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ΔΙΠΛΩΜΑΤΙΚΗ ΕΡΓΑΣΙΑ

Παροχή παιδοκαρδιοχειρουργικής φροντίδας σε δυσμενή περιβάλλοντα, μια συστηματική ανασκόπηση

Pediatric heart surgery care provision in developing countries, a systematic review

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ABSTRACT

Background: Many children in low resourced countries do not have access to treatment for congenital heart diseases, as the needed infrastructure and equipment is limited. Pediatric heart surgeons and medical staff have a long history of humanitarian aid, through the establishment of mission trips to developing countries.

Methods: Our systematic review evaluates the humanitarian pediatric heart surgery mission programs that have been published in Pubmed all over the yearsas well as programs for capacity building in developing countries.

Results: A single country outreach and the significant role of many missions' provider organizations is being underlined in our review.The CHD patients included in the assessed reports ranged from 20 to 1580 patients.The most frequent surgery types referred in the studies were VSD, ASD and TOF repairs, as well as PDA ligation.

Mortality rates differed among the reports. The enrolment of institutions into a multicenter congenital heart database showed improved outcomes in terms of reduction in length of ICU stay and mortality.

Conclusion: The establishment of mission programs to low resourced countries has saved many children with CHD, who otherwise would have died because ofthe absence of needed infrastructure and training of the local staff.

Keywords:humanitarian cardiac surgery, congenital heart disease, mission, developing country

ΣΥΝΟΨΗ

Υπόβαθρο: Πολλά παιδιά σε αναπτυσσόμενες χώρες δεν έχουν πρόσβαση στη θεραπεία για συγγενείς καρδιοπάθειες, καθώς η απαιτούμενη υποδομή και εξοπλισμός είναι περιορισμένα. Οι παιδοκαρδιοχειρουργοί και το ιατρικό προσωπικό έχουν μακρά ιστορία παροχής ανθρωπιστικής βοήθειας, μέσω της εγκαθίδρυσης ιατρικών αποστολών στις υποανάπτυκτες χώρες.

Μέθοδοι: Η παρούσα συστηματική ανασκόπηση αξιολογεί τα παιδοκαρδιοχειρουργικά προγράμματα ανθρωπιστικών αποστολών που έχουν δημοσιευτεί στην Pubmed καθ' όλη τη διάρκεια των ετών, καθώς και προγράμματα ανάπτυξης των ιατρικών ικανοτήτων στις αναπτυσσόμενες χώρες.

Αποτελέσματα: Στην ανασκόπηση υπογραμμίζεται η αριθμητική υπεροχή των ιατρικών αποστολών σε μία χώρα ανά αποστολή σε σχέση με εκείνες με πολυεθνική εμβέλεια, καθώς και ο σημαντικός ρόλος των διάφορων οργανισμών ανθρωπιστικής βοήθειας. Ο αριθμός των ασθενών με συγγενείς καρδιοπάθειες στις συμπεριλαμβανόμενες μελέτες κυμάνθηκε μεταξύ 20 και 1580 ασθενών. Οι συχνότεροι τύποιεπεμβάσεων που παρατηρήθηκαν ήταν οι διορθώσεις του μεσοκοιλιακού ελλείμματος (VSD), του μεσοκολπικού ελλείμματος (ASD), της τετραλογίας Fallot (TOF) και του βατού αρτηριακού πόρου (PDA). Τα ποσοστά θνησιμότητας διέφεραν μεταξύ των μελετών. Η εισαγωγή των ιδρυμάτων σε μια πολυκεντρική βάση δεδομένων για συγγενείς καρδιακές παθήσεις έδειξε βελτιωμένα αποτελέσματα όσον αφορά τη μείωση της διάρκειας παραμονής στη ΜΕΘ και της θνησιμότητας.

Συμπέρασμα: Η θέσπιση προγραμμάτων ανθρωπιστικών αποστολών σε χώρες με χαμηλό εισόδημα έχει διαφυλάξει τις ζωές πολλών παιδιών με συγγενή καρδιακή νόσο, τα οποία διαφορετικά θα πέθαιναν εξαιτίας της έλλειψης της αναγκαίας υποδομής και της κατάρτισης του ιατρικού προσωπικού.

Λέξεις κλειδιά: ανθρωπιστική εγχείρηση καρδιάς, συγγενής καρδιοπάθεια, αποστολή, αναπτυσσόμενη χώρα

INTRODUCTION

Congenital malformations constitute one of the leading causes of infant mortality and long-term disability among the children who survive. About 28% of all major congenital anomalies are heart defects. Annually one in 100 children is born with heart disease and approximately 90% of these children receive suboptimal care or have no access to care. In many developing countries the existing infrastructure to diagnose and treat congenital heart disease (CHD) is limited with an unequal distribution of resources.Moreover, excessive prolongation of the operation waiting time has several consequences, such as increased mortality, socioeconomic issues and emotional stress in related family members.The shortage of trained specialists to treat and manage congenital heart disease in middle- and low-income countries, should motivate physicians and surgeons from all over the world, to be actively involved in the global health networkand serve society's health care interests.

Humanitarian efforts are being made to establish an increased number of cardiac surgery programs and to enhance many of the programs that already exist. Each of these efforts may include short-term surgical missions and educational initiatives designed to build local capacity. The most common model is a visiting-short term surgical team arriving to an era with a high burden of CHD, operating for several weeks, frequently bringing the entirety of their equipment with them. Most often, the services are delivered in cooperation with local hospitals or clinics,with patients screened ahead of time by local providers who may also provide follow-up care. The financial support for the humanitarian outreach activities is provided, in the majority of cases, by many non-government organizations (NGOs), which commonly seek

approval and partnership with Ministries of Health and forge ongoing relationships for recurring missions.

Although short-term missions remain common, there is a lack of significant data collection in the most publications. While evidence regarding the practice and performance of medical missions is limited and scattered, the efficacy and sustainability of this model is being questioned. This review evaluates the programs of humanitarian medical and surgical missions in many developing countries that have been published in Pubmed. The focus is on the different kinds of CHDs being operated, the patients' demographics and clinical characteristics, as well as some quantitative data such as mortality rate and the number of operations being conducted.

METHODS

This review adheres to the recommendations of the PRISMA (Preferred Reporting Items for Systematic reviews and Meta-Analyses) statement(Figure 1).

Search Strategy

Our search was undertaken within the electronic database "Pubmed". The main search was conducted by using the algorithm (humanitarian OR mission OR war OR disaster) AND (cardiac OR heart OR cardiothoracic) AND (surgery OR surgical) AND (pediatr* OR paediatr* OR child* OR congenital). In addition, some key words (e.g., "paediatric surgery missions", "pediatric surgery missions", "pediatric heart surgery in developing countries", "humanitarian heart surgery") were used.Neither language

nor publication date filter was applied. Finally, we hand-searched the reference lists of all full-text reports. Our last search was conducted on September 5, 2018.

Study inclusion criteria

The articles were screened for relevance and full text was acquired if studies provided data on 1) humanitarian pediatric heart surgery missions or 2) programs for capacity building in developing countries.The surgeries were not limited to pediatric open-heart surgeries, but included interventional catheterization undertaken in children.Regarding average patients' age, we included reviews in which the 80% of the studying population were <18 years.

Exclusion criteria

We excluded all case reports, editorials and commentaries. Studies about children with rheumatic or acquired heart disease were excluded, as well as articles about programs in developed countries. Furthermore, studies about the strategy of transferring children to first-world countries for surgical care were not included in our systematic review.

Data extraction and management

All of the titles, abstracts and full-text papers for objectives 1 and 2 were screened. Data was extracted from all included full-text papers for the respective objectives. In more detail, we captured information onmission's recipient country, mission's provider (organization and country), date and duration, total mission visits, financial support, number of children with CHD, type of CHD,sex ratio, description of travelling and local team, patients' age, success and mortality rate, RACHS-1

classification,number of procedures being undertaken, surgery type, as well as the number of reoperations and finally, length of in-hospital and ICU stay. Moreover, we were interested in each mission's plan, namely frequency, duration, logistics, patients' screening and transition phase.

For objective 2, there were included studies that examine the efficacy, the sustainability and the impact of programs or missions with educational transition phase.

Data synthesis

Summary study and population characteristics were reported descriptively using medians and ranges. Due to the observed clinical heterogeneity, a quantitative synthesis was not deemed appropriate.

RESULTS

Study retrieval

We retrieved 636 records from the electronic database Pubmed through the algorithm referred in Methods (Figure 1). Another 327 records were collected through the utilization of keywords. Fourteen studies were included in our final review, of which 10 contained data relevant to objective 1 and 4 to objective 2. For 55 excluded full-text reports, the reasons of exclusion are the following; unclear mission's plan(n=13), no outcome of interest(n=32), reference only in prevalence/incidence data(n=8) and missions' overlap(n=2).

Characteristics of studies included in objective 1

General mission characteristics

Ten papers were included in objective 1. Regarding the mission recipient continent, it was observed that in 2 studies[1, 2] there was a multinational outreach and in the remaining 8 studies[3-10] there was a single country outreach. In 3 studies the main[8] or the only[1, 3] mission's provider organization was the International Children's Heart Foundation(ICHF). The aim of ICHF is to bring the skills, technology and knowledge to cure and care for children with congenital heart disease in developing nations. ICHF does this regardless of country of origin, race, religion or gender. ICHF members work toward this goal through medical mission trips, where they operate on children and educate local healthcare professionals.The other studies were referred in various organizations which were in charge of the mission, such as Voom Foundation, save a Heart Nigeria, Innova Children's Heart Hospital in Hyderabad[2], Children's Heart Fund of Ethiopia(CHFE) and International charity organiazations[5], Heart to Heart International Children's Medical Alliance[6], Heal the Children Northeast Asociacion del Coracon[10] and Heart Saving Project[7]. In the majority of the studies (6 papers), USA is the main country that provides the mission[1-3, 6, 8, 10]. UK[2], India[2], Brazil[9], Japan[7] and Portugal[4]were some additional mission provider countries. There was one report[1] referring to the participation of volunteers recruited from many countries, as ICHF members. These countries are USA, UK, Belgium, Australia, Denmark, Spain, Croatia, Peru, Chile, Costa Rica, Germany, Serbia, Turkey and Switzerland. The assessed programs had a duration that varied from 2 weeks to 11 years (median 5,5 years). The majority of the missions were carried out during the decade 2000-2009[6-9], whereas one mission began in the 90's[1] and another occurred in 1995[10]. The rest of the

missions either carried on during the decade 2010-2019[3, 5-7] or occurred in the present decade[2, 4] (Figure 2).

Characteristics of patients/surgeries

The sample size of the CHD patients included in the assessed reports[1, 2, 4, 5, 7-10] ranged from 20 to 1580 patients (median 249,5).Not all studies provided detailed information regarding the type of CHD of the included patients; only 4 papers[2-5] provided quantitative data. Of these, one report[2] assessed complex CHD only in a population of 20 Nigerian patients; the remaining three assessed larger general populations where the four most frequent CHDswere Ventricular Septal Defect(VSD), Atrial Septal Defect(ASD), Tetralogy of Fallot (TOF) and Patent Ductus Arteriosus (PDA).Of the 5682 CHD patients assessed in these 3 studies[3-5], 3405 patients (60%) belonged to one of the aforementioned groups. However, the frequency and relative frequency of these 4 CHD entities varied considerablywithin studies with the exception of TOF patients (12,5% to 17%); VSD frequency ranged from 16% to 25%, ASD frequency ranged from 3% to 23%, and PDA frequency ranged from 6% to 23%. Gender balance was observed in most of the studies (median 48% male patients) with only two studies showing over imbalance[7, 10]. Regarding the patients' age, five studies[3, 4, 6, 8, 9] includedneonates as well, while the remaining assessed infants and children only. Two studies[1, 5] provided no quantitative data about age distribution and two studies[9, 10] provided information only for the age range. Two papers[7, 8] had the same median of 38 months of the examined children and one study[3] had a mean age of 5 years with a median age of 2,8 years. In another report[2] a mean age of 1,7 years was mentioned. Additionally, the last

study with data on age distribution[4] analyzed that 76 children had a median of 20 days, 675 children had a median of 6 months and 921 children had a median of 5,5 years.

Risk adjustment for congenital heart surgery (RACHS-1) was developed to compare outcome data for pediatric patients undergoing cardiac surgery. RACHS-1 stratifies anatomic diversity into 6 categories based on age, type of surgery performed, and similar in-hospital mortality. Combining the data from the different studies[1, 4, 6], it became apparent that most of the patients pertain to RACHS-1 category 2(median 703) and no patients pertain to RACHS-1 category 5.

The 10 studies included in the analyses had an aggregate of 18.800 procedures conducted in the missions, which comprised cardiac surgeries and percutaneous catheter interventions.

In 2 studies[2, 9]the repairs were divided into palliative with rates 33,3% and 23,8% respectively, and definitive with rates 72.7% and 86%. The most frequent surgery types referred in the studies were VSD, ASD and TOF repairs, as well as PDA ligation. There was not detailed information about the surgery type in every study. In the three reports with quantitative data[3, 4, 10], 5607 children were operated. Of these, 1186 (21%) children had VSD repair, 697(12,4%) had TOF repair, 544(10%) underwent ASD repair and 540(9,6%) had PDA ligation (540, 9,6%).

Characteristics of clinical outcomes

One study[7], included the successful interventions which were 219/224 PDA occlusions, 23/23 pulmonary valvuloplasties and 6/6 aortic coarctation dilations.

Mortality rates differed among the reports. The highest mortality rate was referred in the study about the management of complex CHD in Nigeria[2], where there were 5 deaths in 25 patients, giving a mortality rate of 25%. On the contrary, the lowest mortality rate was 0,004%, representing 1 death (after 48 hours) in 255 catheter interventions due to complications[7]. One late death among 42 patients with surgically repaired lesions (mortality rate 0,02%) was referred in the 1995's mission[10]. Wallen et al[3] presented a significant drop in overall mortality (2,29% from 8,2%, p=0,0003) after the adjustment of a program's policy changes (requirement of the host site to have a fully functional blood bank and access to medical subspecialties, the ICHF providing 24-hour intensivist coverage, and not performing surgery on patients weighing less than 10 kg until local capacity has been developed). The estimated median mortality rate of the other studies[1, 4, 8, 9] was 8% with a range of 4,5% to 13,7%.

Four studies[3, 4, 8, 9] reported the number of reoperations that have been conducted. The mean number of reoperations was 135 with a range of 2 to 421.

Two studies[2, 4] included quantitative data about the length of in hospital and ICU stay. The median in hospital stay in the first paper was 4,5 days and the median ICU stay was 3,5 days. In the second study the median duration of in hospital stay is 9 days with a range of 1 to 76 days and the median duration of ICU stay is 2.4 days with a range of 1 to 57 days.

Mission's plan characteristics

Regarding the mission's plan, the number of total trips that were performed, as presented in the studies with quantitative information[1, 3, 5] ranged between 1 to

223 trips. The frequency of missions varied among the different reports[1, 5, 7, 10] from 1 to 15 missions per year. The duration of the team stay in the mission's recipient country ranged from 9 to 16 days, with an exception of a 2 years stay of a North American doctor in Guatemala[8]. The logistics of the mission, in the majority of the cases were organized by the visiting team or/and ICHF. Furthermore, the screening of the patients with CHD in order to select those who are in need of a procedure was conducted either by a forward visiting team[3, 5, 6] or by local physicians and cardiologists[1, 4] or both[3, 8, 10]. Delivering of education allowing a near- autonomous cardiac care of children with CHD in low resourced countries was ensured through several educational means; traditional didactics and multidisciplinary rounding[3],observation, meetings, educational conferences, reports, traveling scholar fellowships[2] as well as theory and practical courses[1, 4]. The formation of the volunteering teams was multidisciplinary, consisting mainly of surgeons, pediatric cardiologists, anesthesiologists, perfusionists, intensive care and scrub nurses.

Characteristics of studies included in objective 2

Four papers were included in objective 2. One of the studies[11] was about the establishment of an extracorporeal membrane oxygenation program(ECMO) in a Southern American country, which concluded that ECMO therapy can be implemented successfully in a developing country. Moreover, the study quoted the aforementioned success by describing the ECMO care model regarding staff, training process, care protocol, ECMO circuits and costs. Another paper described a comprehensive pediatric cardiac care program in Guatemala[12], where Aldo

Castaneda Foundation(ACF) was established locally and in the USA in 1997 to serve as a fundraising instrument of pediatric cardiac unit. The rest of Guatemala's cardiac program's goals were to provide diagnosis and treatment to all native children with CHD and to provide training of local staff. Atotal of 2630 surgical procedures were performed during a ten-year period (1997-2007), with an increasing number of operations each year. The median ICU and in hospital stay was 3 and 6 days respectively. The overall mortality rate was 8.3%. The remaining studies dealt with the inclusion of an institution in India[13] as well as 28 sites in 17 developing countries[14] (Pakistan, Colombia, Russia, China, Dominican Republic, Brazil, Argentina, El Salvador, Ukraine, Peru, Mexico, Belarus, Vietnam, Uganda, India, Guatemala, Bangladesh) in the International Quality Improvement Collaborative(IQIC) project and registry. The enrolment into a multicenter congenital heart database showed improved outcomes in terms of reduction in bacterial sepsis and surgical site infections, in length of ICU stay and mortality.

CONCLUSION

The disparity in access to pediatric heart surgery care in globally acceptable levels and the need for clinical services and education throughout the underserved areas of the world are evident. Bridging the gap by initiating several missions with educational transition phase for the local staff has successfully allowed the treatment of many children with CHD in their own environment and has reduced the high mortality rates of the previous years. The adoption and the faithful adherence to an applicable mission's plan as well as motivation provision for education to physicians and other medical staff demonstrate a concentrate approach that is very

promising for the achievement of the ultimate goal; saving as many children as possible.

Figure 1. PRISMA Flow Diagram

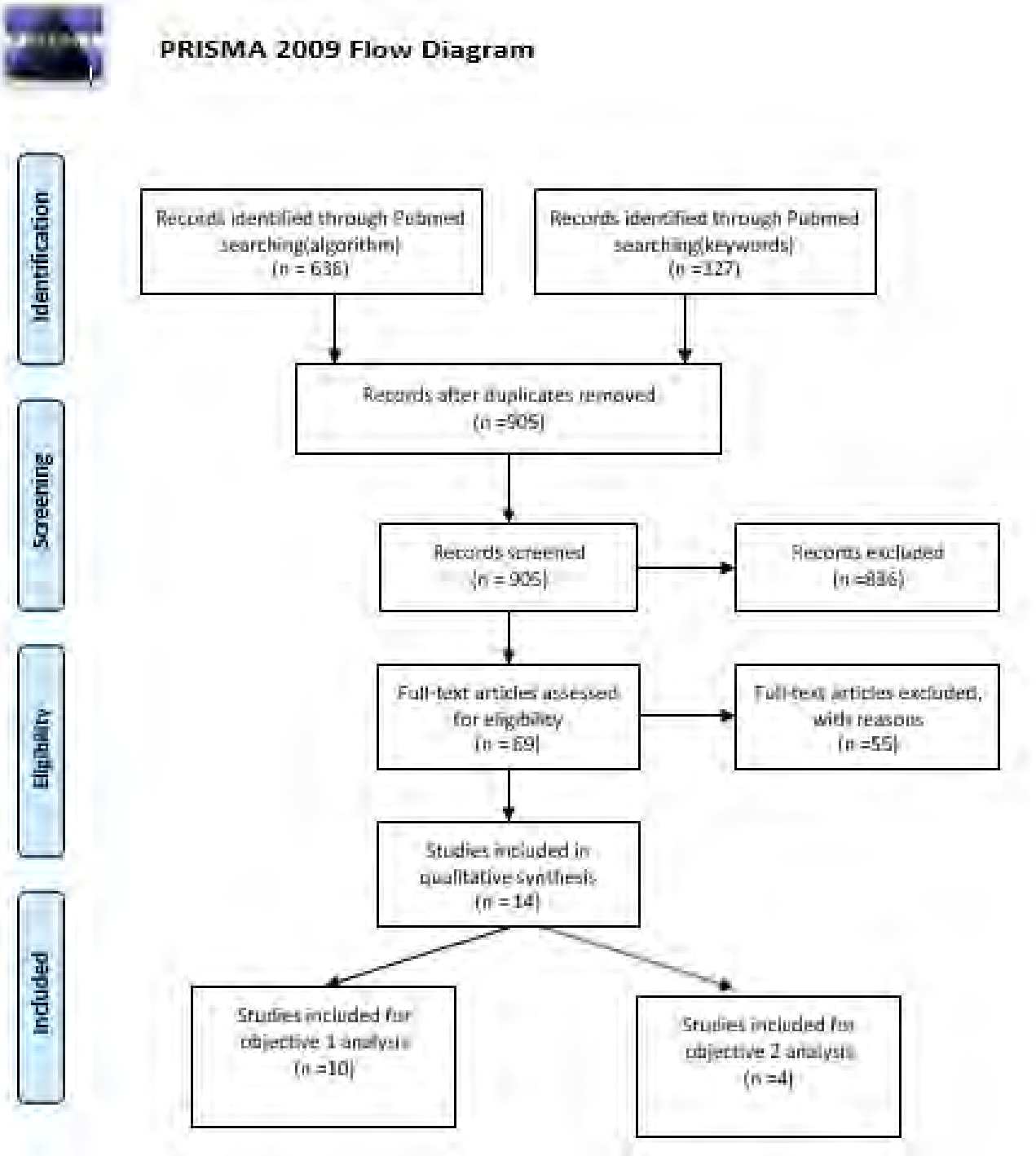
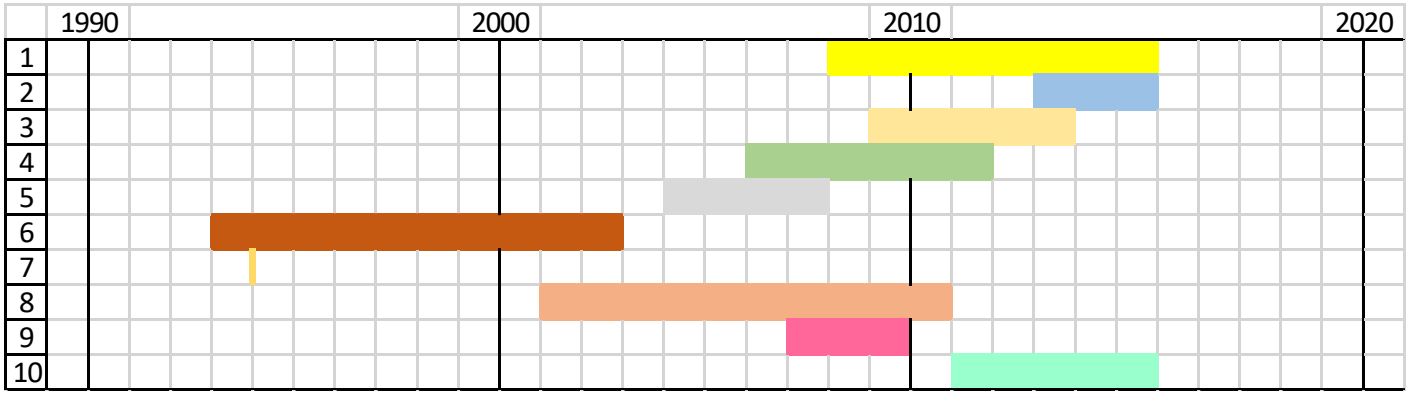


Figure 2. Mission program duration span



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