Head and Neck Myopericytoma (MPC): A Case Report of Double Synchronous Sinonasal MPC

Ann. Ital. Chir., 2024 95, 5: 760–766
https://doi.org/10.62713/aic.3484

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AIM: Myopericytoma (MPC) is a rare tumour characterized by a perivascular proliferation of pericytic cells with myoid differentiation and a typical spindle shape. Except for the rare malignant cases, MPC mostly shows a benign course. Symptoms are often non-specific, and the diagnosis could be accidental. Simple biopsies are often non-diagnostic and do not provide any information about the benign or malignant course of the disease. General agreement for its management is lacking.

CASE PRESENTATION: An old patient was referred to our tertiary cancer centre for left nasal obstruction for the previous three months. No worker risk factors were reported. The nasal endoscopy with enhanced endoscopic systems equipped with digital post-processing image enhancement technology (I-SCAN) and Narrow Band Imaging (NBI) revealed a non-bleeding reddish mass located at the anterior third of the left nasal fossa floor, about 1 cm in size and posteriorly a second more minor similar lesion at the level of the nasal septum. The patient underwent a radical endoscopic excision with diagnostic and curative intent.

RESULTS: No significant intra-peri- and postoperative complications were recorded. The final histopathological exam revealed a double synchronous sinonasal myopericitoma removed with safe margins. The patient is still alive with no evidence of disease after three years from surgery.

CONCLUSIONS: MPC is a challenging disease that must be considered in the differential diagnosis of all the vascular lesions of the head and neck region. Surgery should be performed with radical margins to provide a definitive cure. The endoscopic approach may allow a radical removal with a low risk of surgical complications, allowing the possibility of removing representative material for an accurate histopathological diagnosis.

Keywords: myopericitoma; head and neck; sinonasal surgery; vascular tumour; clinical pearls

Introduction

Myopericytoma (MPC) is a rare mesenchymal tumour characterized by a perivascular proliferation of pericytic cells with myoid differentiation and a typical spindle shape. According to the World Health Organization Classification of Soft Tissue Tumours, Glomus tumour Not Otherwise Specified (NOS), MPC, Myofibroma, and Angioleiomyoma belong to the same pericytic tumours group, including a broad spectrum of perivascular neoplasia with variable contractile phenotype [1]. MPC mainly affects middle-aged men and is often found at the level of the skin and soft tissues of the extremities. MPC rarely occurs intramuscularly, intraosseous, intra-viscerally, or in the central nervous system [2, 3]. Generally, most lesions tend to exhibit a benign clinical course, and a complete surgical excision is curative. However, some recurrence and malignant behavior cases have been described [4, 5]. Nowadays, diagnosing MPC is often challenging, and in most cases, it is an accidental finding during clinical visits for other reasons. Tailored therapies and treatments of MPC vary based on the benign or malignant course of the disease. Unfortunately, general agreement regarding their management has been lacking because of the rarity and inhomogeneity of the cases described in the literature. We described the first clinical case of a double synchronous sinonasal myopericytoma, reporting an update on all known knowledge about this rare tumour of the head and neck district.

Submitted: 6 June 2024 Revised: 7 July 2024 Accepted: 22 July 2024 Published: 20 October 2024
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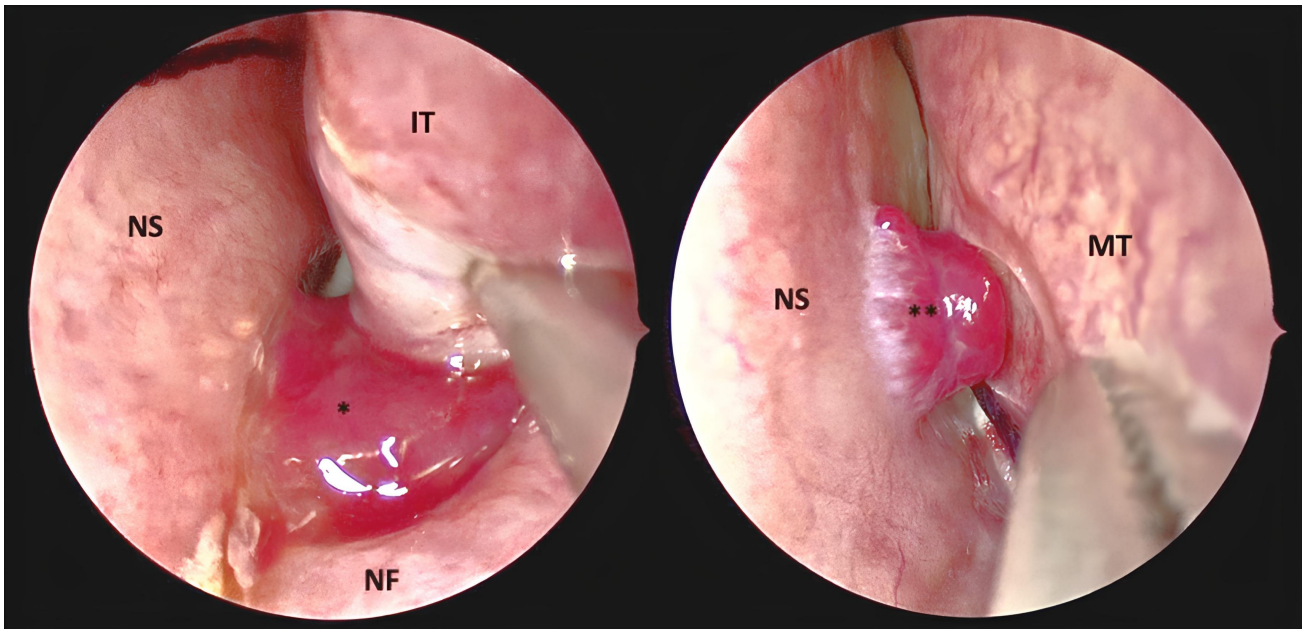

Fig. 1. Pre-operative endonasal endoscopy. NS, nasal septum; IT, inferior turbinate; NF, nasal floor; *, nasal floor neoformation; MT, middle turbinate; **, nasal septum neoformation.

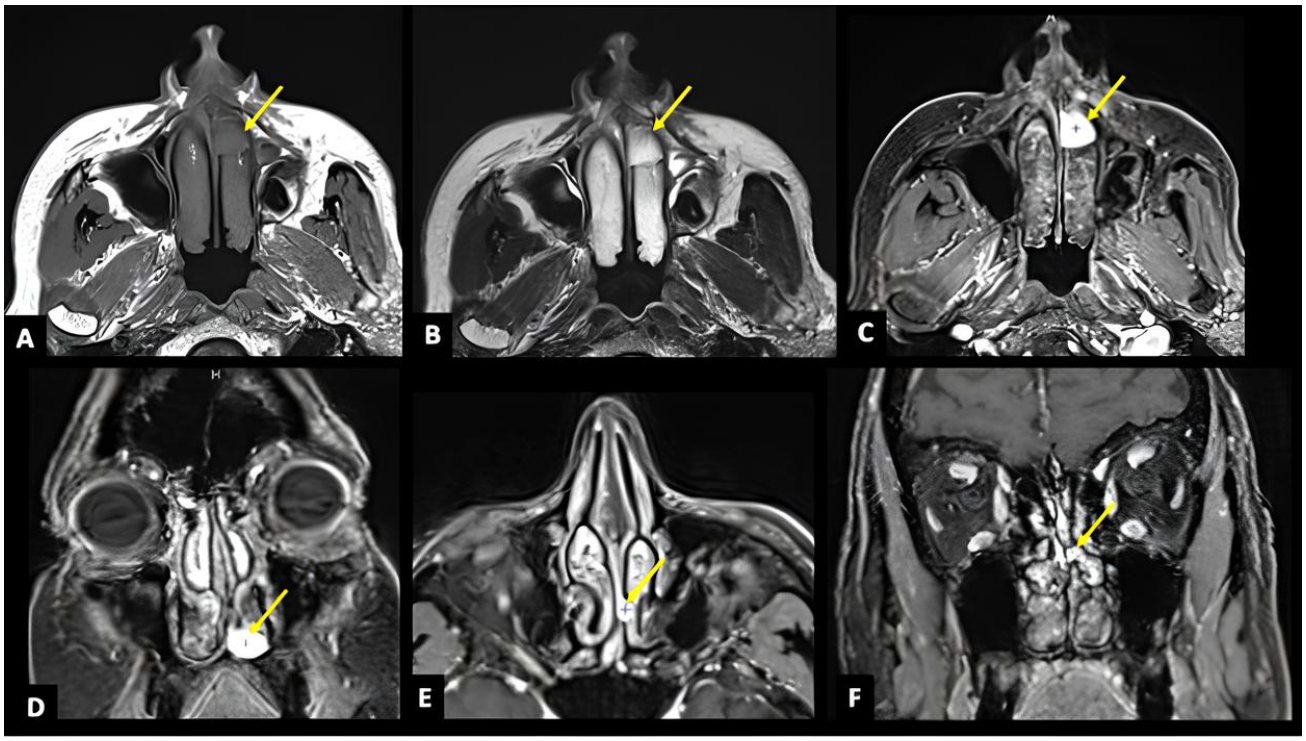

Fig. 2. Pre-operative Magnetic Resonance Imaging (MRI). Images (A–D) show the anterior nasal floor mass (yellow arrows); (E,F) show the nasal septal mass (yellow arrows). (A) Axial T1 weighted. (B) Axial T2 weighted. (C) Axial T1 with contrast enhancement. (D) Coronal T1 with contrast enhancement. (E) Axial T1 with contrast enhancement. (F) Coronal T1 with contrast enhancement.

Case Presentation

An 82-year-old woman was referred to our tertiary cancer centre, complaining about left nasal obstruction for the previous three months. The patient had no relevant past medical history. She had no rhinorrhea, epistaxis or facial pain. She was never exposed to wood or leather dust in her past and had no history of smoke or alcohol/drug abuse. According to the current standard of care, the patient underwent a nasal endoscopy with enhanced endoscopic systems equipped with I-SCAN and Narrow Band Imaging (NBI),

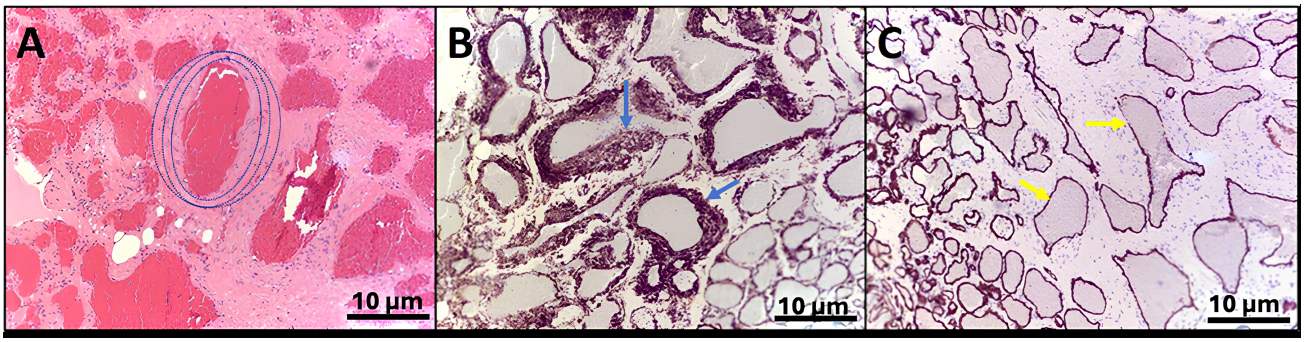

Fig. 3. Histopathologic features (Original magnification: 5×). (A) The hematoxylin and eosin exam shows vessels filled with blood and surrounded by muscle layers in an onion fashion (blue dotted lines). (B) Alpha-Smooth Muscle Actin (ASMA) staining shows a smooth muscle cell proliferating around a vessel (blue arrows) in an onion fashion. (C) Cluster of Differentiation 34 (CD34) staining shows luminal vascular cell (yellow arrows).

which revealed a non-bleeding reddish mass located at the anterior third of the left nasal fossa floor, about 1 cm in size. Posteriorly to the first mass, a second, more minor lesion was observed at the level of the nasal septum with the same endoscopic features. The right nasal fossa was inspected, and no other suspicious masses were observed at the level of the pharynx, larynx, and oral cavity (Fig. 1). The computed tomography (CT) scan showed two soft-tissue masses occupying the left nasal fossa without reabsorption or infiltration of the nasal floor's bony structures and the nasal septum. Magnetic Resonance Imaging (MRI) with gadolinium contrast enhancement confirmed the presence of both lesions, isointense to muscle in T1 sequences, slightly hyperintense in T2 sequences and characterized by homogeneous contrast enhancement (Fig. 2). The case was discussed with the multidisciplinary team board, including head and neck surgeons and a dedicated radiologist. The panel reviewed the recorded nasal endoscopy and CT/MRI imaging material. Due to the suspected lesion's vascular nature, small size, and benign endoscopic and radiologic characteristics, the patient was referred to surgery with diagnostic and curative intent under endoscopic control.

Surgical Procedure

The patient was laid supine in an anti-Trendelenburg position under general anesthesia with orotracheal intubation. Careful preparation and decongestion of the nasal cavities were obtained using nasal pledgets soaked with a 0.2% nafazoline hydrochloride solution. Surgery was performed using 0° and 45°, 4 mm endoscopes and 4K cameras with professional image enhancement systems CLARA (TH121) and CHROMA apps (XP0317224K) (Karl-Storz®, Tuttlingen, Germany). After nasal decongestion, a partial inferior turbinectomy was performed to achieve an optimal visualization of the anterior lesion on the floor of the nasal fossa. Even intraoperatively, neither lesion showed any sign of bony or septal infiltration. Thus, we performed a radical excision with safe margins. No significant intraoperative bleeding was recorded. The left nasal fossa was filled with an antibiotic ointment, followed by a nasal packing removed on the second postoperative day. Follow-up visits were scheduled after three weeks and three months: the postoperative endoscopic evaluations showed regular healing without disease recurrence. The final histopathological findings revealed a proliferation of vessels surrounded by oval spindle-epitheliod cells with eosinophilic cytoplasm and a concentric perivascular pattern with irregular hemangiopericytoma-like vessels. The neoplasm showed a vascular space of homogeneous diameter filled with blood, and the luminal endothelial cells were immunoreactive for Cluster of Differentiation 34 (CD34) (Fig. 3A–C). Immunohistochemistry underlined positivity for Cluster of Differentiation 34 (CD34) and Alpha-Smooth Muscle Actin (ASMA). The lesion did not show malignant features such as increased mitosis, atypia and necrosis. Based on the morphology and immunohistochemistry, the final diagnosis was myopericitoma (Fig. 3A–C). No significant peri- and postoperative complications were recorded. The patient is still alive with no evidence of disease after three years from surgery.

The patient realized a written informed consensus for using data for scientific purposes. The study was approved by the Institutional Ethical Committee (European Institute of Oncology IRCCS) (IEO code 1615) and complied with the principles stated in the Declaration of Helsinki "Ethical Principles for Medical Research Involving "Human Subject".

Table 1 (Ref. [2, 4, 5, 6, 7, 8, 9, 10, 11, 12, 13, 14, 15, 16, 17, 18, 19, 20, 21, 22, 23, 24, 25, 26, 27, 28, 29, 30, 31, 32, 33, 34, 35, 36, 37, 38, 39]) summarizes the most updated information on the main features of head and neck myopericytoma taken from the review of the scientific knowledge known so far. To make the review more homogenous and selectively focused on the head and neck pathology, vertebral and intracranial myopericytoma were excluded.

Table 1. Contemporary review on the main features of the head and neck myopericytoma.

Year	Author	Site	Age	Sex	Max size (cm)	Recurrence/persistence	Follow up (months)
2020	Chaskes MB *et al*. [2]	DST	23	F	2.5	No	5
2002	McMenamin ME *et al*. [4]	SST	81	F	2	Yes (liver)	24
2012	Terada T [5]	Oral cavity	61	M	1	No	6
2012	Jung YI *et al*. [6]	Multicentric	32	F	2.5	No	48
2009	Lau PPL *et al*. [7]	Multicentric	42	M		Yes (liver and spleen)	48
2010; 2013	Xia L *et al*. [8]; Wu F *et al*. [9]	Multicentric	43	F	5.6	No	60
2019	Ju WT *et al*. [10]	Oral cavity	41	F	1.6	No	27
		Oral cavity	61	M	0.5	No	25
		Multicentric	46	F	3.7	No	56
		DST	62	F	3.5	No	8
		Oral cavity	10	M	3.8	No	51
2009	Chu ZG *et al*. [11]	Multicentric	41	F	5.6	No	9
2008	Laga AC *et al*. [12]	Nasal	64	M	1.5	No	19
		Oral cavity	72	F	1	No	19
2010	Kuczkowski J *et al*. [13]	Salivary gland	65	M	1.4	No	24
2016	Prado-Calleros HM *et al*. [14]	DST	38	F	5	No	96
2010	Terada T [15]	SST	56	F	3	No	48
2012	Lee SK and Kwon SY [16]	SST	75	F	2	No	20
2013	Kim EK *et al*. [17]	SST	44	F	0.7	-	-
2018	Rubino S *et al*. [18]	Oral cavity	48	M	3.2	No	-
2007	Wilson T *et al*. [19]	Nasal	18	M	-	No	6
2009	Lee SK and Kwon SY [20]	DST	51	M	3.9	-	-
2011	Rho BH *et al*. [21]	SST	70	F	2	No	20
2018	Strayer E *et al*. [22]	Oral cavity	42	F	3	No	3
2013	Chotey NA *et al*. [23]	SST	18	F	1.2	No	6
2009	Sapelli S *et al*. [24]	Oral cavity	28	M	1.5	No	36
2015	Mathew NK *et al*. [25]	Mandible	12	M	4.5	No	24
2014	Bates AS *et al*. [26]	Salivary gland	66	M	1.2	No	18
2013	Akbulut S *et al*. [27]	Oral cavity	61	F	2	No	18
2006	Dray MS *et al*. [28]	SST	35	F	1	-	-
2006	Mentzel T *et al*. [29]	SST	17	M	-	-	-
		Deep soft tissues	22	M	-	-	-
		SST	47	M	-	-	-
		SST	53	M	-	-	-
2007	Datta V *et al*. [30]	Oral cavity	36	F	0.5	-	-
2007	Ide F *et al*. [31]	Oral cavity	45	F	2	No	108
2008	Ide F *et al*. [32]	Oral cavity	50	M	1.3	-	-
		Oral cavity	67	M	1.2	-	-
2008	Maheshwari V *et al*. [33]	SST	42	M	1.5	-	-
2011	Lee YB *et al*. [34]	Nasal	68	F	0.5	-	-
2012	Bansal S *et al*. [35]	Oral cavity	40	F	5	No	6
2015	Vasenwala SM *et al*. [36]	Oral cavity	14	M	2	No	6
2018	Arden RL *et al*. [37]	Nasal	40	F	0.8	-	-
2019	Ralli M *et al*. [38]	Oral cavity	46	M	-	No	8
2020	Almeida LKY *et al*. [39]	Oral cavity	12	F	-	No	60

Legend: SST, superficial soft tissues; DST, deep soft tissues; F, female; M, male.

Discussion

As shown by our contemporary Literature review, MPC is confirmed to be a rare neoplasm that mostly shows a benign course. The rarity of this neoplasm is responsible for the lack of uniformity in its management from diagnosis to treatment, leaving the surgeon astonished when he accidentally steps into a diagnosis of MPC in his clinical practice.

Specifically for the head and neck region, MPCs are frequently described as superficial lesions involving the dermis or sub-cutis or occurring in the deep soft tissue, includ-

ing the oral cavity, tongue, nasal cavities, salivary glands, deep neck muscles and para-pharyngeal space. Independently of the site of its origin, symptoms are often non-specific: both large deep cervical [8, 9, 14, 15, 16, 17] and small superficial lesions [18] may present with local pain or swelling. Even from a histopathological point of view, MPC is reported back to be easily confused with other tumours originating from the pericyte cellular line [19]. According to the histopathological examination, MPC is characterized by the presence of perivascular myoid cells, sharing features of both smooth muscle cells and glomus cells and for these aspects, MPC is reported back to be easily misdiagnosed with other soft tissue tumours [28]. For this purpose, the immunohistochemical features have the utmost importance for a correct differential diagnosis, and the positive result of ASMA, Muscle-specific actin, vimentin and h-Caldesmon analysis will finally orientate towards MPC [9, 15].

In Literature, superficial and mucosal MPCs demand a careful inspection of all the head and neck districts since they share a multifocal pattern with the oral cavity being the most affected sub-site. Salivary gland MPCs are usually investigated with an ultrasound (US) examination, although, even in these cases, the radiologic characteristics are non-specific and rarely diagnostic [40]. In some reports, authors describe MPCs as well-demarcated, heterogeneous, markedly hypoechoic solid masses with a prominent colour signal to the power Doppler US [16, 20, 25, 26]. CT scan/MRI imaging is the most common preoperative investigation for deep MPCs; nonetheless, the radiologic features do not allow a differential diagnosis from other benign neoplasms, such as schwannomas, Castleman disease of hyaline vascular type, angioleiomyomas, solitary fibrous tumours and paragangliomas [16, 20]. Considering CT scans, a wide range of radiological presentations has been reported in Literature, describing MPC as a well-demarcated-sharply defined vascular lesion [16, 20] or a poorly defined mass [19], or even like a homogeneously enhancing mass [16, 20] with heterogeneous attenuation, peripheral enhancement and a central irregular non-enhancement region [8, 9, 19] without calcification or invasion of the adjacent structures [19]. According to MRI, MPCs are characterized by a low signal intensity on T1 and a high signal on T2, presenting as a heterogeneous lesion with a homogeneous contrast enhancement [27], surrounded by ectasic vascular structures [19]. In all described and investigated cases with a preoperative fine needle aspiration cytology (FNAC), none of the cytological reports was diagnostic [13, 14, 16, 19]. Similarly, fine needle aspiration biopsy (FNAB) was diagnostic only in 2 out of 5 cases [10, 16, 21].

Despite all the difficulties in MPC management, the first key point that emerged from the Literature review and was confirmed by our experience is that, when a single isolated lesion compatible with MPC is suspected, a radical excision should always be perceived. Thus, incisional biopsies [22, 23, 24] should be avoided, as they disrupt the integrity of the tumour [10], arguably paving the way to neoplastic field dissemination and the risk of multifocal MPC recurrences.

As known by the revision of Literature, most cases with a previous incomplete excision or a non-specified clearness of resection margins [6, 7, 8, 9] subsequently developed multicentric [6, 7, 8, 14, 19], multifocal [12, 13], or disseminated recurrences [4]. These findings support the trigger effect theory of surgical trauma (i.e., incisional biopsies) in the development of multiple lesions or tumour dissemination.

The second key point for mucosal MPCs is endoscopic nasosinusal surgery because it is a safe and effective technique to remove radically all malignant and benign diseases, reducing surgical morbidities compared to open surgery [41].

The present paper reports the first case of a multifocal intranasal MPC which was radically treated, and the patient was free from disease during the last follow-up visit (thirty-six months after the treatment). Particularly, the patient presented multiple and small lesions in the nasal fossa. In only six papers, authors described multicentric foci of MPCs [6, 7, 8, 9, 10, 11, 14], while multifocal localizations of MPC (nose pyramid and parotid) [12, 13] were reported in only two cases.

Finally, even in our case, endoscopic nasal surgery was confirmed to be effective in the treatment of multifocal nasal MPCs.

We know this is a simple case report describing our experience of managing sinonasal multifocal lesions, which is insufficient to define a recognized consensus about the diagnosis and treatment of this kind of disease. However, this first work underlines the feasibility and usefulness of the endoscopic approach for nasal MPCs and its capabilities to ensure a safe diagnosis and radical removal, even in multifocal nasal MPCs.

Conclusions

MPC is a rare and challenging disease that must be considered by the otolaryngologist in the differential diagnosis of all the vascular lesions of the head and neck region. When biopsies reveal a histopathological diagnosis, the endoscopic naso-sinus surgery performed with a radical intent (clear resection margins) is an effective approach for single and multifocal MPCs, reducing the risk of tumour dissemination and local or distant relapses.

Availability of Data and Materials

The data used and analyzed during the current study are available from the corresponding authors.

Author Contributions

PC, MA designed the research study. FB and FC analyzed the data and drafted the manuscript. RDB interpreted the data and revised the manuscript. LMS, GP and MT car-

ried out literature review and made acquisition of data. FM made acquisition of data, provided the histopathological analysis and the iconographic materials. All authors revised the manuscript critically for important intellectual content. All authors read and approved the final manuscript. All authors have participated sufficiently in the work and agreed to be accountable for all aspects of the work.

Ethics Approval and Consent to Participate

The patient released a written informed consensus for the use of data for scientific purposes. The study was approved by the Institutional Ethical Committee (European Institute of Oncology IRCCS) (IEO code 1615) and complied with the principles stated in the Declaration of Helsinki "Ethical Principles for Medical Research Involving "Human Subject".

Acknowledgment

The authors thank Prof. Linda De Berardinis for assistance with the English text.

Funding

This research received no external funding.

Conflict of Interest

The authors declare no conflict of interest.

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