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Case Report of a Patient with Swyer-James-MacLeod Syndrome Undergoing Breast Surgery under Regional Anaesthesia and Review of Literature

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Abstract

Swyer-James-MacLeod Syndrome is a rare acquired pulmonary disorder that develops secondary to infectious etiologies in early childhood. Patients who are affected have the potential of developing perioperative respiratory complications. While regional anaesthetic techniques are often performed as adjuncts to general anaesthesia, there is less data on breast operations being done solely under regional anaesthesia. We herein describe a patient with Swyer-James-MacLeod Syndrome who underwent breast lesion wide excision under combined paravertebral and pectoral nerves block, supplemented with propofol infusion for sedation. Choice of blocks was decided upon based on knowledge on the anatomy. Sole regional anaesthetic techniques have been the safest approach in some circumstances and should always be considered in patients who are of high risk under general anaesthesia.

Keywords

Breast Surgery, Regional Anaesthesia, Paravertebral Nerve Block, Pecs Block, Swyer-James-MacLeod Syndrome

1. Introduction

Swyer-James-MacLeod Syndrome (SJMS) is a rare, acquired post-infectious condition characterised by emphysematous changes or bronchiectasis which may lead to fibrosis to cause pulmonary capillary bed destruction. Patients with severe lung disease presenting for anaesthesia are a challenge because intraoperative and postoperative respiratory complications are more common in them, and these may lead to prolonged hospital stay and increased mortality. Regional

anaesthetic techniques are often used adjunctively for breast surgeries under general anaesthesia (GA), but there are limited reports on breast surgeries done solely under regional anaesthesia (RA). We describe a patient with SJMS who successfully underwent breast surgery under regional anaesthesia.

Written consent was obtained from the patient for publication of this case report.

2. Case Report

The patient is a 51-year-old female with a history of Swyer-James-MacLeod Syndrome (SJMS) who required stereotactic hook-wire localization, wide excision of left breast micro calcifications. Her only surgical history was an uneventful laparoscopic bilateral salpingo-oophorectomy earlier in the same year performed under combined spinal-epidural, for which she required postoperative intensive care monitoring. Although she was NYHA class 2, her recent lung function test was grossly abnormal: FEV1 1.04 L (46% predicted), FVC 1.97 L (76% predicted) and FEV/FVC ratio 53%. Her chest X-ray showed changes congruent with her diagnosis: hypoplastic, bronchiectatic left lung and bronchial wall thickening in the right lung (Figure 1).

She was planned for surgery under regional anaesthesia. Single shot paravertebral, medial and lateral pectoral nerve blocks were performed under conscious sedation with intravenous midazolam 1mg and fentanyl 50 mcg. She was first positioned prone and left T3 and T4 paravertebral spaces were located with ultrasound (Figure 2 and Figure 3). Paravertebral block was performed with a 50 mm Pajunk Sono Plex Stim cannula and Sonosite Edge HFL50x transducer in

Figure 1. There are bronchiectatic changes in the left lower zone with some cystic lucencies and airway wall thickening. There is reduced volume of the left lung with relatively lucent appearance of the left hemithorax. Some bronchial wall thickening is also present in the right lung.

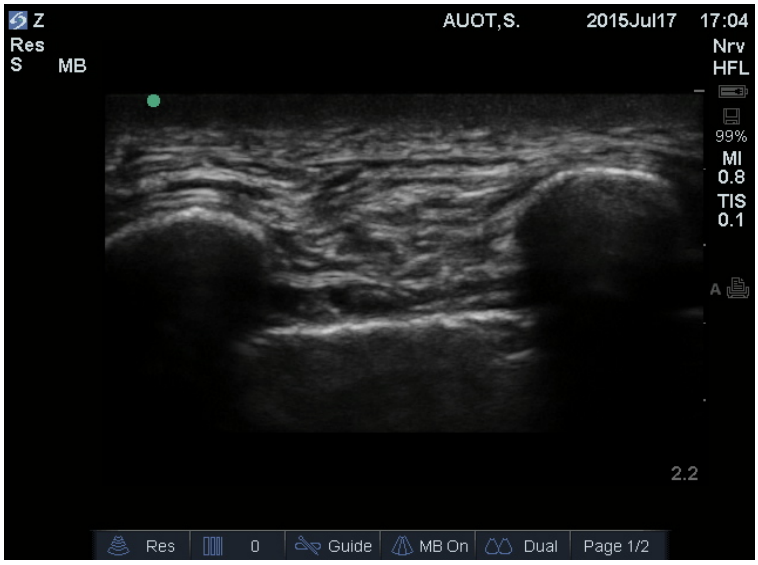

Figure 2. Ultrasound image of left paravertebral space with the transducer in the sagittal plane.

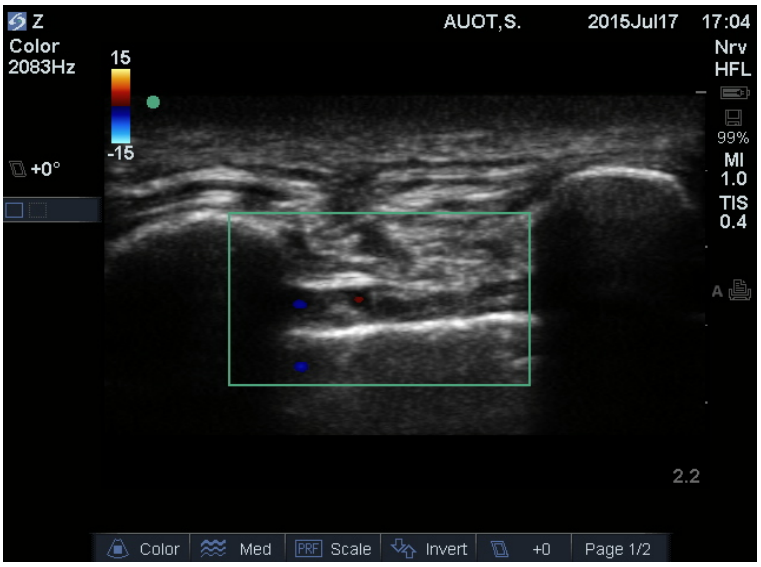

Figure 3. Ultrasound image of the same space, with frame indicating boundaries of colour Doppler applied, to reveal location of blood vessels before injection using out-of-plane technique.

the sagittal plane using out of plane technique. 100 mg lignocaine (5 mcg/ml adrenaline) and 50 mg ropivacaine in 10 ml were administered to each level. She was then positioned supine for medial and lateral pectoral nerve block with 8 ml of 200 mg lignocaine (5 mcg/ml adrenaline) under in-plane ultrasound guidance in the infraclavicular area.

Sedation was started at patient's request with propofol target controlled infusion, Schneider mode at effect site concentration of 1.8 ng/ml. A 3 cm radial skin incision was made 18 minutes after the completion of regional anaesthesia. Planned wide excision proceeded smoothly with excellent surgical condition.

Total surgical time was 20 minutes and patient recovered well enough for same-day discharge.

3. Discussion

Swyer-James-MacLeod Syndrome (SJMS), which was first described in 1953 by Swyer and James, and further detailed by MacLeod, is a rare acquired condition secondary to viral bronchiolitis and pneumonitis in childhood [1]. The condition manifests as a post-infectious bronchiolitis obliterans characterized by emphysematous changes or bronchiectasis which may lead to fibrosis in both bronchioles and interalveolar septum, causing pulmonary capillary bed destruction [2]. Patients' symptomology may range from asymptomatic with incidental diagnostic radiographical findings to severe symptoms, such as dyspnoea and recurrent pulmonary infections.

The anaesthetic considerations must be individualised, depending on the severity of lung involvement, comorbidities as well as type of surgical procedure. In severe cases, such as that in our patient, anaesthetic management parallels that of patients with obstructive lung disease [2]. Local anaesthesia, regional anaesthesia and central neuraxial anaesthesia should be used whenever possible. Care should be taken to avoid anaesthetising the phrenic nerve in patients with severe disease. As these patients are prone to baro- and volu-trauma, mechanical ventilation should be carefully set to avoid high airway pressures and large tidal volumes in cases when general anaesthesia cannot be avoided [3].

In view of her underlying SJMS, regional anaesthesia was performed to avoid potential postoperative respiratory complications. An incision was to be made at the 0700 position of the lower inner quadrant of the left breast. The combination of single shot paravertebral block at T3 and T4 levels, as well as medial and lateral pectoral nerve blocks was chosen based on the knowledge of the anatomy of the breast. The breast is mainly innervated by the anterolateral and anteromedial branches of the thoracic intercostal nerves corresponding to T3 - T5 and these are being targeted by the paravertebral blocks. Supraclavicular nerves from the lower fibers of the cervical plexus provide innervation to the upper and lateral portions of the breast. Researchers believe sensation to the nipple derives largely from the lateral cutaneous branch of T4 [4]. Medial and lateral pectoral nerve blocks, also known as Pecs I block, were administered to incorporate coverage to pectoralis muscle which lies deep to the breast tissue.

A brief literature search of the MEDLINE database via Pubmed was carried out to review the types of RA performed for breast surgery. Articles were included (**Table 1**) if RA was performed on patients undergoing breast surgery, published in English and full texts were available. RA performed included paravertebral blocks, Pecs block, serratus plane block, intercostal blocks, superficial cervical plexus block, complete ante-thoracic block, thoracic and cervical epidural anaesthesia, as well as transverse abdominis plane blocks for operations with transverse rectus abdominis myocutaneous (TRAM) flap reconstructions.

Table 1. Review of regional anaesthesia performed for breast surgery.

First Author (year)	Patient indication	Surgical indication	Type of block	Outcome	Complication
Our presented patient	SJMS with grossly abnormal lung function test	Left lower inner quadrant lumpectomy	PVB and Pecs I block	Excellent surgical condition. Discharged on the same day.	-
Moon EJ (2017) [5]	Refusal of GA	Left breast mass for Breast-Conserving Surgery with Axillary Clearance	Pecs I and II (in combination with sedation)	Analgesic effect lasted 8 hours No post-operative nausea and vomiting (PONV)	-
Versyck B. (2017) [6]	140 patients with breast cancer stage 1 - 3	Mastectomy or lumpectomy with sentinel node or axillary node dissection	Pecs II block or placebo block with saline	Pecs group had significantly less pain during stay in recovery area, required significantly less postoperative opioids	-
Hong B (2017) [7]	27-weeks parturient, with recurrent breast cancer, refused GA	Wide excision of breast mass 3 - 4 cm below the clavicle	Pecs II block and Pecto-intercostal fascial block	No intraoperative analgesics. Postoperative Visual Analogue Scale (VAS) score 1 but did not require further analgesics.	-
Abdallah FW (2017) [8]	Cohort study of 225 patients with breast cancer	Ambulatory breast cancer surgery	3 study groups of (75 per group): pectoralis, serratus block and conventional opioid-based analgesia	Pec and SPB associated with, reduced intraoperative fentanyl requirements, expedited recovery room discharge, reduced post-operative opioid use and PONV compared to control	-
Kim H (2017) [9]	Refusal of GA	Excision of breast giant fibroadenoma	Pecs I, Pecs II and internal intercostal plane block	No intraoperative opioid needed. No postoperative pain	-
Takahashi H (2017) [10]	72 years old, breast cancer at 1 o'clock position of the right breast	Modified radical mastectomy with sentinel node dissection	Complete ante-thoracic (medial, inferior, lateral) block, with sedation using propofol and remifentanil 0.1 mcg/kg/min	No postoperative analgesics.	-
Patel SY (2017) [11]	65 year old with ductal carcinoma-in-situ and mammary dysplasia, history of severe PONV	Unilateral mastectomy and TRAM flap reconstruction without axillary lymph node dissection	Pecs I and II, unilateral abdominal TAP block done before GA	NRS 4 - 5/10 on the side not blocked mild to moderate nausea 0 - 48 hrs RA lasted 24 hrs	-

Continued

Cata JP (2016) [12]	Retrospective study of 792 non-metastatic breast cancer to evaluate if use of PVB prolongs their survival	Mastectomy	198 given PVB, remainder treated with opioid based analgesia	Significantly lower intraoperative fentanyl consumption No significant change in recurrence-free survival	-
Hards M (2016) [13]	Retrospective study of 27 breast cancer patients	Mastectomy	16 had serratus block; 11 had wound infiltration	No patients with SPB had severe pain in recovery or POD 1 2 patients who had wound infiltration had severe pain in recovery, and 3 patients on POD1	-
Bonomi S (2016) [14]	Breast cancer patients	Implant-based breast reconstructions	Bilateral Pecs block, with GA	Eliminated need for post-operative opoid no PONV lower mean VAS score throughout inpatient stay	-
Fallatah S (2016) [15]	40 patients with unilateral breast cancer	Unilateral lumpectomy and axillary lymph nodes dissection	20 patients received peroperative unilateral PVB from T2-6; 20 patients received postoperative PCA morphine; both with GA	Significantly lower pain scores, shorter hospital stay, higher satisfaction score, and less PONV in patients who had PVB	2 patients who had PVB had inadvertent vascular puncture and one with pain at the site of injection that continued beyond a month
Wolf O (2016) [16]	RCT involving 74 breast cancer patients	Prosthetic breast reconstruction	PVB with GA or GA alone	Significantly less intraoperative and postoperative opioid consumption Less antiemetic medication	-
Khemka R (2016) [17]	2 patients with unilateral breast malignancy	breast conservation surgery with dissection of the axillary nodes followed by reconstruction using a LD pedicle flap	Pecs I block and serratus anterior plane block with GA	No intraoperative opioid. 1 had 6 mg morphine 0 - 48 hs. The other had 4 mg of morphine 0 - 48 hs	-
Fusco P (2016) [18]	78 year old severe respiratory insufficiency for pulmonary fibrosis	Radical mastectomy and axillary dissection	Serratus plane block, PECS I block, parasternal LA injections	Complete anaesthesia of mammary and axillary regions. No opioids first 24 hs of surgery	-

Continued

Shah A (2015) [19]	Retrospective review involving 132 patients	Implant-based breast reconstruction	Intercostal nerve blockade	Significant reduction in length of stay, consumption of intravenous morphine and increased cost-savings	-
Sundarathiti P (2015) [20]	RCT involving 70 breast cancer patients	Unilateral mastectomy surgery with axillary dissection	GA or PVB via catheter and injections at 3 different levels	Analgesic consumption, post-operative pain scores significantly greater in patients with GA	-
Kulkarni K (2013) [21]	40 ASA I and II patients with breast cancer	Radical mastectomies	Cervical epidural anaesthesia with either 10 mls of 0.25% bupivacaine or 10 mls of 0.375% ropivacaine	Similar onset of sensory block Significant increase in mean motor blockade score, time to achieve complete blockade and time to grade I motor recovery for patients who received bupivacaine	Respiratory distress in 2 patients requiring GA with intubation
Coopey SB (2013) [22]	Retrospective review of breast cancer patients	Mastectomies with immediate reconstructions	PVB	Significantly less mean length of stay, shorter mean time to conversion to oral narcotics, less incidence of PONV in PVB group	-
Li NL (2011) [23]	40 breast cancer patients	Unilateral breast surgery	GA or (GA and PVB)	Significantly lower pain scores at all time points, incidences of PONV, doses of postoperative analgesics and narcotics and higher Quality of Recovery scores in PVB group	-
Moller JF (2007) [24]	RCT involving 88 breast cancer patients	Breast tumour resection or mastectomy with lymph node biopsy	GA and paravertebral injections with either 0.5% ropivacaine or saline	Postoperative consumption of IV fentanyl and number of patients who reported pain >3 on NRS were significantly less in the Ropivacaine group	-
Singh AP (2006) [25]	50 ASA 1 or 2 breast cancer patients	Modified radical mastectomies	Cervical epidural anaesthesia with 10 mls of 1% lignocaine followed by 0.125% bupivacaine through epidural catheter	No clinically significant variations in perioperative pulse and respiratory rate No fall in mean arterial blood pressure during operation	One case where the procedure was terminated due to accidental dura puncture.
Exadaktylos AK (2006) [26]	Retrospective study of 129 patients with breast cancer	Mastectomy and axillary clearance	50 patients had PVB with GA; 79 patients had GA with morphine analgesia	Recurrence- and metastasis-free survival was significantly higher in the PVB patients	-

Continued

Kolawole IK (2006) [27]	1. 55 years old patient with recurrent left breast tumour with lung metastasis complicated by pleural effusion and pneumonia 2. 60 years old left breast mass with widespread lung metastasis	Palliative simple mastectomy	Intercostal nerves block at thoracic levels 2 - 7; infraclavicular approach to superficial cervical plexus; subcutaneous infiltration in midline to block contralateral intercostal fibres	1) pain-free for 5 hours 2) Uneventful postoperative course	-
Kairaluoma PM (2004) [28]	60 breast cancer patients	Breast resection or mastectomy with and without associated axillary dissection	GA and PVB with bupivacaine or saline	Significantly less post-operative opioid medication, less pain after 24 h, less PONV in the bupivacaine group	One patient had bilateral convulsions right after bupivacaine injection
Stamatiou G (2004) [29]	24 years old, 14 weeks of gestation, with left breast tumour	Wide local excision of tumour and lymph node dissection	T1-T7 thoracic paravertebral block and superficial cervical nerve block	Tolerated 65 min of operation under sedation with propofol infusion Total analgesic requirement was 12 mg morphine and 5 g paracetamol for the first 48 hs	-
Buckenmaier CC 3rd (2002) [30]	72 year old patient with HOCM, emphysema on home oxygen therapy	Left partial mastectomy with axillary dissection for infiltrating ductal carcinoma	Left paravertebral nerve blocks at thoracic levels 1 - 6; left superficial cervical plexus block	Pain free and opioid free on day of operation 4 mg of IV morphine on POD1 Discharged pain free subsequently	Hypotension, attribulated to sedation and possibly hypovolemia from fasting; resolved after fluid challenge
D'Ercole FJ (1999) [31]	38 year old, 29 weeks of gestation with a 10 cm left breast mass	Left modified radical mastectomy with axillary dissection	Left paravertebral nerve blocks at T1-T6	Tolerated procedure Oral non-opioid analgesia 18 h after neural blockade Discharged on POD2	-
Atanassoff PG (1994) [32]	RCT involving 48 ASA I and II patients	lumpectomy	3 groups of patients receiving intercostal nerve blockade of T3-6 unilaterally with either 1.5% or 2% lidocaine or 0.5% bupivacaine. 4th group was control who received GA	Significantly higher postoperative pain scores in control group. Number of women requiring post-operative analgesia, total amount of analgesics given during 24 hs post operatively were significantly lower in RA group	-

Several randomised controlled trials [15] [16] [20] [24] [28], comparing the use of PVB and GA to that of GA alone in patients undergoing breast surgeries, showed significant reduction in post-operative pain, post-operative opioid consumption and occurrence of post-operative nausea and vomiting (PONV) in patients receiving PVB. A recent randomised control trial conducted by Versyck B [6] comparing the more novel Pecs II block to placebo block in patients undergoing mastectomy or lumpectomy with axillary node dissection showed significant reduction in pain scores and postoperative opioid use. Other positive outcomes from the review include reduction in length of hospital stay (LOS), increased cost-savings and an increase in satisfaction score. On the other hand, complications were much less frequently described, signifying a favourable risk-benefit ratio of RA. Use of RA avoided post-operative respiratory complications, such as atelectasis, pneumonia and consequently, respiratory failure, associated with use of GA, especially in our patient with SJMS. It also had advantages over thoracic epidural in its ability to maintain haemodynamic stability, preserve lower limb motor power and hence, to allow for same-day discharge.

R. Blanco, who developed the Pecs I and II blocks, came up with the description of the serratus plane block (SPB), which blocks primarily the thoracic intercostal nerves and provides complete analgesia to the lateral part of the thorax [33]. A prospective observational study [34] conducted to compare paravertebral and serratus plane block in patients scheduled for non-reconstructive breast surgery showed no significant differences between the 2 groups in terms of quality of post-anaesthetic recovery, signifying that it could have been an alternative to the paravertebral block. When compared to PVB, Pecs block and SPB could eliminate the risk of posterior midline spread and subsequent hypotension. Pecs block and SPB may also be more advantageous for anticoagulated patients as according to ASRA's evidence-based guidelines, the same precautions should be taken when placing thoracic PVB as when placing an epidural [35]. However, similar to Pecs II block, SPB targets the lateral cutaneous branches of the intercostal nerves and may not sufficiently provide coverage to our patient whose tumour was in the lower inner quadrant of the breast.

Regional techniques have frequently been performed as an adjunct to GA, but there have been less data on it being given as the sole anaesthesia for breast surgery. The use of a thoracic paravertebral block at T1-3 levels with 3 ml solution of 0.75% ropivacaine at each level was described in a left quadrantectomy with axillary lymph node dissection for infiltrating ductal carcinoma in a patient who is at a late stage of amyotrophic lateral sclerosis. No other analgesia was given during the 110-min surgery and for 34 hours post-surgery [36]. Ultrasound-guided Pecs II block and internal intercostal plane block were given for excision of breast giant fibroadenoma in a patient who refused general anaesthesia. 10 and 20 mls of 0.375% ropivacaine was injected for Pecs I and Pecs II block respectively, followed by 10 mls of 0.375% ropivacaine injected for internal intercostal plane block, with propofol infusion given for sedation to the patient

who tolerated 1.5 hours of surgical time [9].

RA, either used alone or as an adjunct to GA, can also be recommended for well patients for the benefit it carries. Basic sciences studies and a retrospective review have suggested that breast cancer patients had better cancer outcomes if they received paravertebral blocks with GA compared to those who had received GA alone [26], although this benefit has yet to be shown in recent studies [11]. A multicentre double-blinded randomised trial (https://www.clinicaltrials.gov/, NCT00418457) is currently underway to determine the rate of cancer recurrence or metastasis in breast cancer patients receiving either RA or GA. Large majority of studies have shown that regional technique reduces intraoperative use of opioids and post-operative pain [16] [37] [38]. A randomised controlled trial [39] with PVB and placebo in patients undergoing mastectomies have not shown that use of RA to be associated with less incidence of chronic pain. There was, however, less severe chronic pain in patients who received PVB, and they too, experienced better physical and mental health-related quality of life compared to patients who received placebo injections. Other benefits of giving RA with or without GA include lower incidence of post-operative nausea and vomiting related to reduced usage of opioids [8] [40] [41], improved patient satisfaction score [39] and reduction in length of hospital stay [22] [40].

Regional anaesthesia has proven to be the safest approach in circumstances where GA would have carried a much higher risk. A patient with Eisenmenger's syndrome had surgical resection of a carotid body tumour done successfully under continuous cervical plexus block and remifentanil infusion, avoiding GA and its associated haemodynamic complications [42]. A supraclavicular block as a sole anaesthetic technique for an elbow incision and drainage procedure for one of a pair of adult craniopagus twins is another example where RA is more ideal than GA. They would otherwise be subjected to unpredictable effects of anaesthetic crossover and potential difficult airway due to anatomy differences. [43] Simultaneous bilateral below-knee amputations were performed under ultrasound-guided combined inguinal femoral and subgluteal sciatic nerve blocks in a coagulopathic patient with chronic ischaemic heart disease [44]. Left below-knee amputation, which took 76 mins, was performed under ultrasound-guided left femoral nerve block and left subgluteal sciatic nerve block using 21 ml and 24 ml of local anaesthetic mixture (containing 1:1 ratio of 1% mepivacaine and 0.75% ropivacaine) respectively. Subsequently, similar blocks using a total of 47 ml of the same local anaesthetic mixture was performed for the right below-knee amputation. Sequential placement of the blocks before corresponding limb surgery allowed successful anaesthesia without local anaesthesia toxicity, and at the same time, avoiding the high risk of GA in this patient.

Apart from the patient's medical condition and choice when considering method of anaesthesia, other practical issues include patient comfort and the ability to provide ideal operating conditions. RA can be combined with sedation to improve intra-operative patient comfort. However, gentle surgical manipula-

tion is required and this may not be well-received by all surgeons. Consequences of potential complications resulting from RA have to be taken into consideration. In this particular patient, it would have been disastrous if a pneumothorax were to result from PVB in the only functional lung.

4. Conclusion

Regional block with sedation is a feasible anaesthetic option for patients going for breast surgeries and this can reduce perioperative respiratory risks dramatically in patients with severe lung pathology. In addition, it may also be recommended to well patients as RA may improve other aspects of postoperative recovery, such as better pain control, lower incidence of PONV, shorter length of stay, which may all indirectly increase cost savings. However, there are other considerations such as surgeon's and patient's acceptability before making RA the default technique for breast operations.

Financial Disclosures

None.

Conflicts of Interest

None.

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