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Complex abdominal wall reconstruction after oncologic resection in a sequela of giant omphalocele: A case report

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ABSTRACT

INTRODUCTION: Trauma injuries and oncologic resection are common aetiologies of complex abdominal wall defect. Reconstruction of abdominal wall is an everlasting question for general, paediatric and reconstructive surgeons. The plethora of techniques, bioprosthetic and engineered tissues offer countless possibilities.

PRESENTATION OF CASE: The patient was a 28 years old woman, with past history of untreated giant liver omphalocele, admitted for a suspicious hepatic tumefaction without specific clinical signs. The thoraco-abdominopelvic CT scan revealed lung metastasis and a bilobed left hepatic tumour. Pre-operative cytologic findings of mild differentiated hepatocellular carcinoma differed from the post-operative findings of hepatoblastoma. The full-thickness abdominal wall defect after a radical resection was reconstructed with a combined acellular dermal matrix, NPWT and skin graft solution. A total epithelization was obtained after 8 weeks follow-up.

DISCUSSION: Hepatoblastoma in adult is rare, with no consensus. A radical resection in context of giant untreated omphalocele is an unusual challenge for the surgical team. The pre-operative evaluation, the defect classification and the general conditions of the patient are paramount steps for an appropriate reconstruction. Primary or delayed reconstruction with myocutaneous flap as gold standard, depends on the oncologic management and anticipated post-operative complications. Acellular dermal matrix used for a bridged fascial repair directly on viscera and covered by NPWT, favoured a healthy granulation tissue. The full-thickness defect was then reconstructed with an ADM, NPWT and skin graft instead of an association with the myocutaneous flap. The patient follow-up was emphasized in the hepatoblastoma, but the complications of this reconstruction strategy are unknown. A total epithelization was obtained, the abdominal bulge or hernia is the first complication under surveillance.

CONCLUSION: Delayed reconstruction after an oncologic large abdominal wall resection has the advantage to manage post-operative complications and prepare alternative solutions. Acellular dermal matrix was not first designed for skin tissue regeneration, some authors as us experimented the conclusion that this matrix could be used for permanent abdominal wall reconstruction.

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Abbreviations: ADM, Acellular dermal matrix; ALD, flap autologous latissimus dorsi flap; ALT, flap antero lateral thigh flap; AWD, abdominal wall defect; BWS, Beckwith-Wiedemann syndrome; CAWD, complex abdominal wall defect; CT, scan computed tomography; GO, giant omphalocele; HIV, human immunodeficiency virus; HBV, hepatitis B virus; HCV, hepatitis C virus; ISOWATS PL, (Identification and diagnosis of postoperative fistula, Sepsis and Source Control, Optimization of Nutrition, Providing and Ensuring Wound Care, Redefining the Anatomy and understanding the pathology at hand, Timing of definitive surgery and/or takedown of fistulas, Definitive Surgery and surgical creativity, Postoperative care, Long-term follow-up); MDT, Multidisciplinary Team; NPWT, Negative Pressure Wound Therapy; TDAP, flap thoraco dorsal artery perforator flap; TFL, flap tensor fascia lata flap; US, ultra sound.

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1. Introduction

Anterior abdominal wall reconstruction these last decades became a full-fledged entity, leading to creation of specialized abdominal wall reconstruction units. Full-thickness abdominal wall defect is observed in traumatic injuries, oncologic resection, omphalocele and laparochisis. Omphalocele is one of the common aetiologies of congenital abdominal wall defect, with a prevalence of 1 in 4000–6000 birth [1]. It is characterized by an umbilical ring herniation of abdominal viscera covered by a multi-layered membrane which includes peritoneum, Wharton's jelly and amnion, with or without syndromic malformation association. Management of omphalocele is multidisciplinary, based on multiple factors as the defect size, the herniation of the liver or not, syndromic mal-

formation association (Beckwith-Wiedemann syndrome, severe pulmonary hypoplasia) [2]. Giant omphalocele is rare, with a staged surgical repair recommendation and many techniques described. The long-term follow up of the GO repair revealed aesthetic and functional sequelae [3].

We report in line with the SCARE guidelines [4] a case of sequela of giant omphalocele with a hepatic tumour, highlighting the oncologic and reconstructive surgical experience with combined solutions through flap reconstruction, acellular dermal matrix, skin graft, and Negative Wound Pressure Therapy (NWPT).

2. Case presentation

The present case is a 28 years old north-African woman, with a poly malformative syndrome including horseshoe kidneys and a congenital anterior abdominal wall defect type omphalocele, non-smoking, BMI 19, negative HIV, HBV and HCV serologies. The type of omphalocele, its clinal history and therapeutic care were unavailable in the medical transfer report. The patient was transferred in the hepatobiliary surgery service for a suspicious hepatic tumour evolving during the past year.

The patient did not complaint about any specific symptoms, and did not present any digestive or respiratory signs. At the physical



Fig. 1. Picture of the tumefaction.

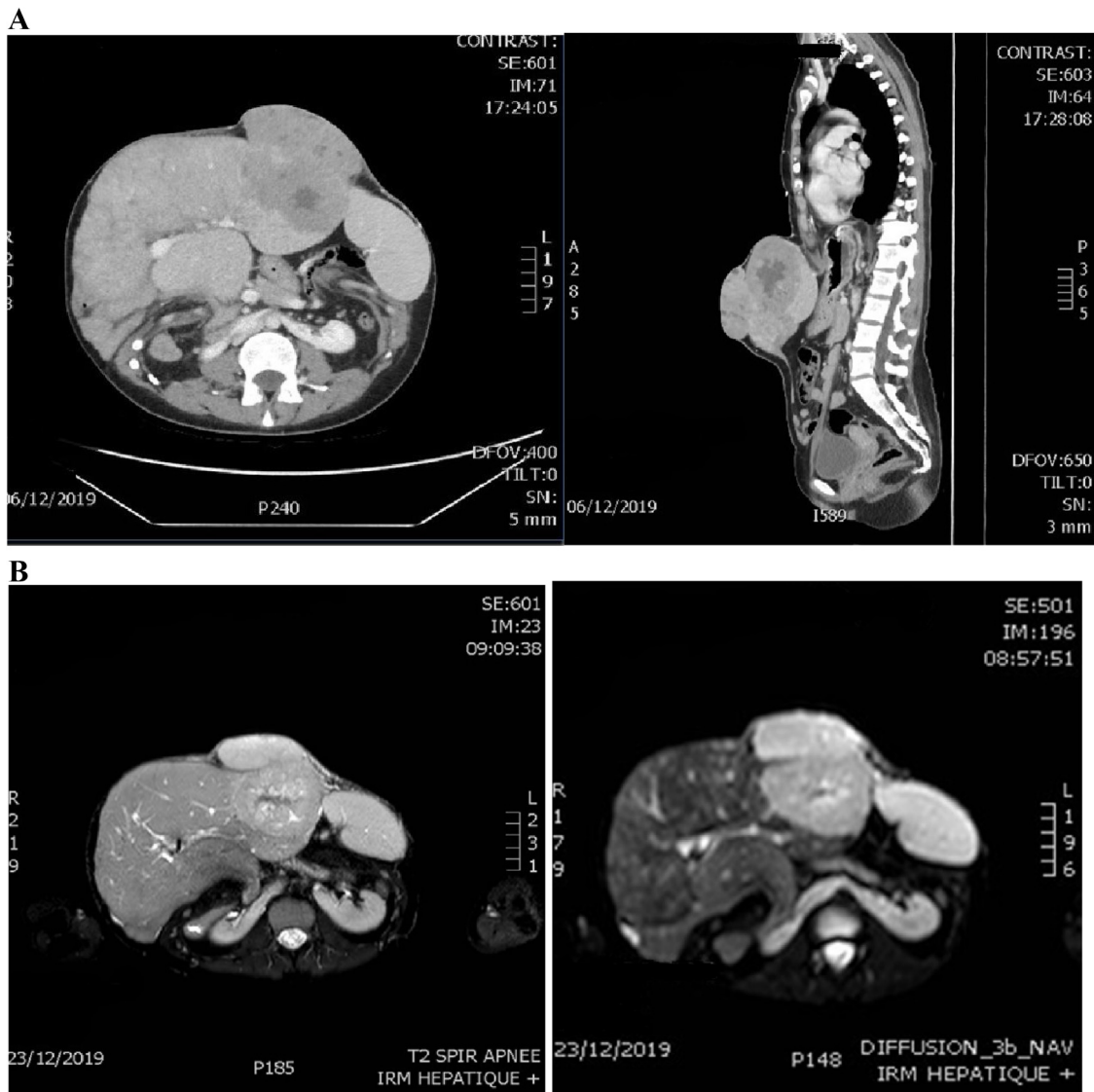


Fig. 2. a. The Thoraco abdominopelvic contrast CT scan.

b. The Hepatic MRI.

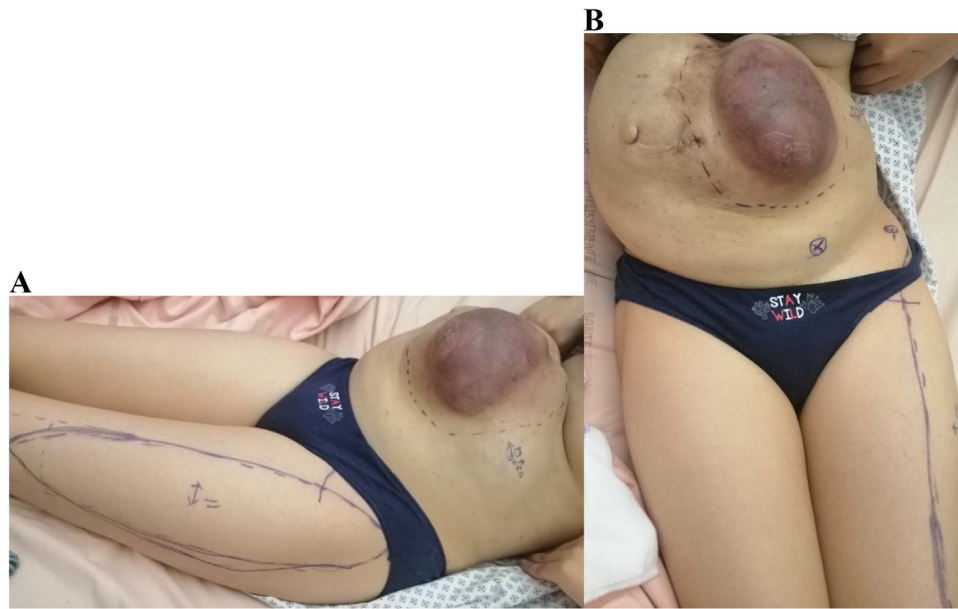


Fig. 3. a. Tensor Fascia Lata Flap ; b marks of DIEP and Circumflex artery.

examination, there was a midline abdominal scar, a 19 cm * 20 cm purplish tumefaction localized at the left hypochondrium and epigastric area, the umbilicus deflected on the right. On palpation, a splenomegaly was found, the tumefaction was painful, non-impulsive, irreducible, heterogenous, deeply fixed, and originate from the liver. (Fig. 1: Picture of the tumefaction) To complete her clinical examination, biological investigations and medical imaging were performed.

The thoraco-abdominopelvic contrast CT scan and hepatic MRI revealed: a 6 mm hilar lymph node and a 4 mm nodule at the superior lobe of the left pulmonary parenchyma, a second 4 mm nodule at the superior lobe of the right pulmonary parenchyma, a congenital abdominal wall defect type giant omphalocele with liver, spleen, pancreas, small bowel herniation, horseshoe kidneys, a voluminous bilobed heterogenous hepatic tumor with central necrotic tissue of 103*85*106 mm localized at the left hepatic lobe enhanced by the contrast and without vascular invasion. (Fig. 2a: The Thoraco abdominopelvic contrast CT scan) (Fig. 2b: The Hepatic MRI) Two weeks later, the anatomopathological findings from a fine-needle hepatic biopsy concluded to a mild differentiated hepatocellular carcinoma.

The oncologic Multidisciplinary Team (MDT) Meeting with oncologist, hepatobiliary surgeon and reconstructive surgeon recommended a R0 left-lobectomy 'en-bloc' resection with the cutaneous tissue and an abdominal wall reconstruction. The surgical team presented two different approaches. The primary closure which included the left lobectomy and abdominal wall reconstruction. The staged closure which included a first time with the left lobectomy and a temporary abdominal closure with a mesh and Negative Pressure Wound Therapy (NPWT), and a second time for the abdominal wall reconstruction.

After a second clinical re-evaluation and medical imaging (abdominal CT scan and Ultrasound (US) doppler of the superficial, deep inferior epigastric and circumflex femoral arteries, two different strategies for the abdominal wall reconstruction were proposed to the patient. The first one was a Tensor Fascia Lata (TFL) flap, as a pedicled or free flap and the second one was an Autologous Latissimus Dorsi (ALD) free flap. (Fig. 3a: Tensor Fascia Lata Flap) (Fig. 3b: marks of DIEP and Circumflex artery)

The staged closure approach was decided based on the risk of losing the flap if there was any complication after the hepatectomy

(haemorrhage, infection, bilioma). Ten days after the MDT recommendation, the left lobectomy was performed with the 'en-bloc' resection including the cutaneous tissue and achieving clear margin superior to 3 cm. (Fig. 4a: Tumour extended 22*16*14.5 with skin flap and 22*16 and hepatic mass 10.5*13.5*7 cm) (Fig. 4b: Picture of the abdominal wall defect after oncologic resection.) The temporary abdominal wall closure was done with a porcine non-crossed-linked, collagen matrix (acellular dermal matrix ADM) mesh CELLIS[®], fixed with a Vicryl[®] 2 interrupted sutures on the sub-cutaneous tissue and recovered with a Negative Pressure Wound Therapy (NPWT) at -80 mmHg. (Fig. 4c: Abdominal closure with the mesh 20*30 cm * 4 mm) (Fig. 4d: NPWT) (Fig. 4e: Dress wounding after 2 weeks of NPWT)

The only post hepatic surgery complication was a bilioma confirmed by the thoraco- abdominopelvic contrast CT scan control after two weeks. After three weeks of NPWT and parenteral nutrition, the wound healing was at a proliferative phase, covering the entire mesh. This result introduced the possibility of a skin graft. A split-thickness skin graft was realized with a dermatome, the antero lateral part of the right thigh was the donor site, and recovered by NPWT. The wound dressing was followed-up during 8 weeks until the total epithelization. (Fig. 5a: Skin graft) (Fig. 5b: First wound dressing 5 days after skin-graft) (Fig. 5c: 2 weeks – dress wounding)

A second MDT meeting after the new anatomopathological findings of a mixed hepatoblastoma with microscopic clear margin of 0.45 mm and lung metastasis recommended a fine-needle cytopunction of the lung lymph node and a chemotherapy.

3. Discussion

Congenital anterior abdominal wall defects (AWD) are rare, with an estimated prevalence of 5.49 per 10 000 birth in Europe [5]. Omphalocele or exomphalos and gastroschisis are the most common congenital abdominal wall defects, with a prenatal diagnostic at 11–14 weeks of gestation [6]. She presented an omphalocele containing the liver, pancreas, spleen, a part of the stomach and small intestine, defined as a giant omphalocele. Her past therapeutic care is not well-known, but her midline scar suggested a first step for an abdominal wall reconstruction. Several syndromes are associated with omphalocele. Beckwith-Wiedemann

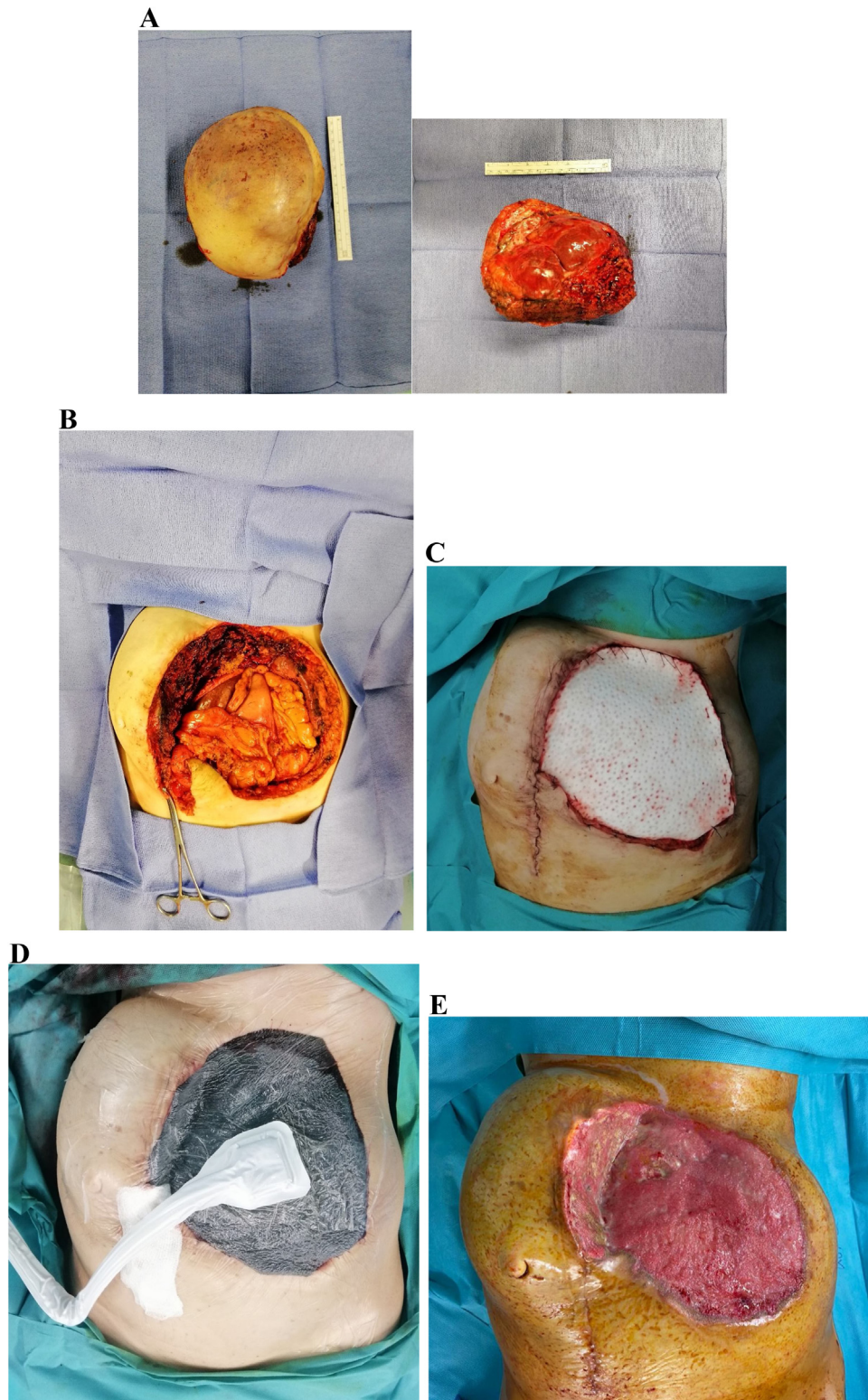


Fig. 4. a. Tumour extended 22*16*14.5 with skin flap and 22*16 and hepatic mass 10.5*13.5*7 cm.
 b. Picture of the abdominal wall defect after oncologic resection.
 c. Abdominal closure with the mesh 20*30 cm * 4 mm.
 d. NPWT.
 e. Dress wounding after 2 weeks of NPWT.

syndrome (BWS) is the most common, with a polymorph clinical presentation and high risk of embryonal tumours like hepatoblastoma, neuroblastoma or Wilms tumour [7]. However, isolate omphaloceles are more common, a genetic diagnosis for BWS can

be recommended in her case according to some expert consensus [8]. Adult hepatoblastoma is very rare, 50 cases described in literature, and aggressive tumour, without no therapeutic consensus [9].

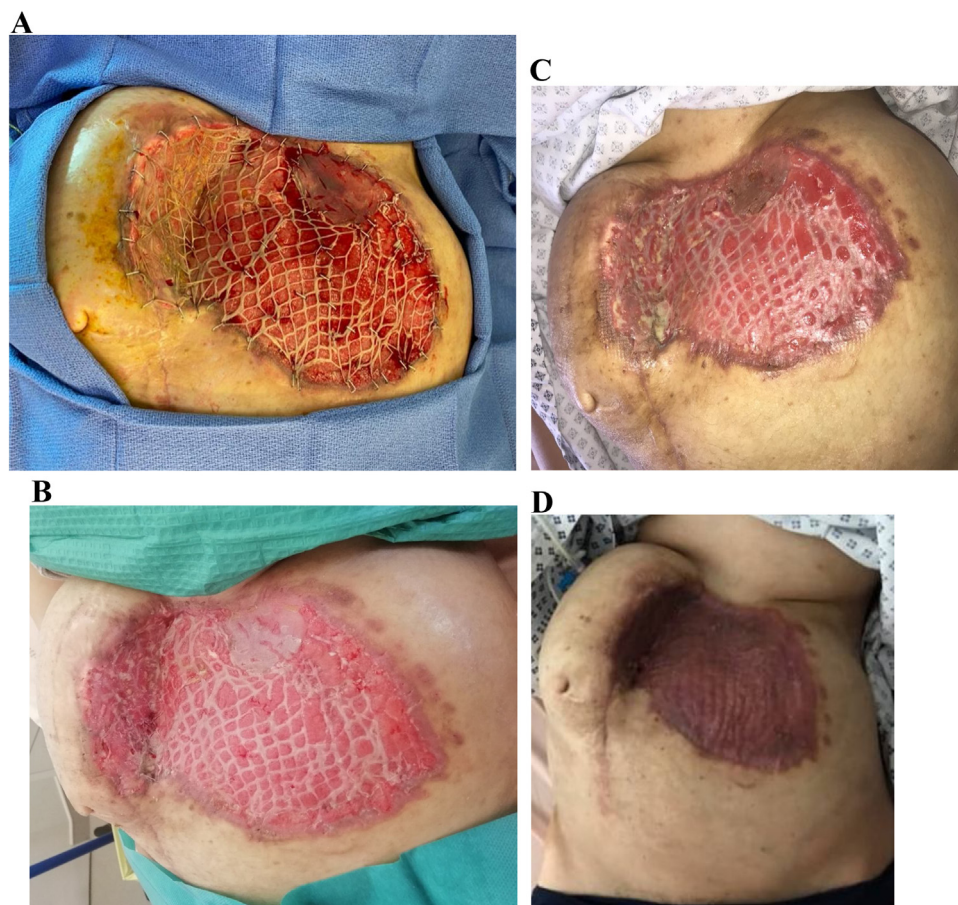


Fig. 5. a. Skin graft. b. First wound dressing 5 days after skin-graft. c. 2 weeks – dress wounding. d. Total epithelialization.

The particularities of this case are:

- An untreated sequelae of giant congenital liver anterior abdominal wall defect in adulthood which is very rare [10].
- A Full-thickness abdominal wall defect after radical ‘en-bloc’ oncologic resection of hepatoblastoma extended to the skin in adult, with lung metastasis.
- The management of a complex abdominal wall reconstruction with a complicated post-operative period.

Complex abdominal wall reconstruction is a challenge for both digestive and reconstructive surgeons. The reconstructive project must consider the localization, type, depth and size of the defect, the presence of contamination, the type of tumour as well as the general conditions of the patient [11].

Three main objectives must be achieved during a complex abdominal reconstruction: restore the abdominal wall integrity and functionality, protect the intra-abdominal viscera, prevent ventral hernia [12]. This should integrate the oncologic difficulties by managing bacterial contamination, radiated soft-tissue injury, resecting clean margins which can lead to a full-thickness abdominal wall defect. Many classifications of AWD have been proposed, which help the surgical team to have a holistic approach and advanced reconstructive techniques [11]. According to the Anderson classification [12], the patient had a type V any defect involving two or more types (abdominal localization), subtype C skin, subcutaneous tissue and any component of the musculofascial abdominal wall. (Fig. 6: M.D Anderson oncologic abdominal wall reconstruction classification system.)

The pre-operative period is a paramount step for the management of a CAWD, helpful to reduce the intra and post-operative morbidity and mortality. In this case the reconstructive challenge was double with the congenital abdominal wall defect and the large full-thickness defect after the oncologic resection. The nine-steps ISOWATS PL strategy is a disciplined protocol, with a CAWD mortality of 10% [13]. Our approach during the MDT and surgical meeting reviewed many items of this protocol.

The first question was the timing of closure, primary or staged. Post hepatic surgery has an overall morbidity from 4.1% to 47.7%, involving several intra and post-operative complications [14]. With a second time for AWR lengthening the intervention time, it could lead to a septic situation, failure of the flap. Early flap coverage is beneficial after a full thickness oncologic resection, offering a primary and definitive closure, reducing the local tissue injury and pro-inflammatory response [15]. Considering all these parameters, we recommended a delayed staged reconstruction, to have a life-boat for a re-intervention.

The second question was the full-thickness abdominal wall defect in this context of sequelae omphalocele. Clinically omphaloceles are classified as small, giant or ruptured. There is no consensus for giant omphalocele (GO) definition, but a defect > 5 cm, containing the liver or other viscera, and a disproportion between the abdominal viscera and abdominal cavity is considered as GO [1]. The indication of omphalocele management is based mainly on this clinical classification and malformation association [2] (Table 1).

The fascia repair ideally must be dynamic using the component separation technique, gold standard in absence of abdominis rectus [16,17], which was inappropriate in this case. A bridged repair mesh was indicated here instead of its greater rate of ventral her-

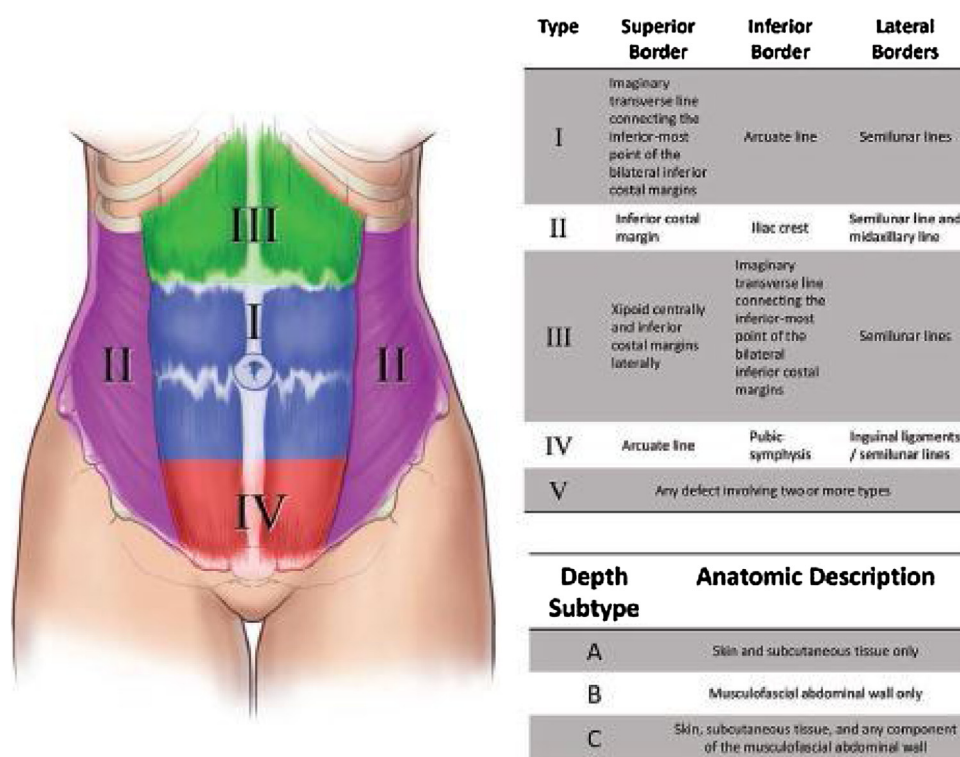


Fig. 6. M.D Anderson oncologic abdominal wall reconstruction classification system [9].

Table 1
Indication in omphalocele management [2].

Indication	Type of omphalocele	Non-surgical techniques	Surgical techniques
Immediate repair	<ul style="list-style-type: none"> • Small omphalocele • Giant omphalocele without syndromic malformation 	Topical medication: <ul style="list-style-type: none"> • Silver sulfadiazine • povidone-iodine (Betadine) • 70% alcohol solution • 2% merbromin (Mercurochrome) solution • Silver nitrate. 	<ul style="list-style-type: none"> • Umbilical cord ligation • Fascial closure • Primary midline fascial closure with umbilical cord preservation • Skin flap – umbilicoplasty • Dermal advancement sutures
Staged repair	<ul style="list-style-type: none"> • Giant omphalocele with or with liver and viscera herniation • Failure of immediate repair • Sequelae of primary omphalocele repair 	Topical medication: <ul style="list-style-type: none"> • Silver-based • Iodine-based • Manuka honey • 2% aqueous eosin • Negative Pressure Wound Therapy (NPWT) [1] 	Gradual viscera reduction: <ul style="list-style-type: none"> • Gross technique • Schuster technique • Sequential sac ligation Tissue expanders (intraabdominal, intramuscular, subcutaneous) Fascial closure techniques: <ul style="list-style-type: none"> • Simple midline fascial closure • Component separation • Mesh ideally bioprosthetic • Skin Flap closure Other: <ul style="list-style-type: none"> • Multilayered flap technique • Turn over flap with prosthetic mesh [3] • Approximation rectus abdominis [3]

nia than fascial approximation with mesh reinforcement [11]. After the oncologic resection, the defect was a full-thickness abdominal wall defect of 23*20 cm, with residual fascia. For large defect bioprosthetic human or porcine acellular dermal matrix are recommended. The non-crossed-linked porcine ADM presents multiples advantages like improving cellular infiltration and neovascularization, reducing bacterial counts, maintaining tensile strength and conservative treatment in case of infection or exposure [18]. The bilioma was the only post-operative complication, without secondary infection and drained. The combination of porcine ADM and NPWT in this case led to a wound bed with a healthy granula-

tion tissue over the mesh, allowing a split-thickness graft, with a low morbidity [11]. The acellular bilayer matrices as Integra®, featured a synthetic epidermal layer are primary designed as complete skin substitute instead of ADMs [19]. Providing specific host cells to ADM as scaffold will lead to a tissue regeneration, as observed in this case with the ADM-skin graft combination allowing skin cells colonization with a dermal bed [20].

The third question was a skin graft vs an autologous flap as definitive reconstruction. It was compulsory for this case to manage the aggressivity of the cancer which needed an adjuvant chemotherapy and be aware of an oncologic recurrence as the

Table 2
Flap reconstruction for abdominal wall reconstruction indication [15].

	Local	Pedicled	Free
Epigastric	Transposition IM, IC, SE Keystone	Rectus Omentum	Thigh-based (ALT, AMT, VL, TFL, RF, STF) Back-based (LD, TAP, Scap/Para)
Periumbilical	Bipedicled Fasciocutaneous Transposition DIEP, SIEP, TLP Keystone	Rectus Omentum	Thigh-based (ALT, AMT, VL, TFL, RF, STF) Back-based (LD, TAP, Scap/Para)
Hypogastric	Bipedicled Fasciocutaneous Transposition DIEP, SIEP, TLP Keystone	Thigh-based Rectus Omentum	Thigh-based (ALT, AMT, VL, TFL, RF, STF) Back-based (LD, TAP, Scap/Para)
	Bipedicled Fasciocutaneous	Thigh-based	

Abbreviations: ALT, anterolateral thigh flap; AMT, anteromedial thigh flap; IC, intercostal epigastric artery perforator flap; IM, internal mammary artery perforator flap; LD, latissimus dorsi flap; RF, rectus femoris flap; Scap/Para, scapular/parascapular flap; SE, superior epigastric artery perforator flap; STF, subtotal thigh flap; TAP, thoracodorsal artery perforator flap; TFL, tensor fascial lata flap; VL, vastus lateralis flap, DIEP, deep inferior artery perforator flap, SIEP, superficial inferior epigastric artery perforator flap, TLP, thoracolumbar perforator flap.

resection margin was less than 1 cm [21], and there was the presence of lung metastasis. Sparing the flap option, reduced the risk to lose it in case of recidivism and preserved it for a definitive reconstruction after oncologic adjuvant treatment. Skin graft allowed the observation of local oncologic recurrence as the margin were uncertain [11], and the possibility of reoperation through the ADM mesh [19].

The autologous flap was the life-boat reconstructive option. Thigh-based flaps Tensor Fascia Lata (TFL), Antero Lateral Thigh (ALT) and Back-based flaps Autologous Latissimus Dorsi (ALD), Thoracodorsal Artery Perforator (TDAP) are well indicated for complex abdominal wall reconstruction [15] (Table 2).

All of these flaps can be harvested for a myocutaneous flap, indicated in case of large full-thickness defect. The TFL flap in this case was proposed as a pedicled flap in pre-operative, for the proximity with the AWD, which allows a well-tolerated rotation and extension to the area to be covered, and preserves the own vascularization of the flap [22]. But we were in front of a massive abdominal defect after the resection. The free flaps with the TFL [23], ALT Thigh Flap [24] and ALD flap [25], were suggested for the definitive reconstruction.

We combined the bridged fascial mesh repair with NPWT and skin graft for a temporary complex abdominal wall closure after oncologic resection. Due to the aggressivity of the hepatoblastoma, the temporary solution became a permanent solution. Some case reports proposed this combination synthetic or biologic fascial mesh repair and skin graft for definitive abdominal wall reconstruction [26,27].

4. Conclusion

The acellular dermal matrix can be placed directly over a viscera and used as both temporary and permanent wound dressing in AWR [19]. This case was challenging for both digestive and reconstructive surgeons. Complex abdominal wall defects are singular by the past history of the patient (sequelae of a severe congenital abdominal wall defect), the general conditions, the aetiology (traumatic, oncologic resection), and the type (localization, size, depth) of AWD. Reconstruction of complex abdominal wall defect must consider all of these particularities, the surgeons experience and the combined solution with bioengineered tissue [20].

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Ethical approval

The ethical committee of the hospital gave the agreement to report this case.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

- Antonio Ianneli: Surgery, writing and proofreading.
- Lionel Sebastianelli: Surgery, writing and proofreading.
- Frederica Jessie Tchoungui Ritz: Surgery, writing and proofreading.
- Marie Anne Poumellec: Surgery, writing and proofreading.
- Alexandra Maertens: Surgery and proofreading.
- Olivier Camuzard: proofreading.
- Thierry Balaguer: surgery and proofreading.
- All authors have revised and agreed to the submission of this manuscript.

Registration of research studies

Not applicable.

Guarantor

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Declaration of Competing Interest

The authors report no declarations of interest.

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