Annals of Pediatric Surgery

CASE REPORTS Open Access

Giant foreign body granuloma after endoscopic management of vesicoureteric reflux: a case report

Saket Davera[1], Anjan Dhua[1*], Shipra Agarwal[2] and Mohamed Sulaiman[2]

Abstract

Background: Deflux®, a dextranomer/hyaluronic acid copolymer, is extensively used as an endoscopic bulking agent for management of vesicoureteric reflux (VUR). The complications following Deflux® treatment as described in the literature are clinically insignificant transient obstruction and infections of the urinary tract. We report a rare case of a giant Deflux® granuloma presenting as an intraoperative surprise while undergoing open ureteric reimplantation for the failure of prior endoscopic management.

Case presentation: A 2.5-year-old boy with a primary VUR needed Anderson Hyne's pyeloplasty of the affected side for concomitant pelviureteric junction obstruction. During removal of the double "J "stent in the postoperative period, subureteric Deflux® was injected, hoping that a more invasive procedure could be avoided. At a later date, as VUR persisted, an open ureteric reimplantation, was performed. Intraoperatively, a large Deflux® granuloma was noted at the site of previously injected site.

Conclusions: Giant Deflux® granuloma is a rare complication of this modality of VUR management. A focussed radiological assessment in such pateints during follow-up is required, especially those planned for surgical intervention in the vesicoureteric region later.

Keywords: Case report, Dextranomer, Deflux, Vesicoureteric reflux, Foreign body granuloma

Background

The endoscopic injection method for the treatment of vesicoureteral reflux (VUR) is well established. Currently, the most frequently used agent is Deflux®, a dextranomer/hyaluronic acid copolymer. The technique is simple and generally done as a day care procedure. The complications following Deflux® therapy are infrequent and include clinically insignificant transient obstruction and infections of the urinary tract [1]. We describe a case of a giant foreign body granuloma at the site of previous Deflux® injection. This case is being reported to highlight this complication of Deflux® therapy, which is reported scantily in the pediatric urology literature [1]. An awareness of this phenomena may have an essential bearing on the management options selected by the treating surgeon.

Case presentation

A 2.5-year-old boy with a primary VUR was being followed up since birth for antenatally detected right-sided hydronephrosis. A renal dynamic scan earlier had shown an obstructive drainage pattern with a selective renal function of 36%. He also had grade 4 VUR on the right side on a preoperative micturition cystourethrography (MCUG) (Fig. 1). He underwent a right sided open Anderson-Hynes pyeloplasty (AH pyeloplasty) with an indwelling double J stent (DJ) earlier at the age of 6 months. During the procedure, the ureters were slightly

*Correspondence: anjandhua@hotmail.com

[1] Department of Paediatric Surgery, All India Institute of Medical Sciences (AIIMS), Ansari Nagar, New Delhi 110029, India
Full list of author information is available at the end of the article

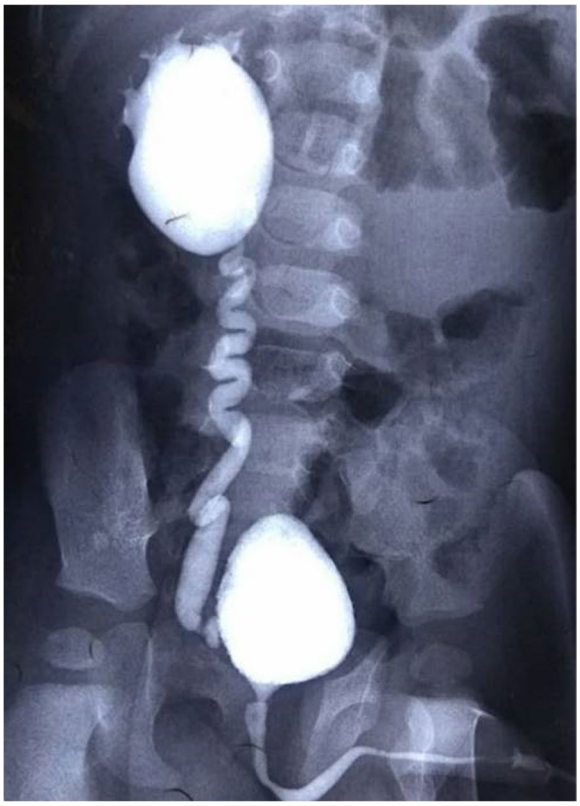

Fig. 1 Preoperative MCUG is showing grade 4 VUR on the right side with a dilated pelvis

dilated with a distinct short segment narrowing at the pelviureteric junction. After ensuring a normal urine microscopy report, the DJ stent was removed cystoscopically 4 weeks after pyeloplasty, and 1.0 ml of Deflux® was also injected sub-ureterically on the right side by the STING (subtrigonal injection) technique. By dynamic hydrostatic distension classification system, the right ureteric orifice was a H2 type, and the contralateral ureteric orifice was of H0 type.

In follow-up visits, improved renal drainage was observed on a renal dynamic scan done with a bladder catheter, but the VUR did not resolve as documented on a repeat MCUG (both done after 6 months of AH pyeloplasty). The MCUG did not reveal any other abnormality other than the reflux. An ultrasonography (USG) performed at this time showed right sided hydroureteronephrosis and no other abnormalities. In the subsequent follow-up period, he had two episodes of culture-proven urinary tract infection (UTI) requiring intravenous antibiotics for treatment. A dimercaptosuccinic acid scan showed multiple cortical breaks in the right kidney; hence, open reimplantation was planned. At the time of surgery (done after an interval of months from AH pyeloplasty), a 4×3 cm foreign body granuloma of Deflux® was discovered at the previous injection site (Fig. 2A). A plane could be developed between the granuloma and the ureter, and it was subsequently enucleated out. In the process, it required a long incision over the granuloma for access. After removal of the granuloma and closure of the long incision on the bladder mucosa, a cross trigonal reimplantation of the ureter was performed. Postoperatively, the child had prolonged hematuria which resolved conservatively. The histopathology report suggested an intense inflammatory reaction with foreign body material and giant cell reaction with a fibromuscular wall (Fig. 2B). In the follow-up period, he remains symptom and UTI free, and the upper tracts well-preserved after 18 months of the last procedure.

Discussion

The endoscopic management of VUR is now widely practised by pediatric urologists.

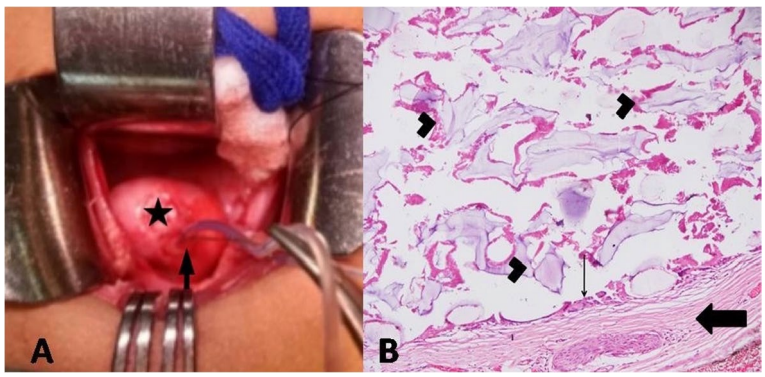

Fig. 2 **A** Intra operative findings of a large granuloma (star) encasing the right ureteric orifice (arrow showing the exit of a catheter from the ureteric orifice). **B** Abundant amorphous foreign body material (arrow heads)with giant cell reaction (arrow) within the fibromuscular wall (bold arrow), H&E, × 200

Various agents like Teflon paste (Mentor, Santa Barbara, California), Macroplastique (Uroplasty Inc., Minneapolis, Minnesota), and cross-linked bovine collagen have been used earlier. Subsequently, Deflux® (Q Med, Uppsala, Sweden) was approved by the United States Food and Drug Administration in 2001 [2]. There has been a steady increase in the usage of Deflux® since then, not only in VUR but a trend towards the use of Deflux ® for other urological conditions have also emerged in the medical literature.

A foreign body type of local reaction is expected around any implant. This is also true for Deflux®, and studies have shown that though there ensues a granulomatous response around the lower ureter, but it is not so intense that it precludes subsequent ureteric reimplantation [3]. All these properties have made Deflux® an ideal option in the armamentarium of the pediatric urologist as a bulking agent for treating VUR.

With this background information, a literature search showed that Życzkowski et al. in 2012 reported two cases very similar to the case being reported. In both the cases, a hard lumpy granuloma was seen at the site of previous Deflux® injection. The excised material sent for histopathological examination revealed chronic inflammatory lymphocytic infiltrate with a cluster of a foreign body surrounded by polynucleic histiocytes as was also found in our case. In one of the two patients, the granuloma was so infiltrative that it required excision of the terminal ureter as it was found unsuitable for reimplantation. This was unlike our case, where a plane of dissection was available, and the "Defluxoma" could be enucleated, albeit requiring a more elaborate incision above the ureteric ostia. The phenomena of granuloma formation is distinct from calcification of the Deflux ® implant which is relatively common. The calcific implant often mimic distal ureteric calculi radiologically but usually are of no clinical consequence unlike granulomas which can take giant dimensions [4, 5].

Granulomas at the site of injection of bulking agent was first described in 1991 by Bonnet et al. in cases where Teflon was used and subsequently other workers also reported similar findings with Teflon [6]. Deflux®, on the other hand, has been quite safe, and no such adverse effects have been reported in the literature, except the two cases described above [1]. This along with our experience of this index case leads us to believe that despite being a synthetic bulking agent with an acceptable safety profile, the formation of giant granulomas (Defluxoma) is still possible, although rarely. The cause of the granuloma formation in the index case still eludes us, although it may be speculated that the patient's inappropriate immune response, the technique of injection, and the sterility of the implant may have a role. The last two factors are modifiable but cannot be firmly attributed to since we lack any evidence against them. In an ideal situation, the granuloma should have been picked up in the preoperative radiological work-up (USG and MCUG) before the reimplantation surgery; however, in the index case, probably it was missed. A focussed examination by an experienced radiologist may probably would have given us same additional information. If detected in preoperative workup, they can point the surgeon to anticipate difficult planes of dissection, and this may even dictate the choice of access (open versus minimally invasive) and the actual technique of ureteric reimplantation, depending on tissue characteristics remaining after excising the granuloma. Loss of the length of ureter may also occur, and this may further test the preparedness of the surgeon. Our case required an extensive incision around the ureteric ostia to enable us to dissect and free the terminal ureter for subsequent reimplantation. Our case highlights that whenever endoscopic injection therapy is embarked upon a radiological screening for such patients must be performed in the follow-up. By screening, these rare complications may be picked up more readily than in the current scenario.

Conclusion

Giant granuloma formation is a possibility after injection therapy for VUR. A focussed radiological assessment in such pateints during follow-up is required to unearth this rare complication especially those who require additional surgical procedure to avoid intraoperative surprises.

Abbreviations

AH-pyeloplasty: Anderson-Hynes pyeloplasty; VUR: Vesicoureteric reflux; DJ: Double "J"; MCUG: Micturition cystourethrography; STING: Subtrigonal injection; UTI: Urinary tract infection; USG: Ultrasonography.

Acknowledgements

Not applicable

Authors' contributions

All authors have read and approved the manuscript. Concepts: AD, SA, MS. Design: SD, AD. Definition of intellectual content: AD, SA, MS. Literature search: SD, AD. Data acquisition: SD, AD. Manuscript preparation: SD, AD. Manuscript editing: AD, SA. Manuscript review: AD, SA, MS. Guarantor: AD.

Funding

None

Availability of data and materials

Available upon request.

Declarations

Ethics approval and consent to participate

Since the patient concerned is a child, written informed consent was obtained from the parent.

Consent for publication
Written informed consent was obtained from the parent for publication of this case report and accompanying images.

Competing interests
The authors declare that they have no competing interests.

Author details
[1]Department of Paediatric Surgery, All India Institute of Medical Sciences (AIIMS), Ansari Nagar, New Delhi 110029, India. [2]Department of Pathology, All India Institute of Medical Sciences (AIIMS), Ansari Nagar, New Delhi 110029, India.

Received: 21 September 2020 Accepted: 7 November 2022
Published online: 15 December 2022

References

1. Życzkowski M, Prokopowicz G, Zajęcki W, Paradysz A. Complications following endoscopic treatment of vesicoureteric reflux with Deflux® – two case studies. Cent European J Urol. 2012;65:230–1.
2. Seibold J, Werther M, Alloussi SH, Aufderklamm S, Georgios G, Todenhöfer T, et al. Long-term results after endoscopic Vur-treatment using dextranomer/hyaluronic acid copolymer– 5-year experience in a single-center. Cent European J Urol. 2011;64:84–6.
3. Stenberg A, Larsson E, Lackgren G. Endoscopic treatment with dextranomer-hyaluronic acid for vesicoureteral reflux: histological findings. J Urol. 2003;169:1109–13.
4. Cerwinka WH, Grattan-Smith JD, Scherz HC, Kirsch AJ. Appearance of Deflux implants with magnetic resonance imaging after endoscopic treatment of vesicoureteral reflux in children. J Pediatr Urol. 2009;5:114–8.
5. Yankovic F, Swartz R, Cuckow P, Hiorns M, Marks SD, Cherian A, et al. Incidence of Deflux® calcification masquerading as distal ureteric calculi on ultrasound. J Pediatr Urol. 2013;9:820–4.
6. Bonnet O, Grandcoin P, Sibert L, Gobet F, Hemet J, Grise P. Peri-ureteral granuloma after teflon injection for vesico-ureteral reflux. Prog Urol. 1991;1:1046–50.

Publisher's Note