The role of kyphoplasty in the management of osteogenesis imperfecta: risk or benefit?

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Abstract Kyphoplasty is a recognized treatment option in the management of symptomatic osteoporotic compression fractures, osteolytic vertebral metastases or haemangioma. To our knowledge, kyphoplasty with polymethylmethacrylate in a patient with type I osteogenesis imperfecta (OI) and a vertebral compression fracture has not been reported so far. We report on a 58-year-old patient with type I OI and a vertebral compression fracture at L2 with undislocated posterior vertebral wall and an additional older L1 fracture. Because of severe back pain resistant to conservative therapy over 5 months the indication for percutaneous kyphoplasty was made. Preoperative adjacent endplates of L2 were nearly parallel. Radiologically a minimal loss of height of the L2 vertebra was seen without adjacent fractures at 9 months follow-up. A slight increase of the preoperative kyphotic angle of overlying vertebrae L1 ($8.7^{\circ}/10.3^{\circ}$) and T12 ($10.4^{\circ}/11.0^{\circ}$) was apparent. The visual analogue scale showed decrease of low back pain from 10 to 2 allowing mobilization with a walking frame. Kyphoplasty constitutes a minimal invasive therapeutic alternative in the treatment of vertebral fractures in type I OI and pain, resistant to conservative treatment. Similar to the results of osteoporotic fractures

the immediate reduction of pain and stabilization of the fracture in undislocated fragments can be achieved. No adjacent fractures occurred 9 months postoperatively after kyphoplasty in type I OI. Preoperative parallelism of the endplates seems to protect from adjacent fractures.

Keywords Osteogenesis imperfecta · Kyphoplasty · Compression fracture · Polymethylmethacrylate · Spine

Introduction

Painful vertebral compression fractures caused by osteoporosis or osteogenesis imperfecta (OI) are disease-related fractures and require an adequate treatment in order to relieve postural pain and restore activities of daily living [10, 14]. Similar to patients suffering from severe osteoporosis, patients with OI are at risk to develop insufficiency fractures. These fractures may have an impact on pulmonary function due to kyphotic deformity and an increased mortality rate [8, 15]. On the basis of vertebroplasty, kyphoplasty was developed in 1998 with the intention to combine the analgesic and vertebral consolidation effect of vertebroplasty with restoration of the physiological height of the collapsed vertebral body [13]. During the last 20 years this technique becomes established in compression fractures due to osteoporosis, in hemangiomas or neoplastic processes [1, 5]. We extended the indication for percutaneous kyphoplasty to a compression fracture of L2 in a 59-year-old man with type I OI and severe pain resistant to conservative treatment. To our knowledge, kyphoplasty with polymethylmethacrylate (PMMA) has not yet been reported in patients with type I OI.

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Case report

History and examination

A 58-year-old man with type I OI, coronary heart disease, renal failure and dilatation of ascending aorta presented in our institution with severe back pain over 5 months without preceding injury. Because of severe posttraumatic gonarthrosis he was only able to walk either with a walking frame or with crutches. He contracted about 70 fractures since the age of 1 year without external force and was incapacitated for work since 20 years. Further he reports on having a daughter and grandchild suffering from OI.

Conservative treatment with pain killers and bracing failed. Using visual analog scale (VAS) the patient described severe lumbago (VAS score 10 of 10) without neurological deficits. Physical examination revealed a local tenderness of the lumbar spine without irradiation. Oswestry disability index (ODI) was 54%, constituting severe disability. X-rays showed a compression fracture at L2 with undislocated posterior vertebral wall, an additional old L1 compression fracture and radiographic appearance of osteoporosis. Preoperative adjacent endplates of L2 were nearly parallel, whereas the kyphotic angle of L1 was measured 8.7° and of T12 10.4° (Fig. 1a, b). Magnetic resonance imaging confirmed multiple roof plate fractures with an increased T2-weighted signals corresponding to bone marrow oedema of L2 (Fig. 2a, b). The integrity of the posterior vertebral wall was evaluated using computed

tomography. Further there were no signs of compression of the cauda equina. He therefore underwent kyphoplasty of L2 in which PMMA was used as bone void filler with particular interest in restoring the anterior third of the vertebral body.

Operation

The patient was placed in prone position using the bilateral transpedicular approach. Two voids were created in the L2 vertebra using a balloon catheter (Kyphon). After inflating the balloon catheters with contrast material and controlling under fluoroscopic monitoring, the PMMA was delivered slowly into the cavities (Fig. 3a, b). The amount of the cement was between 4 and 6 ml and mainly injected into the anterior part of the vertebral body with the intention to restore and strengthen the vertebra. The patient tolerated the intervention well without pulmonary or neurological complications.

Postoperative course

Postoperative X-rays of the lumbar spine were obtained, which showed the PMMA within the vertebral body. A slight increase of the preoperative kyphotic angle of overlying vertebrae L1 ($8.7^\circ/10.3^\circ/10.3^\circ$) and T12 ($10.4^\circ/11.0^\circ/11.0^\circ$) was apparent comparing preoperative with 1 and 9 month follow-up X-rays (Fig. 4a, b). The visual analogue scale (VAS) showed decrease of low back pain

Fig. 1 a, b Preoperative a.p. and lateral radiographs of the lumbar spine

Fig. 2 a, b Preoperative sagittal magnetic resonance image of the lumbar spine (T1- and T2-weighted)

Fig. 3 a, b Intraoperative lateral view of the lumbar spine during injection of PMMA into the bone void

from 10 to 2 allowing mobilization with a walking frame. The ODI was nearly maintained (52%) and was related to patients' disease at the latest follow-up appointment.

Discussion

Osteogenesis imperfecta is caused by a genetic defect of the gene that encodes type I collagen with the clinical manifestation of an inherited disorder of connective tissue. Characterized by abnormal osseous fragility leading to numerous fractures, the type I OI is the most common and mildest form and compatible with life [14].

Posterior stabilization was not an adequate treatment option in this patient due to poor quality of bone and the elevated risk of loosening. Further, radiation therapy may have resulted in a delayed pain relief [2]. The advantage of immediate pain relief in patients with osteoporotic fractures undergoing kyphoplasty is well known [4]. Kyphoplasty is used in the treatment of type A1 and A3 compression fractures according to the AO classification [12]. Although there are some drawbacks in kyphoplasty, like extravasation of PMMA [11] with injury to the spinal cord or vascular structures or fractures of adjacent vertebrae [6], it is a minimal invasive technique with immediate therapeutic success. The authors identified six case reports [6, 9, 10, 14, 16, 17] describing the treatment options of either vertebroplasty (five cases) or kyphoplasty (one case) in patients with OI (Table 1). The reported drawbacks of vertebroplasty in OI [16, 17] may be caused by the architecture of trabecular bone in this specific disease leading to an increasing risk of leaks, pulmonary embolism and resulting pulmonary and cardiac failure. On the other side the formation of "bone shell" in kyphoplasty may reduce the risk of cement leakage and embolism.

Fig. 4 a, b Follow-up (9 months) a.p. and lateral radiographs of the lumbar spine

Table 1 Reports of kypho- or vertebroplasty in osteogenesis imperfecta

Authors	Year	Study	Operation	Cement	Complication	Follow-up
Khoury et al. [10]	2008	Case report	Vertebroplasty	PMMA	None	13 months
Hardenbrook and Lombardo [6]	2006	Case report	Kyphoplasty	Silicate-substituted calcium phosphate	None	9 months
Kasó et al. [9]	2004	Case report	Vertebroplasty	PMMA	Successive fractures	11 months
Tozzi et al. [16]	2002	Case report	Vertebroplasty	PMMA	Pulmonary embolism	3 months
Rami et al. [14]	2002	Case report	Vertebroplasty	PMMA	None	17 months
Vasconcelos et al. [17]	2001	Case report	Vertebroplasty	PMMA	Transient arterial hypotension	n/a

n/a not applicable, PMMA polymethylmethacrylate

The literature on adjacent segment fractures after kyphoplasty reports on an incidence of 3–29% [3]. In a retrospective study, Fribourg et al. [3] pointed out that 21% of subsequent vertebral fractures occurred 2 months after kyphoplasty confirming stress loading on adjacent levels. After this time interval only occasional fractures were seen. Further, if the fracture is still mobile, the correction of the segmental kyphosis can influence in a positive way the sagittal balance which can result in a reduction of new fractures at adjacent levels [7].

We believe that restoration of the anterior wall of the lumbar vertebral body provides immediate structural support and prevents development or aggravation of kyphotic angulation. The endplates of L2 were nearly parallel in our patient and there were no adjacent fractures at 9 months follow-up. Our hypothesis on absence of adjacent fractures is the previous remodelling of the L1 vertebra due to an older compression fracture which might have supported

additional stress from the initial procedure. Patients' activity was maintained at 9 months follow-up and shown by the ODI (54–52%).

Using the technique of percutaneous kyphoplasty and applying it to a patient with OI who suffered from severe pain due to vertebral collapse, the authors showed a solution of immediate pain relief. To our knowledge this is the first reported case of kyphoplasty and PMMA in a patient with OI and additional adjacent older compression fracture.

Conclusions

Kyphoplasty constitutes a minimal invasive therapeutic alternative in the treatment of vertebral fractures in type I OI and pain, resistant to conservative treatment. Similar to the results of osteoporotic fractures, the immediate reduction of pain and stabilization of the fracture in undislocated

fragments can be achieved. Preoperative parallelism of endplates and additional older adjacent fracture seem to protect from adjacent fractures.

Conflict of interest statement None of the authors has any potential conflict of interest.

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