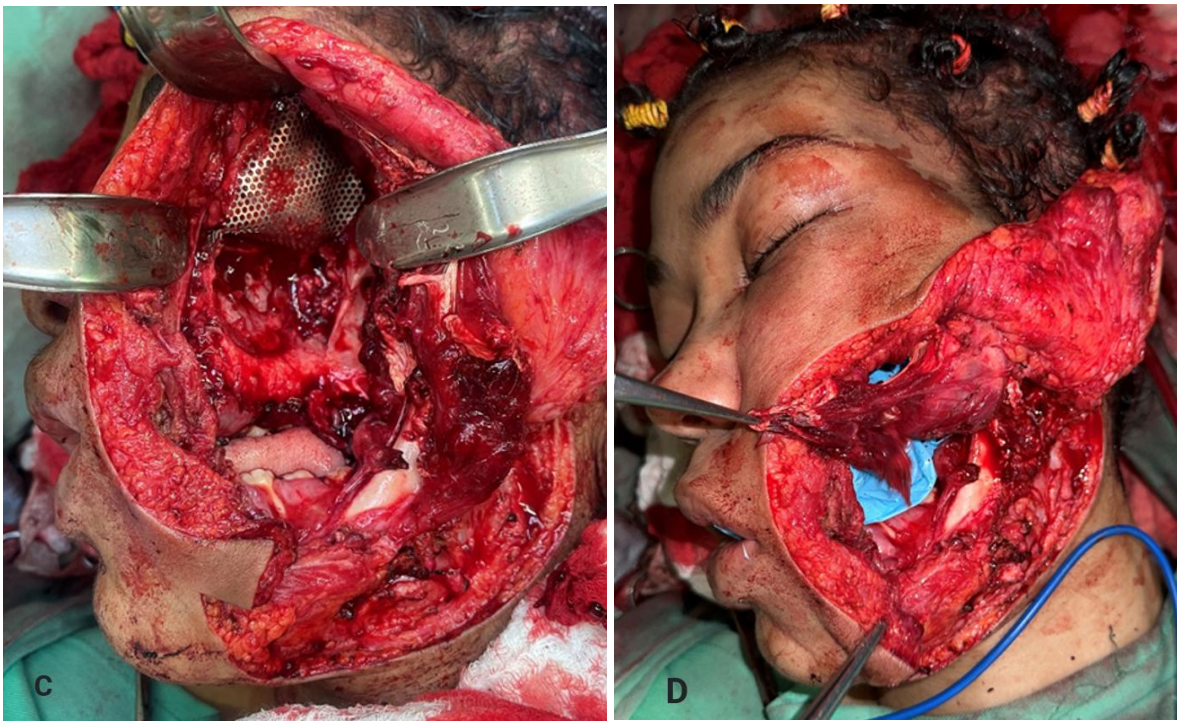


Fig. 5: A) Facial and temporal mass dissection. B) Excised tumor, in blue, area of the oral mucosa removed. C) Orbital floor and lateral wall reconstructed with titaniun mesh. D) Oral Mucosa resconstructed by masseter muscle flap.

Excess skin was excised, two drains were placed in the temporal and cheek regions, and the skin was closed. Early postoperative complications did not occur.The tumor consisted of two parts. It was rubbery and it appeared to be well demarcated and lobulated. The patient was discharged 1 week later. The patient died 6 months later from causes yet to be esta blished.

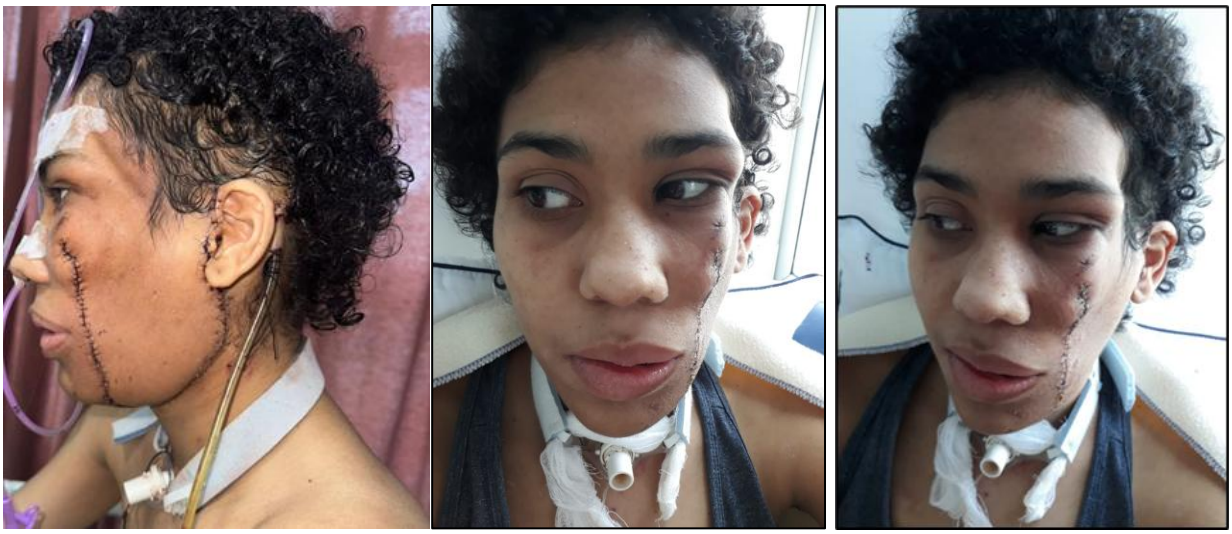


Fig. 6: Immediate post-surgical

DISCUSSION

Liposarcoma is one of the most common malignant mesenchymal tumors in adults. Since it was first described by Virchow in 1857. Dedifferentiated liposarcoma (DDL) was first described by Evans in 1979 as a tumor composed of areas of a non-lipogenic sarcoma associated with an atypical lipomatous tumor (ALT)/ well differentiated liposarcoma (WDL)

The buccal fat pad was first described by Bichat in 1802. Today, this anatomic structure is under focus again due to the widespread application of facial rejuvenation procedures. Anatomic studies reveal that the buccal fat pad is always

located under SMAS, it is surrounded by a thin capsule, and it provides a gliding surface far the chewing muscles. The main extensions are the temporal, malar, and buccal ones.

The patient presented here suffered from a liposarcoma originating from the buccal fat pad. The interesting point about this patient was that the entire buccal fat pad its body as well as the temporal, malar, and buccal extensions was invaded by the tumor. it was limited to the capsule of the buccal fat pad. It expanded and filled the adjacent anatomic spaces, but did not invade the adjacent tissues such as bone, muscle, and skin. Because of this, surgical extirpation was relatively easy and the tumor was removed completely.

Dedifferentiated liposarcoma is a high grade malignant adipocytic tumor that most commonly arises in the retroperitoneum and deep soft tissues of the extremities [3,4]. Its occurrence in the head and neck region is rare, accounting for approximately 1% of cases, and facial involvement is exceptionally uncommon [5-8].

Most reported cases affect older adults, predominantly males, making the present case of a 21-year-old female with facial DDLPS particularly unusual [3,6]. Only 51 cases of Dedifferentiated Liposarcoma of the Cheek have been reported, and only 5 in the cheek and east of the buccal fat pad [8].

Facial and cheek liposarcomas pose significant diagnostic challenges due to their rarity and nonspecific clinical presentation. These tumors are frequently misdiagnosed preoperatively as benign lipomatous lesions or salivary gland tumors [6, 7, 10]. In addition, preoperative biopsy may be unreliable, as small tissue samples can miss areas of dedifferentiation, leading to delayed or incorrect diagnosis [6,8].

Histologically, DDLPS is defined by the coexistence of a well-differentiated liposarcoma component and a non-lipogenic sarcomatous component [2, 3]. Molecular confirmation using amplification of MDM2 and CDK4 is considered a valuable diagnostic tool, particularly in challenging anatomic locations such as the head and neck [3,4].

Surgical resection remains the cornerstone of treatment for localized DDLPS [3,4,8]. Achieving clear margins in the facial region can be challenging due to the complex anatomy and the need to preserve function and aesthetics. Nevertheless, complete excision is essential, as DDLPS is associated with a high risk of local recurrence [3]. The role of adjuvant radiotherapy or systemic therapy in head and neck DDLPS remains controversial.

Although DDLPS generally carries a poorer prognosis compared to well-differentiated liposarcoma, outcomes depend on tumor location, histological grade, and completeness of surgical resection [3,4]. Data about long-term outcomes of facial DDLPS are limited due to the low numbers of reported cases.

CONCLUSION

Dedifferentiated liposarcoma of the facial region is an exceptionally rare malignancy, particularly in young female patients. Its uncommon location and nonspecific clinical presentation make early diagnosis challenging, and definitive identification often relies on postoperative histopathological examination.

Surgical excision remains the mainstay of treatment for localized disease, although achieving adequate margins in the facial region may be technically demanding.

Dedifferentiation in head and neck liposarcomas is very unusual, and we report the first case of the buccal fat pat dedifferenciated liposarcoma.

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