CASE SERIES

The use of acellular dermal matrix in palatoplasty to decrease the rate of postoperative oronasal fistula

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Abstract

Different cleft palate repair techniques have been described to achieve optimum results and minimise complications. Postoperative fistulae are one of the most challenging complications after palate repair. In this clinical study, we reviewed the records of patients who underwent palatoplasty using acellular dermal matrix (ADM) as an addition to facilitate difficult cleft palate and palatal fistula closure.

It was a retrospective, comparative, single-centre study, in which records of patients who underwent cleft palate surgeries between 2015 and 2018 were reviewed. Patients who underwent cleft palate or palatal fistula repair with and without ADM were included. Fischer's exact test was used to compare the two groups (primary cleft palate repair with and without ADM) in relation to the rate of fistula occurrence postoperatively.

Charts of a total of 31 patients were reviewed. ADM was used in 13 patients; 8(61.5%) were primary repairs and 5(38.5%) were fistula repairs. Eighteen patients were repaired without ADM, of whom 16(88.9%) were primary cleft palate repairs and 2(11.1%) were fistula repairs. The statistical analysis showed no significant difference in fistula formation rate or recurrence in both the groups.

ADM is a simple, safe, and helpful tool for augmenting cleft palate repair, mainly in relatively wide and high-tension cleft palate repairs. In our study, a trend showing decreased complications with ADM was observed. Therefore, we recommend a multi-centre study with a larger sample to assess the significance of ADM in cleft palate and palatal fistulae repair.

Keywords: Cleft palate, Palatal fistula, Palatoplasty, Fistula repair, Acellular dermal matrix.

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Introduction

Palatoplasty is a procedure used to close the palatal cleft, aiming at improvement in speech with minimal maxillary growth interruption.[1] Despite the description of various techniques aimed at increasing the chances of a successful repair, complications are not uncommon. One of these complications which causes significant functional sequalae is oronasal fistula.[2,3] The reported prevalence ranges from 0% to 76%.[4] They usually occur at the junction of the soft and hard palates,[5] however, they may occur at any location along the repair line. Multiple factors have been associated with higher rates of fistula formation.[6] Tension along the repair line resulting from wide cleft or shortage of local tissue is the most common factor.[5] Fistulae are a further burden on the management of cleft palate patients in terms of morbidity and cost.[2,7]

Surgical repair of fistula in cleft palate patients is a challenge due to underlying fibrosis in the bed, compromised vascularity, and limited mobility of the tissue.[8,9] These factors also increase the chance of recurrence after surgical repair by up to 65%.[10,11] Hence, it is crucial to prevent the initial occurrence of fistula in primary cleft palate repair. In this regard, many techniques have been published to try to decrease the rate of fistula formation and facilitate a tension-free repair and/or reinforce the closure site.[1]

The use of acellular dermal matrix (ADM) as an extra layer for re-enforcement of closure in palatoplasty has been published in 2003.[8] ADM is widely used in the field of plastic surgery and is used in cleft palate surgery to reinforce both primary palatoplasty and closure of oronasal fistula.[4,8,12] Theoretically, ADM has several advantages in palatoplasty and fistula repair. It is safe, easy to use, widely available, strong, resists infection and avoids donor morbidity. However, it may not be readily available due to cost in some centres.[8,13,14] The potential benefit of decreasing the rate of formation of fistula after using ADM in palatoplasty has been previously reported.[9,15,16]

The objective of this study was to review and compare the cases of cleft palate repair with and without ADM in a

single centre in Saudi Arabia.

Case Series

This was a retrospective chart review that was conducted in December of 2019 and included all the patients who underwent primary palatoplasty and palatal fistula repair at King Saud University Medical City in Riyadh, Saudi Arabia, by three senior authors of this study between January 2015 and December 2018. Data collected from medical records included the age at the time of palate and/or fistula repair, type of clefting (according to the Veau classification),[17] presence of syndromes, palatoplasty repair technique, length of follow-up, rate of fistula development and recurrence. Exclusion criteria were, patients operated on in another hospital, patients who were operated by other than the three senior authors of this study, all patients who were lost to follow-up, and those repaired with techniques other than flap closure (such as tongue flaps or FAMM flaps). Allocation to groups was based on availability of ADM at the time of performing the procedure. Informed consent for surgery, use of ADM, photography and usage of the data for research was obtained from parents or legal guardians of all eligible candidates. Data were analysed using Statistical Package for Social Studies (SPSS 22; IBM Corp., New York, NY, USA). Continuous variables were expressed as mean ± standard deviation. Categorical variables were expressed as percentages. Mann-Whitney U test was used for continuous variables without normal distribution. Chi-square test and Fisher exact test were used for categorical variables. Shapiro-Wilk test was used to assess the normality of the data. P value less than 0.05 was considered statistically significant. The consent of the patient/guardian was taken prior to the writing of the manuscript.

Surgical Technique

Primary Cleft Palate Repair: All cleft palates were repaired with either Von Langenbeck or Bardach techniques.[18,19] In Von Langenbeck technique,[18] traditional bipedicled mucoperiosteal flap palatoplasty with intravelar veloplasty and muscle retro-positioning technique was used. In Bardach technique,[19] unipedicled mucoperiosteal flap palatoplasty with intravelar veloplasty was used. The Levator Veli Palatini muscle was dissected and freed from all abnormal attachments of the hard palate. Medial transection of the tensor aponeurosis as it courses medially around the hamulus was performed. Vomer flap(s) was used when necessary to ensure complete nasal layer closure. Following the nasal layer closure, ADM (ultrathin layer less than 1mm), as shown in Figure-1 a, was placed over the nasal lining as shown in Figure-1 b. The subsequent two layers were closed after the ADM was applied. No sutures were used to secure the ADM.

Fistula Repair: All cleft fistula cases were repaired by the closure of the fistula in layers. After separating both the layers surrounding the fistula (oral and nasal), a wide subperiosteal dissection of the oral and nasal mucosa and complete dense scar tissue surrounding the fistula was released. After nasal layer closure, ADM was placed over the repair line without sutures. Oral mucosa was closed subsequently. This process was followed in all fistula cases except one case which was treated with a palatoplasty redo using the Baradach technique combined with Intervelar veloplasty due to the wide nature of the fistula.

Postoperative Care: Postoperatively, all patients were hospitalised for one night, started on liquid diet, and received three doses of intravenous antibiotics and oral antibiotics upon discharge for seven days. Liquid diet was continued for a minimum of five days and then advanced to a soft diet for three weeks. All patients were followed postoperatively in the clinic at one week, three weeks, three months, and then yearly. Routine surveillance of fistula, dehiscence, and infections was documented.

Results

Demographics: Charts of 31 patients were included in the final analysis. Of the total 31 patients, 14 (45.2%) were male and 17 (54.8%) were female. Mean age at first presentation was (44.38 ± 33) months, while the mean age at the time of the first surgery was (51.32 ± 40) months. The mean follow-up duration after surgery was (47.10 ± 20) months. A total of 24 (77.4%) patients presented with primary cleft palate, and 7 (22.6%) presented with fistula secondary to operations done in

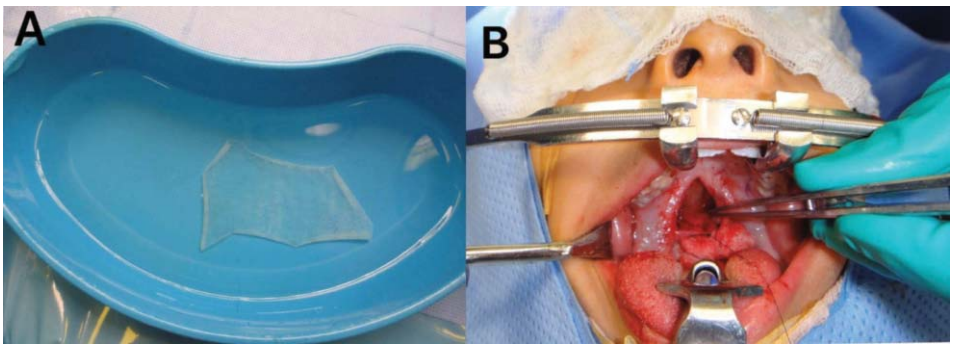

Figure-1: (A) ADM in kidney dish prior to application. (B) In primary palatoplasty, thin ADM is placed over the nasal lining following nasal layer closure.

other hospitals. Six patients were syndromic and were diagnosed with one of the following syndromes: Apert, Dandy-Walker, Joubert, Pierre Robin sequence, Stickler, and Down syndrome. Fifteen (48.4%) patients had a cleft lip and palate, while 16 (51.6%) had a cleft palate with no cleft lip. Fourteen (45.2%) patients had an alveolar cleft associated with cleft palate, while 17 (54.8%) had an intact alveolus. Sixteen (51.6%) patients had a complete cleft palate, while 8 (25.8%) had an incomplete cleft palate. With respect to Veau classification[17] for patients who had primary cleft palate at presentation, 6 (25%) patients had class 1, 10 (41.7%) patients had class 2, and 8 (33.3%) had class 4. None of our patients had class 3 primary cleft palate. With regard to the fistulae that occurred, all developed at the junction of the soft and hard palate. Wide fistulas were defined as fistulae wider than 1cm in the greatest width.

Surgical details: Bardach technique[19] with intravelar veloplasty was performed in 11 (35.3%) patients, Von-Langenbeck[18] in 9 (29%) patients, Redo Bardach in 5 (16.1%) patients, Bardach in 4 (12.9%) patients, Bardach with intravelar veloplasty and vomer flap in 1 (3.2%) patient, and Right Von-Langenbeck and left V-Y pushback in 1 (3.2%) patient. ADM was used in 13 (41.9%), (8 patients had a primary cleft palate and 5 patients had initial fistulae at presentation), while 18 (58,1%) patients were treated without the use of ADM, (16 patients had primary cleft palate and 2 had initial fistulae at presentation).

Complication rate in primary repair groups: A Total of 24 (77.4%) patients presented with primary cleft palate. ADM was used in only 8 (33.4%) patients, as shown in Table-1. Of the non-ADM group, 3 (18.8%) patients developed a fistula whereas 13 (81.3%) had no fistula on follow-up. In the ADM group, one (12.5%) patient developed fistula, while 7 (87.5%) had no fistula on follow-up. The location of the developed fistula was in the junction between soft and hard palate. There was no significant difference between primary repair groups in complication rate. There was no significant difference between primary repair groups in terms of gender, age at repair, Veau classification, syndromes, comorbidities, or surgical techniques.

Table-1: Summary of patients with acellular dermal matrix use in primary palatoplasty.

Patients	Cleft Type (Veau)	Age at Operation (Months)	Outcome
1	IV	11	Complete closure
2	II	31	Complete closure
3	II	24	Complete closure
4	IV	14	Lost follow up
5	IV	15	Complete closure
6	IV	19	Complete closure
7	II	27	Small fistula
8	II	120	Complete closure

Complication rate between fistula repair groups: Seven (22.6%) patients presented to our centre with fistula secondary to primary repairs performed in other hospitals. All fistulae were located at the junction of soft and hard palate. Two (28.6%), patients were treated without ADM; while ADM was used in the remaining 5 (71.4%) as evident in Table-2. Of all the patients treated without ADM, 2 (100%) developed a fistula. In the ADM group, 2 of 5 (40%) patients developed a fistula while 3 of 5 (60%) patients had no fistula on follow-up. There was no significant difference between the fistula repair group in complication rate. There was no significant difference between fistula repair group in terms of gender, age at repair, Veau classification, syndromes, comorbidities, or surgical techniques.

Table-2: Summary of patients with acellular dermal matrix use in fistula repair.

Patients	Age at operation (Year)	Follow up (Months)	Outcome
1	11	24	Complete closure
2	20	1	Complete closure
3	5	24	Recurrence
4	4	1	Complete closure
5	32	12	Recurrence

Discussion

Many techniques have been described for surgical closure of cleft palate, all having the objective of maximising palate functionality and decreasing the rate of complications.[1] However, even with meticulous surgical techniques, complications still occur. Fistula is one of the main complications. It has a significant negative burden on functionality.[2,3] The reported rate of fistula occurrence is highly variable, reaching up to 76%.[4] This variable incidence is caused by different repair techniques and fistulae locations.[4] In a systematic analysis of 44 studies that included 9,294 patients, the fistula occurrence rate was 5.4% among patients with Veau 1&2 and 17.9% among Veau 3&4.[20] Mostly, it occurs at the junction of the soft and hard palate.[5] The rate of oronasal fistulae formation was strongly related to cleft type, cleft width, mucosal tear, damage to the vascular pedicle, post-operative haemorrhage, infection, absence of a multilayer closure, upper respiratory tract infection and surgeon's experience.[6] Wide cleft or shortage of local tissues are the

most critical factors significantly influencing tension on the repaired palate.[5] Occurrence of fistula post repair causes significant functional limitation and imposes higher cost of treatment.[7] Nasal regurgitation of food or liquid, fetor, chronic inflammation, hearing loss, nasal escape, hypernasality, and velopharyngeal incompetence all can occur with fistula post repair.[3,21]

Surgical repair of the fistula is even more difficult than repairing a primary cleft palate. Fibrosis, compromised vascularity, limited mobility of the tissue, all add to the difficulty of the repair.[8,9] The rate of recurrence is reported to be high (65%).[10,11] Hence, prevention of occurrence of fistula during primary palate repair is crucial. Many techniques have been published aiming to decrease the rate of fistula occurrence. Basically, these techniques have focused on a tension-free repair and/or enforcement of closure site. Proper incision planning, extended relaxing incisions, meticulous wide subperiosteal dissection, vomer flaps, total release of tensor veli palatine muscle aponeurosis, hamulus and foramina lamina fracture, greater palatine vascular pedicle release, and adding an extra layer by using ADM can help to achieve a tension-free repair of both nasal and oral layers.[1]

ADM is nowadays widely used in plastic surgery.[12] Its use as an extra layer to enforce closure site in palatoplasty was initially published in 2003.[8] Its use was described as an adjunct in wide cleft palate and fistula repair.[1,8] In 2006, ADM was also used in repairing palatal fistula.[16] Since then, several reports have studied the use of ADM in primary cleft palate repair and fistula repair.[1,4] ADM is cadaveric skin that has been decellularised before use. It is safe, easy to use, widely available, strong, resists infection and avoids further donor morbidity. Additionally, it permits good healing with no functional deficits such as contracture or contour deformity. It provides a scaffold for tissue in growth, revascularisation, and mucosal epithelisation.[8,13,14] The exact mechanism by which it reduces fistula formation is not clearly understood.[1,7] Theoretically, it adds an additional layer of closure to provide a barrier layer in cases of minor dehiscence of the oral or nasal layers. It also adds more thickness to thin repaired tissues and minimises the dead space between the layers.[1,16] Moreover, there is some data that showed ADM does not interfere with palatal dynamics.[1,8,9,15,22] However, it is associated with a higher cost and, thus, is not affordable by all centres. Several studies reported its potential benefit in primary palatoplasty and repair of fistulae.[4,23] To this point, there are no clear indication criteria for ADM use. Some authors recommend using it in cases which carry higher risk of dehiscence like clefts wider than 15mm, syndromic patients, bilateral clefts, hypoplastic mucosa and muscles, and high tension closure line, etc.[1,8,24] A recent meta-analysis demonstrated that there is a paucity of data that clearly confirms its potential benefits.[25]

In our study, we divided the patients into two groups: The first group presented with primary cleft palate, while the second group presented with oronasal fistula after repair of primary cleft palate from other hospitals. In our study, ADM was used in primary palatoplasty in eight patients; only one (12.5%) developed a fistula on follow-up. ADM was used to repair fistula in five patients, of which two (40%) had recurrence of fistula on follow-up. A higher incidence of fistula recurrence in the fistula repair group was noted. This might be due to scarring caused by the original repair and lack of available tissue during closure, leading to significant tension in redo surgery. This is consistent with literature which reported a higher incidence of fistula recurrence in redo surgeries.[10,11] In both the groups, we did not find any statistically significant difference in fistula occurrence with the use of ADM. Nor did we find any statistically significant difference in fistula occurrence post-primary palatoplasty/repair, gender, age at repair, Veau classification, syndromes, comorbidities, and surgical techniques. In our study, increase in fistula occurrence with post-primary palatoplasty in severe Veau classes was noted. This might be because of our small sample size and unequal distribution of our sample in different Veau classes. In contrast to our data, multiple reports support the use of ADM in reducing fistula rate in primary palatoplasty.[4,22] However, most of these reports were of level 3 evidence or lower.[25] Recently, a prospective study revealed a beneficial effect of routine ADM use on primary cleft palate repair, i.e., decreased fistula rates (level 2).[7] The use of ADM in repairing palatal fistula and reducing recurrence rates is published in multiple studies.[8,9,15,16] Kirschner et al, Losee et al and others reported a decrease in the rate of fistula recurrence with ADM.[4,16] In literature, the post-primary palatoplasty occurrence of fistula was noted to be higher in more severe palate Veau classification.[20] On the other hand, no relation between the patient's age at repair and rate of fistula occurrence was noted in our study. Our result is consistent with a prospective study by Gilardino et al.[7]

In the literature, there is no particular surgical technique that has been shown to reliably decrease post repair fistula rate especially in wide clefts, which is consistent with our results.[1,7] The recurrence rate or fistula rate is different amongst various repairs.[4] However, we compared the rate of fistula collectively with and without ADM as against the collectively reported rate. This is the first study done using

ADM in cleft palate repair in Saudi Arabia. Here, ADM was used in primary cleft palate repair and fistula repair. However, our study has several limitations such as the relatively small sample size, retrospective nature of the study, and patients belonging to a single centre. In this study, ADM was used to augment repair in primary palatoplasty and palatal fistula. Even though our results were statistically not significant, we still believe that the use of ADM is beneficial in selected cases such as wide cleft and recurrent fistula. Small fistulae could be repaired using conventional methods. However, with large, recurrent, anterior fistula the risk of post repair recurrence dramatically increases. In such cases, the use of ADM may be of significant benefit. We recommend further prospective studies to assess the real benefit of ADM in palatoplasty and fistula repair. To our knowledge, this is the first study in the Middle East reporting experience with ADM in palatal repair or fistula repair.

Conclusion

The use of ADM in cleft palate repair or fistula repair has been shown, by many surgeons, to be an adjunct to palatal repair. In our experience, although the use of ADM for both primary palatoplasty and fistula repair did not show any statistical significance, it tends to show a trend favouring the use of ADM. This is the first study done using ADM in cleft palate repair in Saudi Arabia and the Middle East. We recommend further prospective studies to assess the real benefit of ADM in palatoplasty and fistula repair.

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References

1. Ağir H, Eren G,Yasar EK. Acellular Dermal Matrix Use in Cleft Palate and Palatal Fistula Repair: A Potential Benefit? J Craniofac Surg 2015; 26: 1517-22.
2. Witt PD, Marsh JL. Advances in Assessing Outcome of Surgical Repair of Cleft Lip and Cleft Palate.Plast Reconstr Surg1997; 100: 1907-17.
3. Wilhelmi BJ, Appelt EA, Hill L, Blackwell SJ. Palatal Fistulas: Rare with the Two-Flap Palatoplasty Repair. Plast Reconstr Surg 2001; 107: 315-8.
4. Losee JE, Smith DM, Afifi AM, Jiang S, Ford M, Vecchione L, et al. A Successful Algorithm for Limiting Postoperative Fistulae following Palatal Procedures in the Patient with Orofacial Clefting. Plast Reconstr Surg 2008; 122: 544-54.
5. Amaratunga NA. Occurrence of oronasal fistulas in operated cleft palate patients. J Oral Maxillofac Surg 1988; 46: 834-7.
6. de Agostino Biella Passos V, de Carvalho Carrara CF, da Silva Dalben G, Costa B, Gomide MR. Prevalence, cause, and location of palatal fistula in operated complete unilateral cleft lip and palate: retrospective study. Cleft Palate Craniofac J 2014; 51: 158-64.
7. Gilardino MS, Aldekhayel S, Govshievich A. A Prospective Study Investigating Fistula Rate Following Primary Palatoplasty Using Acellular Dermal Matrix. Plast Reconstr Surg Glob Open 2018; 6: 18-26.
8. Clark J, Saffold SH, Israel JM. Decellularised dermal grafting in cleft palate repair. Arch Facial Plast Surg 2003; 5: 40-4.
9. Cole P, Horn TW, Thaller S. The Use of Decellularised Dermal Grafting (AlloDerm) in Persistent Oro-nasal Fistulas after Tertiary Cleft Palate Repair. J Craniofac Surg 2006; 17: 636-41.
10. Schultz RC. Management and Timing of Cleft Palate Fistula Repair. Plast Reconstr Surg 1986; 78: 739-45.
11. Cohen SR, Kalinowski J, LaRossa D, Randall P. Cleft Palate Fistulas: A Multivariate Statistical Analysis of Prevalence, Etiology, and Surgical Management. Plast Reconstr Surg 1991; 87: 1041-7.
12. Simpson A, Samargandi OA, Wong A, Graham ME, Bezuhly M. Repair of Primary Cleft Palate and Oronasal Fistula With Acellular Dermal Matrix: A Systematic Review and Surgeon Survey. Cleft Palate Craniofac J 2018; 56: 187-95.
13. Sclafani AP, Romo T 3rd, Jacono AA, McCormick S, Cocker R, Parker A. Evaluation of acellular dermal graft in sheet (alloderm) and injectable (micronised alloderm) forms for soft tissue augmentation: Clinical observations and histological analysis. Arch Facial Plast Surg 2000; 2: 130-6.
14. Yim H, Cho YS, Seo CH, Lee BC, Ko JH, Kim D, et al. The use of AlloDerm on major burn patients: AlloDerm prevents post-burn joint contracture. Burns 2010; 36: 322-8.
15. Steele MH, Seagle MB. Palatal Fistula Repair Using Acellular Dermal Matrix: The University of Florida Experience. Ann Plast Surg 2006; 56: 50-3.
16. Kirschner RE, Cabiling DS, Slemp AE, Siddiqi F, LaRossa DD, Losee JE. Repair of Oronasal Fistulae with Acellular Dermal Matrices. Plast Reconstr Surg 2006; 118: 1431-40.
17. Veau. V. Classification of clefting. In: Bet-de-Lievre. pp. l-5. Masson, Paris. 1938 PP1-5
18. Von-Langenbeck B. Uranoplasty by means of detachment of the mucous-periosteal palate covering. Arch Klin Chir 1861; 2: 205-87.
19. Bardach J. Two-flap palatoplasty: Bardach's Technique. Operative Tech Plast Reconstr Surg 1995; 2: 211–4.
20. Hardwicke JT, Landini G, Richard BM. Fistula Incidence after Primary Cleft Palate Repair. A Systematic Review of the Literature. Plast Reconstr Surg 2014; 134: 618-27.
21. Jeffery SL, Boorman JG, Dive DC. Use of cartilage grafts for closure of cleft palate fistulae. Br J Plast Surg 2000; 53: 551-4.
22. Helling ER, Dev VR, Garza J, Barone C, Nelluri P, Wang PTH. Low fistula rate in palatal clefts closed with the Furlow technique using decellularized dermis. Plast Reconstr Surg 2006; 117: 2361-5.
23. Aziz SR, Rhee ST, Ziccardi VB. Acellular Dermal Graft Augmentation of Primary Palatoplasty.Case Report and Review of the Literature. J Oral Maxillofac Surg 2011; 69: 1221-4.
24. El-Kassaby MA, Khalifah MA, Metwally SA, Abd-ElKader KA. Acellular dermal matrix allograft: An effective adjunct to oronasal fistula repair in patients with cleft palate. Ann Maxillofac Surg 2014; 4: 158-61.
25. Aldekhayel SA, Sinno H, Gilardino MS. Acellular dermal matrix in cleft palate repair: an evidence-based review. Plast Reconstr Surg 2012; 130: 177-82.