CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 81 (2021) 105718

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Laparoscopic insertion of pelvic tissue expander prior radiotherapy for sacral metastasis of alveolar maxillary rhabdomyosarcoma to prevent radiation enteritis

Andrej Omejc[a], Lucija Vegan[a], Mirko Omejc[b,*], Vaneja Velenik[c]

[a] Medical Faculty, University of Ljubljana, Slovenia
[b] Department of Abdominal Surgery, University Medical Centre Ljubljana, Zaloska 7, 1000 Ljubljana, Slovenia
[c] Institute of Oncology, Ljubljana, Slovenia

ARTICLE INFO

Article history:
Received 9 February 2021
Received in revised form 27 February 2021
Accepted 28 February 2021
Available online 3 March 2021

Keywords:
Rhabdomyosarcoma
Radiotherapy
Laparoscopic insertion
Tissue expander

ABSTRACT

INTRODUCTION: Rhabdomyosarcoma is the most common soft tissue sarcoma seen in childhood and adolescence. The most frequent sites are head and neck.
PRESENTATION OF CASE: A young female with maxillary rhabdomyosarcoma involving region of maxillary sinus with skeletal metastases was primary treated acccording to RMS 2005 protocol. She received 9 cycles of chemotherapy. Primary tumor of maxillary sinus was surgicaly removed after 4 cycles of chemotherapy, with 6th cycle of chemotherapy a radical radiotherapy of primary tumor location and metastasis in spinal vertebras, ribs, pelvic bone and left femoral bone started what leads to complete regression of skeletal metastases. In course of maintenance therapy MRI scan showed 12 × 28 × 23 mm lesion in sacrum in the vicinity of right sacroiliacal joint with caracteristics of metastasis. Because the region of right sacroiliacal joint with bowel was already included in primary radiation treatment, tissue expander was laparoscopicaly inserted in lower pelvis to displace bowel loops from radiation field to prevent radiation enteritis. After external beam radiotherapy to her sacrum, a good response without any side effects was achieved.
DISCUSSION: Laparoscopic insertion of pelvic tissue expander prior EBRT and it's subsequent removal after EBRT is safe and effective method for displacing loops of bowel out of the pelvis. With minimal morbidity converts untreatable disease to treatable by allowing delivering high doses of radiation to the patient.
CONCLUSION: After 2 years of follow up the disease is in remission and the patient without any major complaint.

1. Introduction

In childhood and adolescence rhabdomyosarcoma is regarded as the most common soft tisue sarcoma. It is most commonly found in the head and neck, accounting for 40 % of all cases. Other involved sites can be genitourinary tract, retroperitoneum, and rarely extremities. In this case report, prepared in line with the updated SCARE-criteria [1], we report of a young female with maxillary rhabdomyosarcoma with translocation of PAX3-FOXO1 involving region of maxillary sinus with bone metastases at the time of diagnosis.

2. Presentation of case

In September 2016, a 16-year-old female was diagnosed with metastatic alveolar rhabdomyosarcoma with translocation of PAX3-FOXO1 of the right maxillary sinus. Symptoms referred by the patient included stuffy nose, impairing speech started two months before. Later pain in the upper molar region, swelling in the region of maxillar sinus and disturbed vision joined. Diagnostic staging procedures (a computed tomography -CT scan of paranasal cavities and nuclear magnetic resonance-NMR) showed tumor located in the right maxillar sinus with the destruction of ethmoidal cells, right orbit, lower nasal shell and septum nasi. Positron emission tomography PET-CT revealed metastases in retropharyngeal, parapharygeal, subangular, right retromandibular lymph nodes and in tuberositas ossis ischii. NMR of pelvis showed multiple metastases in pelvis, both femurs, spine and in ribs. The patient was planned to nine blocks of chemotherapy (ChT) according to the RMS 2005 scheme (4xIVADo and 5xIVA) [2]. After 5 blocks of ChT a significant

* Corresponding author.
E-mail address: mirko.omejc@mf.uni-lj.si (M. Omejc).

https://doi.org/10.1016/j.ijscr.2021.105718

Fig. 1. Insertion of tissue expander.

regression of primary tumor was observed and she had surgical removal of primary tumor. As complete local removal of the tumor was not possible, external beam radiotherapy (EVRT) of surgical bed (50.4 Gy in daily fraction of 1.8 Gy) with 2 blocks of ChT were performed. Afterwards, all previously visible bone metastases were also irradiated (40 Gy in 20 fractions). The complete regression of all metastatic bone lesions was observed on PET-CT. In November 2017, during maintainance ChT, a 28 mm metastasis in right iliac bone near sacroiliacal joint and another smaller in right acetabulum were visible on PET-CT. She started with second line ChT (4xVIT) and EBRT of both metastases was planned. Small bowel was a dose limiting structure for reiradiation, so it needed to be excluded from radiation field. Because the region of right sacroiliacal joint with bowel was already included in primary radiation treatment in our patient, tissue expander was laparoscopicaly inserted in lower pelvis to displace bowel loops from radiation field to enable radiotherapy to the sacrum without developing radiation enteritis.

There was no bowel preparation required. A 10 mm supraumbilical incision was made and an open Hasson technique used to achieve pneumoperitoneum, with the placement of a 10 mm port at the umbilicus and 5 mm ports in both iliac fossa. Soft adhesions in lower pelvis were easily divided by scissored dissection. In lithotomy and steep Trendelenburg positioning, the suprapubic incision was dilated up to 20 mm. A MENTOR® Smooth Rectangle Tissue Expander (10.6 × 9.3 × 6.6 cm) 400 mL made of silicone with attached silicone tubing was then rolled tight, lubricated with water soluble lubricant and inserted via the suprapubic port and placed laparoscopically in the pelvis, leaving the normal-saline inflation port attached externally (Fig. 1).

A running dissolvable 2/0 polydioxanone (PDS) purse-string stitch was then sutured to the peritoneum of the sacral promontory, and anterior and side walls of the pelvis below the level of the common iliac vessels, and tied to keep the expander in the pelvis. With a Huber® needle inserted into the inflation port, the tissue expander was then filled with 340 mL of normal saline until the expander began to bulge against the retaining stitch (Fig. 2).

Gas was then released from abdominal cavity and the fascia of incisions closed. The connecting port of the expander was placed in a small subcutaneous space suprapubicaly and sutured to the fascia. Skin incisions were closed in the usual manner (Fig. 3).

Subsequent CT scan confirmed adequate placement of the expander in the lower pelvis with intestinal loops out of the planned radiation field (Fig. 4).

Her recovery after surgery was uneventful being discharged after two days. Metastases were irradiated to 40 Gy in 20 fractions using volumetric modulated arc therapy (VMAT) (Fig. 5).

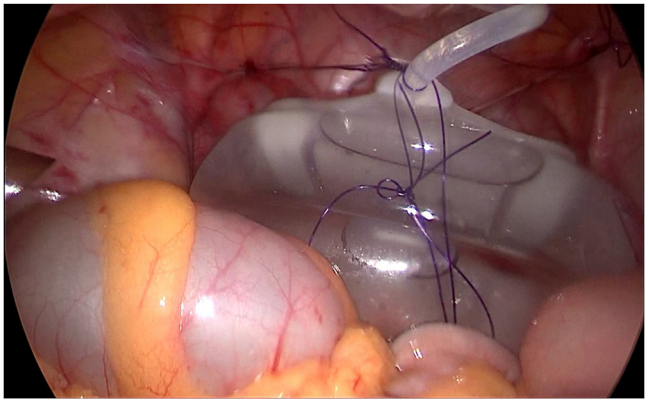

Fig. 2. Bulging of the expander.

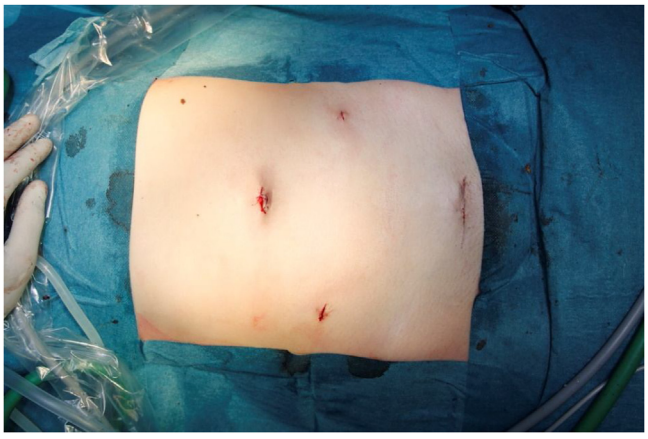

Fig. 3. Skin incisions closed.

Fig. 4. CT scan after tissue expander insertion: bowel loops out of the pelvis.

During radiation treatment, the patient experienced no toxicity. After completing radiotherapy the tissue expander was removed laparoscopically using the initial incisions. Laparoscopic removal was easy as there were only minor adhesions to the silicone implant. The PDS suture was intact, and broke easily with only a gentle pull. After 2 years of follow up the disease is in remission and the patient without any major complaint. Ultrasound examinations and skeletal scintigraphy were both negative regarding the progress of rhabdomyosarcoma.

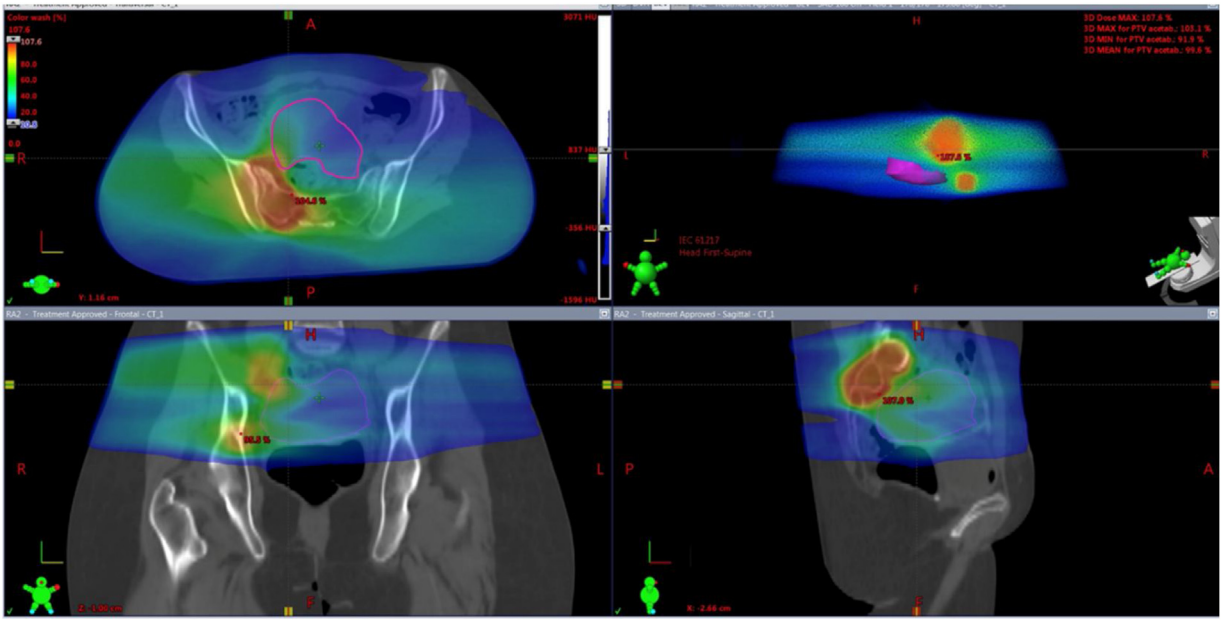

Fig. 5. Radiation field for reirradiation.

3. Discussion

Conventional radiation treatment can cause significant morbidity of patients from radiation enteritis. The aim of our treatment was to avoid this disability as much as possible. When radiation in abdominal cavity is expected, special attention should be given to those patients who have a low peritoneal reflection, and loops of bowel within the planned radiation field. A variety of surgical as well as mechanical displacement procedures in the abdominal cavity has been described to avoid exposure of bowel loops to radiation [5–15]. In case reports and small series a variety of approaches using omentum, different types of mesh and tissue expanders has been reported to displace organs. Despite some promising results majority of these methods never get a widespread clinical application. Benefit achieved by radiation was often not worth the additional surgical morbidity. The tolerance of adjacent healthy tissues is the main factor that limits the effectiveness of any form of radiation treatment. Even with displacement of organs the dose to other adjacent structures usually remains high [3,4]. There are many methods of reducing injury of the small bowel. Among them is multi-field conformal therapy with prior three dimensional planning where the profile of the radiation beam is shaped to fit the target. Adjustment in delivery of intensity-modulated radiotherapy can also be done. This improves the ability of treatment volumes to conform to the configuration of the tumour. In spite of all these improved techniques, visceral organs, particularly loops of intestine still occasionally get injured from being in the radiation field. To reduce that kind of injury alternatives to external beam radiotherapy which play some role in selected cases are brachytherapy or cryotherapy.

In situations where external beam radiotherapy is the preferred or most promising choice, non operative as well as operative methods for removing intestinal loops from the radiation field exist. Non-operative method to remove small bowel from the pelvis at the time of giving radiotherapy include extreme Trendelenburg positioning, abdominal wall compression, bladder distension or the use of a belly board device with patient in prone position [6]. Operative approaches described mainly in case report studies include creation of a temporary artificial pneumoperitoneum or insertion of a peritoneal dialysis catheter with the installation of gas or normal saline into the abdominal cavity [7,8]. Experiences with these techniques are limited. Extent of bowel removal from the radiation field is unreliable and unpredictable, they are time consuming, painful, and often need to be repeated. For pathologic lesions in pelvis adequate and stable displacement of intestinal loops out of pelvis is, at present, a promising technique. In first experience with use of tissue expanders complications were frequent when large expanders were left in the pelvis for long time. The risk was especially high for bladder, ureteric and iliac vessel compression [9]. Compression of iliac veins with deep vein thrombosis and pulmonary embolism has been reported. Infection, with abscess formation and fistulisation, has been reported to occur in up to 7 % of cases. Heavy expander can cause constipation due to obstructive defecation [9,10]. When large incision into the planned radiation field for expander insertion is necessary, wound infections are frequent [13]. Tissue expanders do very little to prevent radiation injury to the bladder or rectum, with radiation cystitis [14] and proctitis still common complications. More recent reports where smaller expanders were inserted, are associated with fewer complications [11]. Contemporary saline filled tissue expanders are easy to insert and remove laparoscopicaly. They are non-adherent to peritoneum and small bowel and radioresistant to degradation. When filled with normal saline they are similar in density to visceral tissues, therefore they do not alter the isodose distribution of radiotherapy. In our patient, a conventional 400 mL normal-saline-filled silicone tissue expander without anchoring tabs was used. It was kept in the pelvis by means of a dissolvable polydiaxanone (PDS) purse string suture. However there is a significant risk with use of dissolvable sutures. Too early degradation of suture can cause tissue expander migration, so non-dissolvable suture may also be used. The choice of a conventional sized expander and the avoidance of overfilling were challenging because of literature reports of the risks of ureteric and iliac vessel compression.

4. Conclusion

Laparoscopic insertion of pelvic tissue expander prior EBRT and it's subsequent removal after EBRT is safe and effective method for displacing loops of bowel out of the pelvis. With minimal morbidity converts untreatable disease to treatable by allowing delivering high doses of radiation to the patient. The ease, sim-

plicity, reversibility, and minimally invasive nature of laparoscopic tissue expander insertion are its main attraction. It should be considered as an option for excluding small bowel from the pelvis prior to radiotherapy of sacrum. These combined techniques hold a double hope of more effectively treating a difficult cancer and also diminishing or eliminating the costly and disabling effects often seen with conventional radiation. Opportunities are open for valuable collaboration and innovation in this area, especially in the further development of minimally invasive displacement techniques.

Conflicts of interest

None.

Source of funding

None.

Ethical approval

The study is exempt from ethical approval.

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

Mirko Omejc operated and conceived the case report.

Andrej Omejc, Lucija Vegan collected the data, contributed to the writing.

Vaneja Velenik analysed the data and supervised the work.

All authors discussed the results and contributed to the final manuscript

Research registration

Not applicable.

Guarantor

Mirko Omejc.

Provenance and peer review

Not commissioned, externally peer-reviewed.

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