Three Cases of Intractable Pneumothorax Treated Successfully with Bronchial Occlusion Using Endobronchial Watanabe Spigots and Coagulation Factor XIII

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ABSTRACT — Purpose. We investigated the usefulness of endobronchial Watanabe spigots (EWS) and/or coagulation factor XIII concentrate for intractable pneumothorax with pulmonary emphysema. We used EWS and/or coagulation factor XIII concentrate together for 3 intractable pneumothorax cases with pulmonary emphysema suffering from prolonged pulmonary air leakage. Methods. Despite thoracic tube drainage for more than 2 weeks, the pulmonary air leakage continued. Bronchial occlusion using EWS was then performed. To obtain bronchial occlusion, the affected bronchi were determined by the balloon catheter test or with other clinical methods. If after using EWS bronchial occlusion minor air leakage continued, coagulation factor XIII concentrate was given intravenously for 5 days. Results. In the first case, we treated it with pleurodesis and coagulation factor XIII and did not perform bronchial occlusion. In the other 2 cases, we performed bronchial occlusion using EWS followed by the administration of coagulation factor XIII concentrate. In all 3 cases, the prolonged air leakage was stopped. On chest X-ray films, the collapsed lungs re-expanded and chest tubes could be removed after these treatments. EWS and/or administration of coagulation factor XIII concentrate might be an effective and noninvasive treatment for patients with intractable pneumothorax followed by prolonged air leakage.

KEY WORDS — Intractable pneumothorax, Prolonged air leakage, Endobronchial Watanabe spigots, Coagulation factor XIII, Bronchial occlusion

BACKGROUND

In cases of intractable pneumothorax with severe emphysema, and continual pulmonary air leakage, treatment can be very difficult. Because of the poor general status or low pulmonary function, many patients are at high risk when performing surgical procedures. For patients with decreased pulmonary function, pleurodesis may lead to lowering the QOL of patients because of complications. Therefore it is desirable that the treatment for these patients should be noninvasive. We tried 2 methods for this purpose; bronchial occlusion and the administration of coagulation factor XIII (fibrin stabilizing factor, CF XIII) (Fibrogammin® P, CSL Behring, Tokyo, Japan). Reasons to use CF XIII are that, it has been reported by many investigators that CF XIII promotes healing, particularly fibroblast proliferation.1 At the same time, it plays an important role in tissue repair. There have been reports that prolonged air leakage was stopped with CF XIII. For these reasons, we used CF XIII concentrate. We performed bronchial occlusion with endobronchial Watanabe spigots (EWS) and sequentially used CF XIII concentrate. Here we report 1 case treated with CF XIII only and 2 cases treated with

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Received July 9, 2009; accepted January 19, 2010.

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EWS and CF XIII. It would be best to remove EWS several months after improvement of the condition. Sometimes spigots are naturally expectorated when coughing.

METHODS

First, we identified the intractable pneumothorax as sustained pulmonary air leakage despite thoracic tube drainage for more than 2 weeks. In the first case the collapsed lung was expanded by pleurodesis only, but minor air leakage continued. We therefore used CF XIII to increase the ability of the wound to heal on the surface of lung. In the other 2 cases, we did bronchial occlusion first and then expanded the lung. Secondly, we administered CF XIII. In bronchial occlusion, the affected bronchi were determined by a balloon catheter test, chest CT or other clinical methods. To occlude the bronchi, we used EWS which has studs attached to its cork like surface (Figure 1) and projections for grasping at each end. It is made from high quality silicone. There are 3 sizes of EWS, with diameters of 5, 6 and 7 mm (S, M, L), of which the M size is mainly used at the level of sub-segmental bronchi. These spigots are removed several months after air leakage improved. If air leakage did not stop completely, 1250 units of CF XIII concentrate were given intravenously for 5 days.

RESULTS

The first case (case 1) was a 76-year-old man (Figure 2). He had a history of severe pulmonary emphysema and had undergone lung volume reduction surgery 8 years previously. He developed pneumothorax on his right side. After 15 days of thoracic tube drainage, pleurodesis was first performed, but minor air leakage continued. Then CF XIII concentrate was administered for 5

Figure 1. Endobronchial Watanabe spigots (EWS) are made from high quality silicone similar to the Dumon stent, and are designed with graspable parts at each end. There are 3 sizes of EWS (S, M, L) with diameters of 5, 6 and 7 mm.

Figure 2. Chest X-ray films show the findings before (a) and after (b) treatment in case 1. He presented pneumothorax on the right side. Pleurodesis and coagulation factor administration were performed.
days. In this case, we did not perform bronchial occlusion with EWS. Finally air leakage stopped and we could remove the chest tube. Because of this successful outcome, we decided to use EWS with the administration of CF XIII for the following 2 cases.

The second case (case 2) was a 69-year-old man (Figure 3). He had a history of pulmonary emphysema and squamous-cell carcinoma of the lung. He was on home oxygen therapy. He developed pneumothorax on his left side. After 24 days of chest drainage, bronchial occlusion with EWS was performed. Because of the continued slight air leakage even after bronchial occlusion, he
required bronchial occlusion 4 times. No pleurodesis was performed. After the 4th bronchial occlusion, sequentially CF XIII was administered for 5 days. Finally, air leakage was stopped and the chest tube was removed.

The third case (case 3) was a 78-year-old man (Figure 4). He had a history of severe pulmonary emphysema. He developed pneumothorax on his right side. After 22 days of thoracic tube drainage, bronchial occlusion with EWS was performed twice. Also pleurodesis was performed but was not successful. Then CF XIII was administered. Finally the air leakage halted and the chest tube was removed.

In these 3 cases, we recognized the decrease and stopping of air leakage. On chest X-ray films, the collapsed lungs were re-expanded and the drainage tubes were removed. In these cases of bronchial occlusion, it was difficult to determine the affected bronchi using the balloon test and other clinical methods. Therefore, bronchial occlusion with EWS was needed several times. In such cases, the administration of CF XIII can help to stop air leakage.

DISCUSSION

This is the first report of intractable pneumothorax being successfully treated with bronchial occlusion using EWS and CF XIII. We approached the treatment of prolonged air leakage mainly from 2 aspects. One is the bronchial occlusion of the affected bronchi which are causing the air leakage. Various fillers have been used in the past, such as fibrin glue, oxycel cotton, cyanoacrylate, and gelatin glue for intractable pneumothorax and pulmonary fistula. However these conventional bronchial occlusion methods are not reliable for bronchial blockade. In 1991 Watanabe et al. first reported bronchial occlusion with silicone.2 The results of clinical trials indicated that bronchial occlusion with silicone provided a surer and longer bronchial blockade than conventional methods. Watanabe et al. therefore developed the Endobronchial Watanabe Spigot (EWS), (Novatech, Grasse, France), a silicone-made bronchial filler. Watanabe et al. evaluated 58 cases treated with this procedure in 2003. Air leakage was stopped in 23 (39.7%) cases and was markedly reduced in 22 (37.9%) cases. As a result, the thoracic drainage tube was removed in 32 (57.1%) cases. There were no severe complications that were related to these procedures. The other method is the administration of CF XIII, which can increase the ability of the wound to heal on the surface of lungs. Topical factors in the delay of healing in a wound include hematogenic disorder, infection, foreign bodies, hematoma, radiation-induced tissue damage, external forces and edema. Systemic factors include hypoxemia, anemia, hypoproteinemia, amino acid deficiency, vitamin A, C, E deficiency, anti-cancer drugs, diabetes, zinc deficiency, steroid administration, and CF XIII deficiency.4 CF XIII is known as a calcium-dependent enzyme and is necessary for the production of stable fibrin clots.5 It has already been established that it stabilizes fibrinogen, and makes a fibrin-matrix where fibroblasts proliferate. It has been reported that CF XIII activity is temporarily reduced after gastrointestinal surgical operation or trauma and administration of CF XIII is helpful in dysphagia or fistula.8 Therefore we thought that CF XIII activity may be reduced topically in intractable pneumothorax and we anticipated the effectiveness of administering CF XIII to increase the ability of the wound to heal and stop leakage on the surface of lung. The complete stopping of air leakage may not always be necessary for patients with intractable pneumothorax. In our cases we expected that the combined therapy of EWS and CF XIII administration would cause the reduction of air leakage successfully and ensure more effective pleurodesis. There have been reports that prolonged air leakage was stopped only with CF XIII treatment in cases with lowered CF XIII activity. However, in our cases, we used the coagulation factor on patients with normal CF XIII activity, because we assumed that CF XIII activity was reduced topically, even if not systemically, in refractory pneumothorax cases. We assume that the expansion of the lung by bronchial occlusion makes pleurodesis more successful and raises the ability of the wound to heal on the surface, as well as decreases air leakage in the lungs. EWS is thought to be a very useful and safe instrument in bronchial occlusion. It is made from high quality silicone like the Dumon stent. The safety and biocompatibility of this material have already been established through extensive experience with the Dumon stent as well as CF XIII, which is an important factor for the restoring organization and the healing of wounds.9 Therefore, a fall in the activity of CF XIII is an obstacle to the healing of wounds and may be a cause of prolonged air leakage in intractable pneumot-
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CONCLUSION

EWS and/or administration of CF XIII concentrate can be an effective and noninvasive treatment for patients with low pulmonary function and prolonged air leakage. Further investigation and more experience with various cases are necessary to determine the usefulness of these procedures for patients with prolonged air leakage.

REFERENCES