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Case report

The diagnosis, genetic alternation, and treatment of the primary pleomorphic liposarcoma of the femur in a rare age: Case report and literature review

Haixiao Wu [a,b,1], Ahmed K. Ghanem [c,1], Kirellos Said Abbas [d,1], Hassan Abdalshafy [e], Basel Abdelazeem [f,g], Hangchi Wang [h], Jin Zhang [a,b,**], Chao Zhang [a,b,*]

[a] *Department of Bone and Soft Tissue Tumors, Tianjin Medical University Cancer Institute and Hospital, National Clinical Research Center for Cancer, Key Laboratory of Cancer Prevention and Therapy, Tianjin's Clinical Research Center for Cancer, Tianjin, China*
[b] *The Sino-Russian Joint Research Center for Bone Metastasis in Malignant Tumor, Tianjin, China*
[c] *The Lundquist Institute at Harbor-UCLA, Los Angeles, CA, USA*
[d] *Faculty of Medicine, Alexandria University, Alexandria, Egypt*
[e] *Faculty of Medicine, Cairo University, Cairo, Egypt*
[f] *McLaren Health Care, Flint, MI, USA*
[g] *Michigan State University, East Lansing, MI, USA*
[h] *Clinical Medical College of Tianjin Medical University, Tianjin, China*

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ABSTRACT

Introduction: Liposarcoma of the bone is an extremely rare and aggressive primary bone tumor. We aimed to review all liposarcoma cases in the literature and present our new young female patient with liposarcoma.

Methods: of literature review and case presentation: Electronic databases (PubMed, Scopus, Web of Science (WOS), and Google Scholar) were searched to retrieve the related cases on liposarcoma. Extraction for important clinical data was done independently by two authors to present age, gender, site, histological type, the treatment used, and clinical outcomes of survival or recurrence. In our presented case, we followed the CARE checklist.

Results: A total of 33 patients were included in the literature search: 19 (57.6 %) male and 14 (42.4 %) female, with a mean age of 42.85 ± 18.83. Seven patients (21.2 %) were less than the age of 20 years old. The most frequent subtype was primary pleomorphic liposarcoma. Most treatment options were resection, excision, and amputation with or without chemotherapy. Recurrence was reported in four cases. However, most patients died due to late diagnosis with distant organ metastasis.

Case presentation: we report a case of primary pleomorphic liposarcoma of the left distal femur of a twelve-year-old female. The patient presented with a painful lower thigh swelling, not responding to analgesics. Imaging studies were done and showed a lesion extended across the

* Corresponding author. Department of Bone and Soft Tissue Tumors, Tianjin Medical University Cancer Institute and Hospital, National Clinical Research Center for Cancer, Key Laboratory of Cancer Prevention and Therapy, Tianjin's Clinical Research Center for Cancer, Tianjin, China.
** Corresponding author. Department of Bone and Soft Tissue Tumors, Tianjin Medical University Cancer Institute and Hospital, National Clinical Research Center for Cancer, Key Laboratory of Cancer Prevention and Therapy, Tianjin's Clinical Research Center for Cancer, Tianjin, China.
E-mail addresses: dczhangj@163.com (J. Zhang), drzhangchao@tmu.edu.cn (C. Zhang).
[1] These authors contributed equally.

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distal physeal plate laterally and centrally with scattered lesions medially. Histologically, the tumor showed a uniformly pleomorphic liposarcomatous pattern with extensive necrosis. Genetic analysis showed frequent mutations in LATS2, CREBBP, and SMAD2 genes in addition to deletions and amplifications in different genetic pathways. Two cycles of MAP chemotherapy were completed before tumor excision and total left knee replacement, followed by two other MAP cycles postoperatively. Multiple lung metastases were detected on chest CT 10 months postoperatively. The patient died 13 months postoperatively.

Conclusion: Primary pleomorphic liposarcoma can present as sclerotic and aggressive malignant bone tumors at an early age. Excision, MAP, and MAPI chemotherapy are not enough to treat this tumor.

1. Introduction

Liposarcoma is the most common type of sarcoma, which originates from the mesenchymal tissue. The world health organization (WHO) has classified soft tissue liposarcomas into five distinct histologic subtypes: well-differentiated, dedifferentiated, myxoid, pleomorphic, and mixed types [1]. Generally, the prognosis of liposarcomas depends on its histological type: well-differentiated liposarcoma (WDLPS) type is a low-grade tumor with a favorable prognosis. On the other hand, dedifferentiated liposarcoma (DDLPS) and pleomorphic liposarcoma (PMLPS) types are high-grade, aggressive, and have a bad prognosis [2].

Primary liposarcoma of the bone is an extremely rare primary bone tumor. Only thirty-three primary liposarcoma cases have been reported in the literature (Table 1). Most of these cases originated from tubular bones, with only a few cases originating from the vertebrae and flat bones. Bone liposarcoma presents as a well-defined or an ill-defined painful mass. Previously, it was assumed that liposarcoma of the bone was only osteolytic lesions. However, new cases are reported with sclerotic lesions as well. The diagnosis of primary bone liposarcoma is based on the histological, radiological, immunophenotypes, and molecular features of the tumor.

Most of the reported cases of bone liposarcoma are very aggressive tumors that grow rapidly and metastasize mainly to the lung. With the only little data available about bone liposarcoma treatment, amputation surgeries and neo-adjunctive MAP chemotherapy showed slightly better outcomes [21,3].

We reported the clinical, radiological (including X-ray, MRI, and ECT), pathological, and gene analysis findings of a case of primary pleomorphic liposarcoma of the distal femur of a twelve-year-old female. We also reviewed the literature regarding primary liposarcoma of bone and re-assess diagnostic criteria and treatment options in the light of our findings.

1.1. Methods of systematic review

Databases (PubMed, Scopus, Web of Science, and Google Scholar) were searched to retrieve related previous case reports using these keywords: liposarcoma, primary liposarcoma, pleomorphic liposarcoma, and well-differentiated liposarcoma. An additional manual search was done for any possible missed articles. The literature search and selection process were conducted by two independent reviewers. In cases where there were discrepancies in the inclusion of studies, a third reviewer was consulted to resolve the differences and make the final decision on whether to include the study. The inclusion criteria were as following: 1. the study (case report, case series, retrospective study, or prospective study) reporting primary liposarcoma regardless of age, sex, race, socioeconomic status, ethnicity, geographical area, or publication date; 2. the study must provide detailed information on diagnostic methods, and treatment approaches, along with clear results and conclusions; 3. only studies published in English will be included to ensure consistency in review and data processing (Supplementary Fig. 1). We specifically noted the regions or countries of origin of the patients reported in these studies to provide a comprehensive global perspective. We excluded duplications, editorials, commentaries, books, or discussions. Screening and extraction were done independently by KSA and HS. The disagreement was resolved by AKG.

1.2. Case description

A 12-year-old Chinese girl presented with variable knee pain and showed in severity increase after walking. The pain can be temporarily relieved by analgesics. Compared with the right thigh, swelling can be found in the anterolateral aspect of the left thigh (the left thigh circumference at the tumor was 41.5cm, while the same part in the right was 31.0 cm). No other complaints of constitutional symptoms were given.

The general timeline of the diagnosis and treatment course of the patient is described in Fig. 1. The left femur X-ray identified an osteosclerotic lesion (Fig. 2, A-B). MRI scanning on the lesion site in the left distal femur suggested a bony destruction with the formation of a soft tissue tumor (Fig. 2, C-D). As shown in MRI with T1-/T2-weighted, the lesion site showed heterogeneously hyperintense and heterogeneous contrast enhancement. Whole body bone scan depicted a single bone lesion around the whole-body bone (Fig. 2, E). The lesion can be found across the distal lateral and central physeal plate, and scattered lesions can be found medially. The intramedullary involvement of the distal femur can be found to extend around 8.0 cm from the articular surface. The soft tissue tumor was found to extend around 9.0 cm from the articular surface (Fig. 2, C-D). The popliteal neurovascular bundle was not invaded by the tumor, while it was close to the border of the tumor.

Incisional biopsy was performed, and the features of a high-grade sarcoma were found. H&E staining suggested a uniformly pleomorphic liposarcomatous pattern, and extensive necrosis (Fig. 3, A). The components of osteosarcomatous, chondrosarcomtous, or

Table 1
Summary of Patients with bone liposarcoma from Literature Review.

Author, year of publication	Sex (Age)	Subtype (Site)	Treatment	Region of Institution	Recurrence/ Follow Up (in months)	Comment
Tiemeier et al., 2018 [3]	M (18)	pleomorphic[a] (Tibia)	Resection and Chemotherapy	UK	No (12)	(Preoperative: two cycles of methotrexate, doxorubicin, and cisplatin (MAP) postoperative:5 cycles of ifosfamide and etoposide).
Boanimbek et al., 2020 [4]	F (19)	Unknown (Fibula)	Resection	West Africa	No (6)	None
Zhan et al., 2019 [2]	M (70)	Well-differentiated (Femur)	Excision	China	No (10)	None
Schneider et al., 1980 [5]	M (69)	Unknown (Fibula)	Excision then amputation	Germany	No (24)	None
George et al., 1957 [6]	F (47)	Unknown (Femur)	Amputation	South Africa	NR (1)	Died Within one month of the operation,
Halevi et al., 2015 [7]	M (70)	pleomorphic (Thoracic vertebrae (T5))	Resection, Chemotherapy, and Irradiation (adjuvant external beam radiation therapy (EBRT)).	Israel	Yes (12)	Died year after the initial
Codina et al., 2016 [8]	M (61)	Pleomorphic[a] (Lumbar 1,3 vertebrae)	Resection	Spain	Yes (2)	Recurrence after three months and died because of metastatic disease.
Moraes et al., 2012 [9]	F (60)	Pleomorphic[a] (L4 body vertebrae)	Resection then decompression, Chemotherapy, and Irradiation.	Brazil	Yes (36)	None
Pardo-Mindan et al., 1981 [10]	M (39)	Pleomorphic-like (Humerus)	Excision and Curettage	Spain	No (NR)	None
Goldman et al., 1964 [11]	M (33)	Pleomorphic (Ulna)	Resection and Reconstruction	USA	No (5)	Alive
Larsson et al., 1975 [12]	F (52)	Unknown[a] (Femur)	Curettage and Irradiation	Sweden	No (5)	Died with widespread metastases in the lungs.
Panousopoulos et al., 2004 [13]	M (72)	Pleomorphic (Femur)	Excision	Greece	No (18)	None
Rehbock et al., 1936 [14]	F (56)	Unknown[a] (Femur)	Irradiation	USA	NR (NR)	Died after 14 months.
	M (60)	Unknown (Iliac bone)		USA	NR (NR)	Died after two weeks
johnson et al., 1962 [14]	M (25)	Unknown[a] (Humerus)	Amputation	USA	NR (NR)	Died after 26 months
	M (46)	Unknown[a] (Humerus)	Amputation	USA	NR (NR)	Died about 18 months after amputation
Catto et al., 1963 [15]	F (16)	Unknown[a] (Tibia)	Amputation	UK	NR (9)	Died five months after amputation
Cremer et al., 1981 [16]	F (58)	Unknown[a] (Femur)	Amputation	Germany	Yes (30)	Died after 2.5 years after diagnosis
	F (37)	Unknown[a] (Ileum)	Cordotomy, Chemotherapy (methotrexate) and Irradiation	Germany	NR (36)	Died after three years after the diagnosis
Kenan et al., 1991 [17]	M (57)	Myxoid (Scapula)	Curettage	USA	No (36)	Three years the patient was stable and without significant roentgenographic changes
Seo et al., 2007 [18]	M (69)	Well differentiated (Temporal bone)	Resection	Japan	No (24)	Alive
Macmull et al., 2009 [19]	M (26)	Well differentiated (Femur)	Excision and chemotherapy (preoperative: 2 cycles of neoadjuvant doxorubicin, cisplatin, and methotrexate and postoperative: 4 cycles of adjuvant ifosfamide and etoposide)	UK	No (16)	Alive
Hamlat et al., 2005 [20]	F (45)	Pleomorphic[a] (Thoracic spine)	Laminectomy T7 and T8 and Irradiation (45 Gy)	France	NR (19)	Alive but physical deterioration
Lmejjati et al., 2008 [21]	M (45)	Pleomorphic (L4/L5 vertebral bodies)	Decompression L4/L5 and Irradiation (45 Gy)	Morocco	No (3)	Died
Rasalkar et al., 2011 [22]	M (13)	Pleomorphic[a] (Femur)	Resection and chemotherapy (preoperative: two cycles of MAP	China	No (13)	Alive

(*continued on next page*)

Table 1 (*continued*)

Author, year of publication	Sex (Age)	Subtype (Site)	Treatment	Region of Institution	Recurrence/ Follow Up (in months)	Comment
			(methotrexate, adriamycin, cisplatinum) and postoperative:one cycle combinations of ifosfamide/ etopoiside and adriamycin/ cisplatinum)			
Dawson et al., 1955 [23]	F (28)	Unknown[a] (Femur)	Amputation	UK	NR (11)	Died after nine months after amputation
Retz et al., 1961 [24]	M (40)	Unknown (Tibia)	Amputation	USA	No (24)	Alive
Ross et al., 1968 [25]	M (15)	Unknown[a] (Fibula)	Excision and Irradiation (cobalt teletherapy)	UK	NR (7)	Died after five months from metastases
Addison et al., 1982 [26]	M (19)	Pleomorphic[a] (Humerus)	Amputation and chemotherapy (cyclophosphamide, adriamycin, vincristine, and DTIC (dacarbazine))	USA	NR (10)	Died ten months after diagnosis
Duffy et al., 1938 [27]	M (49)	Unknown[a] (Femur)	Amputation	USA	NR (60)	Alive
Downey et al., 1982 [28]	M (15)	Pleomorphic[a] (Acetabulum of iliac bone)	Hemipelvectomy	USA	NR (2)	Died after 2 months of surgery due to pneumothoraces
Schwartz et al., 1970 [29]	M (49)	Unknown (Tibia)	Amputation	Australia	No (7)	Alive
Torigoe et al., 2016 [30]	F (36)	pleomorphic[a] (Humerus)	Resection and chemotherapy (ifosfamide, cisplatin plus doxorubicin)	Japan	No (8)	Died

[a] Reported distant metastasis to lung, bone, liver, lymph node, brain, or kidney. F; female, M; Male, NR; Not reported.

rhabdomyosarcomtous were not found. Considering there was no specific chemotherapy protocol for bone liposarcoma, based on the osteosarcoma recommendation (EURAMOS-I), preoperative MAP chemotherapy was used (two cycles of high-dose methotrexate, adriamycin, and cisplatinum). The thigh circumference at the tumor site suggested a significant suppression from the regimen (Fig. 4).

With the completion of the two cycles of MAP, at week 20, the girl underwent complete tumor excision (including biopsy incision) and total left-knee replacement. As shown in Fig. 5, inside the resected left distal femur, the tumor can be found in the metaphysis (length 8cm, across 4.5 × 3.5 cm). The tumor eroded through the lateral bone cortex and extended into the epiphysis. The hyaline cartilage was not involved, and the knee joint surface was not destroyed. In the cut split surface of the tumor, a rough surface can be seen. The yellow adipose tissue wrapped with brown bony tissue can be found (Fig. 5). Postoperative rehabilitation exercises were successfully given to the girl (Fig. 2, I).

The pleomorphic liposarcoma in the resected tumor was found with the microscopic examination (Fig. 3, B). The resection margins examination suggested a negative margin. The chemotherapy-induced tumor necrosis was reported to be 60 %, which was considered an unfavorable response. Thus, postoperative, two cycles of chemotherapy consisting of MAPI (methotrexate, Adriamycin, Cisplatinum, Ifosfamide) were given.

This patient's primary tumor specimens were further analyzed through another NGS test targeting 525 cancer-associated genes (all exons or hotspots) at a CAP-certified laboratory (Simcere Co., Nanjing, Jiangsu Province). The detailed somatic genetic alterations are summarized in Table 2.

At ten months post operation, the occurrence of multiple lung metastases was observed through chest CT (Fig. 2, H). The girl and her parents refused to receive further treatment and died around 13 months Postoperative. There was no local recurrence occurrence at the end of the patient's life, and she kept the ability of independently ambulatory.

The patient's family expressed a mix of hope and concern throughout the diagnosis and treatment process. They appreciated the multidisciplinary team's efforts and the comprehensive care plan. The initial diagnosis was a shock, but the family was grateful for the clear communication and support from the medical team. Despite the challenging journey and the eventual outcome, the family valued the dedication and compassionate care provided by the healthcare professionals.

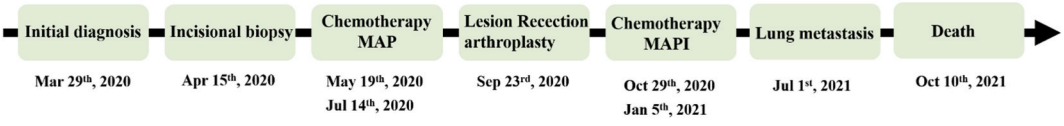

Fig. 1. Illustrating the timeline of the treatment course of the patient.

2. Discussion

Primary liposarcoma of the bone is an extremely rare tumor to the point that it was only reported in the form of a single case report. After reviewing multiple databases, we only found thirty-three case reports of the primary liposarcoma of the bone in the literature (Table 1). Table 1 summarizes the demographics, treatment modalities, and outcomes of patients with primary liposarcoma as reported in the literature. The data highlights the variability in patient age, tumor location, and treatment approaches, emphasizing the need for individualized treatment plans. The study highlights the rarity and aggressive nature of primary pleomorphic liposarcoma of the bone, particularly in younger patients. The cases reviewed originate from diverse geographical regions, including Europe, Asia, Africa, North America, and Australia, indicating that this rare tumor type affects patients worldwide. This international representation underscores the need for a global collaborative approach to better understand and treat this malignant tumor. To our knowledge, our case is the youngest case with the primary liposarcoma in femur. Most of the reported cases presented with painful mass, mainly in the long tubular bones (femur, fibula, tibia, and humerus). Fewer cases were also reported in the vertebrae, scapula, iliac bone, and temporal bone. Nineteen males and 14 females were reported with a wide range of ages from 13 to 70. Only 7 cases were reported before the age of 20 years old. The reported cases included different subtypes of liposarcoma of the bone. The most frequently reported subtype was primary pleomorphic liposarcoma which is known to be a high-grade tumor with a poor prognosis and a high recurrence rate [31].

Patients expired after a short time from receiving treatment because of recurrence or metastasis to vital organs, mainly the lungs. Rehbock et al. [14] reported metastatic nodules in the left ventricle of the heart. Non-cancer causes of death and complications of chemotherapy in cases of distant metastasis should be considered [32,33]. Tiemeier et al.[3] [][3][]reported a significant reduction in the ejection fraction post-operatively and after two cycles of neoadjuvant chemotherapy using MAP. They changed the adjuvant chemotherapy regimen to ifosfamide and etoposide to avoid this cardiotoxicity. WHO defined the liposarcoma of the bone as a malignant neoplasm whose phenotype recapitulates fat and arises within or on the bone surface [34]. Downey et al. [28] also added that histologically, a predominance of immature pleomorphic, often bizarre, uni-globular, and multi-globular lipoblasts should exist beside the primary origin from within the bone for the diagnosis of primary bone liposarcoma to be considered. Based on that, the diagnosis of many of these cases as primary liposarcoma of the bone was questionable because it was made based on the cytomorphological ground of the lesion and the location of the entire tumor in the bone was uncertain as well [3]. On the other hand, our case report fulfilled these two main criteria for diagnosing primary pleomorphic liposarcoma of the bone. First, the tumor originated primarily from the lower end of the femur and did not metastasize from a different site. Second, on histological examination, the tumor cells had almost pure pleomorphic liposarcoma characteristics with the expression of adipocyte biomarkers. Our sample also was negative for the S-100 marker, which is noted to be absent in more than 50 % of the soft tissue pleomorphic liposarcoma [3].

Accurate diagnosis and differentiation between atypical lipomatous tumors (ALT) and the different types of liposarcoma (WDLPS and DDLPS) is challenging. To date, there are no pathognomonic molecular biomarkers for early detection or differentiation of different types of liposarcomas preoperatively. In addition to the anatomical location and imaging techniques, immunohistochemical (IHC) and genetic analysis of the tumor are used for definite diagnosis of the primary liposarcoma of the bone. IHC and gene amplification of MDM2, CDK4, and p16 were reported to be helpful in differentiation between the liposarcomas types [35]. The

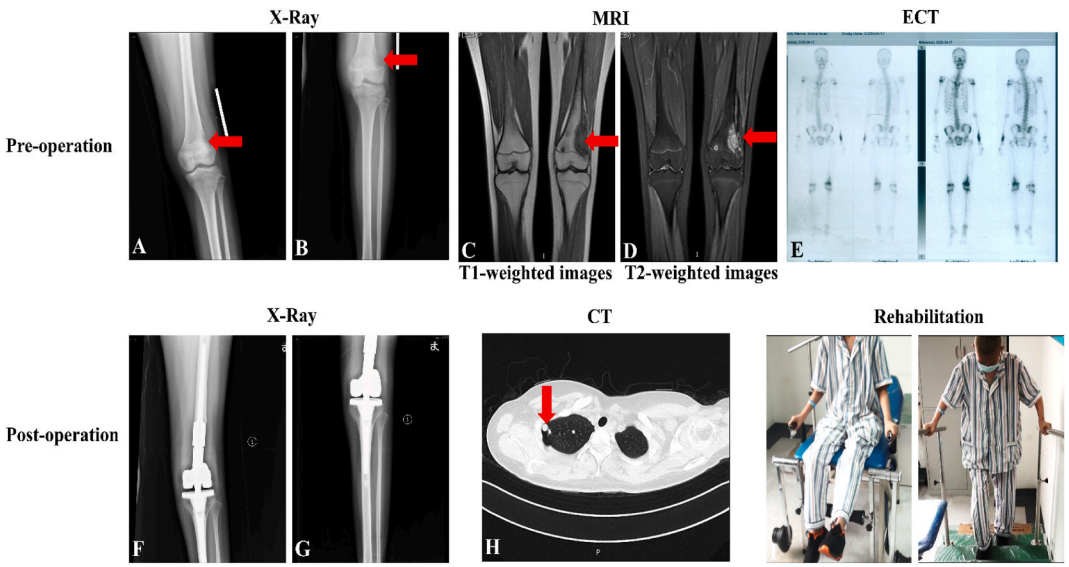

Fig. 2. Pre- and post-operative imaging and rehabilitation. (A) Pre-operative X-Ray of knee lesion (red arrow). (B) Pre-operative X-Ray of tibia lesion (red arrow). (C) Pre-operative MRI T1-weighted image showing the lesion (red arrow). (D) Pre-operative MRI T2-weighted image showing the lesion (red arrow). (E) Pre-operative ECT scan indicating bone activity. (F) Post-operative X-Ray showing knee implant. (G) Post-operative X-Ray showing tibia implant. (H) Post-operative CT scan of chest (red arrow). (I) Rehabilitation process during physical therapy.

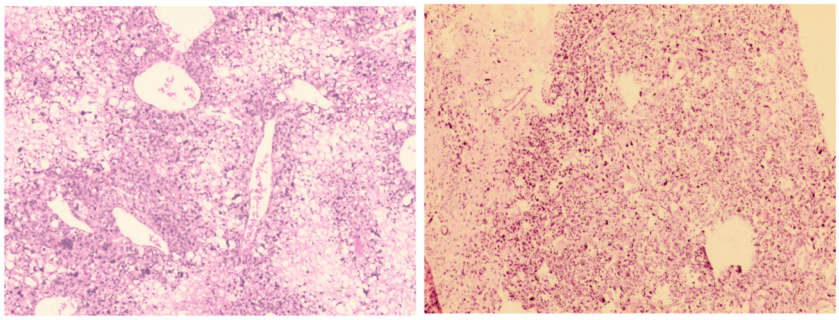

Fig. 3. Pathological images of the lesion site. Incisional biopsy(A) and post complete tumor resection.

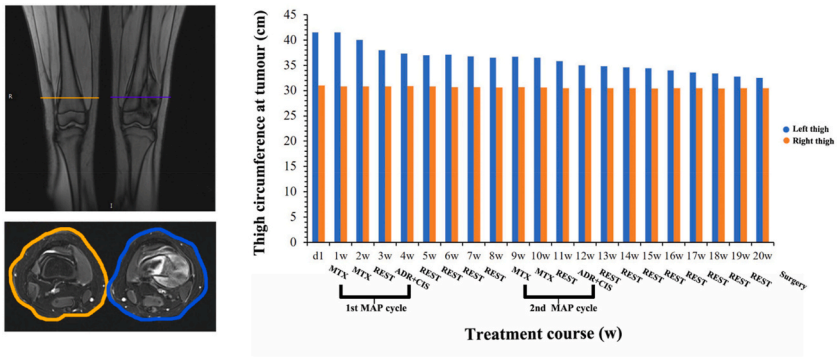

Fig. 4. Tight circumference showing significant change after chemotherapy.

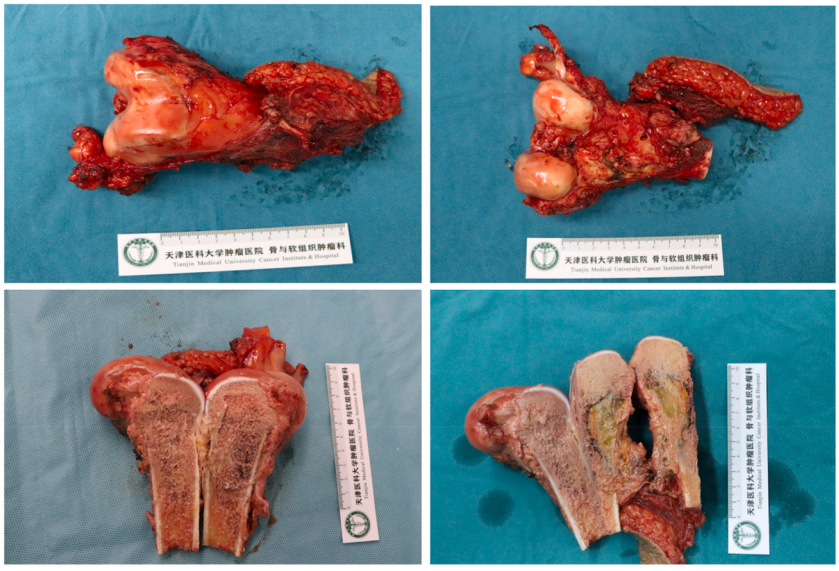

Fig. 5. The complete resection of lesion site. Anterior view(A), posterior view(B), longitudinal section of distal femur(C), and longitudinal section of tumour(D).

combined expression level of three adipose tissue-specific metabolic genes (PNPLA2, LIPE, and PLIN1) is reported to accurately distinguish between ALT, WDLPS and DDLPS as well [36]. IHC analysis of our sample was positive for MDM2. The patient did not have a family history of cancer; however, the genetic analysis of the specimen showed multiple mutations in different genetic pathways. The most frequent mutations were in LATS2 and CREBBP genes (Table 2). The genetic mutations identified in our case, including LATS2, CREBBP, and SMAD2, suggest potential diagnostic markers and therapeutic targets for primary pleomorphic liposarcoma. These

Table 2
Somatic Genetic Alterations of the reported Case.

Pathway	Gene	Mutations	Copy number variations	Frequency (%)
Hippo pathway	LATS2	p.P519L	N/A	47.54 %
gene transcription	CREBBP	p.P1991L	N/A	34.57 %
TGF-β pathway	SMAD2	c.785-17_785 -10delATTTT TTT	N/A	3.90 %
Cell cycle pathway	RB1		Deletion	
	CCNE1		Ampilcation	
			Deletion	
P53 pathway	TP53		Deletion	
Receptor Tyrosine kinase pathway	ERBB4		Deletion	
	FGFR4		Ampilcation	
Apoptosis pathway	CASP8		Deletion	
Immunology signaling pathway	CTLA4		Deletion	
Cellular metabolism signaling pathway	IDH1		Deletion	
Nuclear receptor signaling pathway	ZNF217		Ampilcation	
Notch pathway	PHOX2B		Deletion	
PI3K/AKT signaling pathway	SLC34A2		Deletion	
DNA damage/repair signaling pathway	NSD1		Ampilcation	

mutations indicate disruptions in the Hippo, chromatin remodeling, and TGF-β signaling pathways, respectively, which could be exploited for developing targeted treatments. Alternative or emerging treatments for similar cases could include targeted therapies and novel chemotherapeutic agents. For instance, therapies targeting specific genetic alterations identified in the tumor, such as those affecting the LATS2, CREBBP, or SMAD2 genes, could be explored. Future research should explore these pathways further to understand their roles in tumorigenesis and to develop novel therapies, potentially improving clinical outcomes for patients with similar genetic profiles.

To date, there is no specific treatment protocol for primary bone liposarcomas, and the protocol used for the treatment is based on either soft tissue liposarcoma or osteosarcoma protocols. Radical surgical resection is the main treatment of liposarcoma of the bone. The curative effect of chemotherapy on liposarcoma is questionable. In our case, two cycles of neoadjuvant chemotherapy shrined the tumor size, but it only induced 60 % necrosis. Also, adjuvant chemotherapy did not prevent the recurrence. Perioperative radiotherapy was reported to reduce the liposarcoma size and reduce the recurrence rate [31]. The response to the treatment is mainly dependent on the grade and subtype of the liposarcoma. Eventually, multimodality therapy, including surgical resection, perioperative chemotherapy, and radiotherapy, might improve the prognosis of liposarcoma of the bone and reduce the recurrence rate.

With an average follow-up period of 13.5 months of the reported case reports, only four cases reported recurrence, and 17 cases had distant metastasis, most frequently in the lung. Eighteen cases were reported dead at different times post-diagnosis.

In the course of the literatures review, there are some limitations in this study. First, selection bias may result from including only English-language studies, potentially excluding relevant studies in other languages. Second, most of the previous literature does not provide descriptions of the somatic genetic alterations of the disease, making it impossible for us to conduct comparative references between cases.

3. Conclusion

Primary pleomorphic liposarcoma of the bone is an extremely rare high-grade malignant tumor that can affect children. Accurate diagnosis requires immunohistochemical studies and genetic analysis. Surgical resection combined with perioperative chemo and radiotherapy might give a favorable prognosis compared to single-line treatment. The tumor is very aggressive and has a bad prognosis.

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Ethical approval and informed consent

All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional and national research committees. Written informed consent was obtained from the patient to publish this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Data availability statement

The related data and materials are available for sharing upon request to Jin Zhang, Chao Zhang.

CRediT authorship contribution statement

Haixiao Wu: Writing – original draft, Investigation, Formal analysis, Data curation. **Ahmed K. Ghanem:** Writing – original draft, Methodology, Formal analysis, Data curation. **Kirellos Said Abbas:** Writing – original draft, Methodology, Formal analysis, Data curation. **Hassan Abdalshafy:** Validation, Supervision. **Basel Abdelazeem:** Visualization, Software. **Hangchi Wang:** Visualization, Software. **Jin Zhang:** Writing – review & editing, Project administration, Conceptualization. **Chao Zhang:** Writing – review & editing, Funding acquisition, Conceptualization.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.heliyon.2024.e36953.

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